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Profiling the Language Abilities of Welsh-English Bilingual Children with Down Syndrome

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Profiling the Language Abilities of Welsh-English Bilingual Children with Down Syndrome



PRIFYSGOL
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Doctor of Philosophy in Bilingualism

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Abstract

Down syndrome is the most common chromosomal disability which impacts development on a range of aspects including cognitive and linguistic abilities. Consequently, concerns have been raised as to the extent to which children with this particular language profile are capable of acquiring two (or more) languages. This thesis aimed to address this issue by establishing the linguistic profiles of children with Down syndrome acquiring two languages, specifically Welsh and English in comparison to suitably matched control groups. The linguistic setting in which this research is based (i.e. Welsh-English bilinguals in Wales) is firstly introduced and consideration is given to some of the overarching debates and theories surrounding current literature in bilingualism. Following this, the research to date concerning bilingual language development in children with a developmental disability and more specifically children with Down syndrome is presented.

Children were recruited into one of four target groups and matched on developmental age as well as other important variables such as chronological age and SES. The groups were: Welsh-English bilinguals with Down syndrome, English monolinguals with Down syndrome, typically developing Welsh-English bilinguals and typically developing English monolinguals. The children were assessed on a range of cognitive and linguistic assessments in Welsh and English order to build a comprehensive profile of the children's language abilities. These assessments include measures of core, receptive and expressive language, phonological awareness and working memory. Four case studies of bilingual children with a dual diagnosis of Down syndrome and an Autism Spectrum Disorder are also presented.

Key findings show no negative impact of bilingualism compared to monolingual groups, both for the typically developing children and importantly, also the children with Down syndrome. Comparable profiles are evident for bilinguals and monolinguals with Down syndrome whereby corresponding patterns of strengths and weaknesses were found. Bilinguals with Down syndrome were also comparable to typically developing bilinguals in the additional language (Welsh). The results reveal that there is no evidence to support the view that children with Down syndrome should only be exposed to the majority language as the children in this study were developing appropriate language abilities which were commensurate with their linguistic backgrounds. The case studies document that even children with dual-diagnoses and substantial intellectual and linguistic challenges were developing appropriate bilingual abilities.

In the context of Wales, the findings challenge the view that children with Down syndrome should be placed in English-medium educational settings in favour of bilingual or Welsh-medium settings. On the contrary, bilingual services and provisions should be made available to this population, whilst also considering each child's circumstances and ensuring that there is appropriate support in that setting. Overall, the results increase understanding of bilingualism in Down syndrome and contribute to the growing body of literature documenting no adverse impact of bilingualism for children with developmental disabilities.

Implications arising from this research are discussed which include policy, clinical assessment, interventions, professional recommendations and family support. Future research directions are also proposed.

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Abbreviations

ABC	Aberrant Behaviour Checklist
ASD	Autism Spectrum Disorder
BPVS	British Picture Vocabulary Scale
CA	Chronological Age
CDI	Communicative Development Inventories
CELF-4	Clinical Evaluation of Language Fundamentals 4 th Edition
CELF-P	Clinical Evaluation of Language Fundamentals- Preschool Edition
DD	Developmental Disability
DLD	Developmental Language Disorder
DS	Down Syndrome
DSB	Down Syndrome Bilingual
DSM	Down Syndrome Monolingual
DS-ASD	Down Syndrome-Autism Spectrum Disorder
HFA	High Functioning Autism
IEP	Individualized Education Plan
KBIT-2	Kaufman Brief Intelligence Test – Second Edition
LFA	Low Functioning Autism
LOI	Language of Instruction
MA	Mental Age
MLU	Mean Length of Utterance
MS	Mainstream
MYM	Mudiad Ysgolion Meithrin
NV	Non-Verbal
NVMA	Non-Verbal Mental Age
NWRT	Non-Word Repetition Task
ONS	Office for National Statistics
PA	Phonological Awareness
SEN	Special Educational Needs
SES	Socioeconomic Status
SLT	Speech and Language Therapy
TD	Typically Developing
TDM	Typically Developing Monolingual
TDB	Typically Developing Bilingual
WM	Working Memory
WS	Williams Syndrome

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Introduction

Overview of the thesis

This thesis aims to create the first systematic linguistic profile of Welsh-English bilingual children with Down syndrome (DS). Group comparisons will be made with developmentally matched typically developing (TD) bilinguals and monolinguals alongside bilingual and monolingual children with DS. The first aim of the research is to explore the receptive and expressive language abilities of these groups of children in order to compare the language abilities within and between groups which include measures of vocabulary, language content and language structure. Secondly, the development of phonological awareness (PA) will be explored in order to specifically identify any impact that bilingualism has on this metalinguistic ability in bilinguals with DS. In addition, this research will also investigate if any factors predict successful bilingual outcomes in individuals with DS in order to explain the often-reported large range of individual variation for this population. Finally, language profiles of bilingual children with DS who have an additional diagnosis of an Autism Spectrum Disorder (ASD) will be presented and discussed. This will lead to a unique insight into how bilingual individuals with DS compare on the acquisition of various aspects of language.

Chapter 1 will present an overview of bilingualism which includes highlighting the current linguistic situation in Wales in which the research relates to. This chapter will also briefly summarise some current directions in bilingualism research. The second chapter will outline the research that exists to date on children with a developmental disability (DD) and provide an overview of language development in these individuals including research that explores bilingualism in these populations. The third chapter will explore the linguistic phenotype of children with DS specifically including vocabulary, morphology, syntax and PA. This chapter will also provide an overview of the available research in relation to bilingualism for this population before presenting the aims of the research presented in this thesis. The fourth chapter outlines the general method employed for the research including design, participant recruitment, procedures and methods of data analysis. The fifth chapter will present the findings in relation to the first aim, which explored a number of language abilities in terms of core, receptive and expressive language. The sixth chapter will present the findings in relation to PA abilities, and the seventh chapter will present a case analysis of the children with a dual diagnosis of DS-ASD. The limitations, implications and future directions are considered and evaluated through these chapters. The final chapter of this thesis will conclude by summarising the key findings and discuss these in relation to their impact and practical applications within research, clinical practice and policy. This chapter also discusses the implications within the context of the Welsh-medium education system and clinical practice in Wales alongside highlighting future research directions in this field.

Rationale

The Welsh language has an official status in Wales, meaning that Welsh should not be seen any less favourably than English (Welsh Language Commissioner: Welsh Language Measure, 2011). This has led to a relatively fixed number of Welsh speakers; however, Welsh is still considered a minority language, particularly in comparison to the number of English speakers in Wales. The unique education system in Wales allows for all children to attend Welsh-medium schools, regardless of their family's linguistic backgrounds. At the same time, there has been an increase in the popularity of Welsh-medium schools, for both primary and secondary schools, with more and more parents opting for their children to receive Welsh-medium education. In addition, in English-medium schools it is also compulsory for Welsh to be taught, in some form, in all schools in Wales until the age of 16. Consequently, all children in Wales will receive some exposure to the Welsh language during their time in education and many children develop language capabilities in Welsh and English. Bilingualism is thus a significant feature of education in Wales. Consequently, the growing field of research that explores the impact of bilingualism on language, cognitive and educational development becomes even more imperative and relevant.

Concurrently, there is a growing prevalence of children receiving diagnoses of some form of a learning disorder or developmental disability (DD), with an increase also in the number of children receiving a dual diagnosis of an intellectual and DD (Braun, et al., 2015). Given the increase in children with a DD and the large and growing number of bilingual speakers (especially in Wales), more and more children with DDs will be in bilingual environments or may have the opportunity to be bilingual. Therefore, a logical progression is to question what impact bilingualism will have for children who have language delays or impairments as a result of these developmental and/or intellectual disorders. As a result, questions may arise surrounding the suitability of bilingualism for these populations and considerations of this factor may become more and more recurrent. Children with DS show a markedly different progression of language development from that of typically developing (TD) peers and children with other DDs in terms of language production, comprehension, and grammar. As a result, it is justified to speculate on how development may be impacted by the addition of a second or third language for individuals with DS specifically.

Language development is a complex phenomenon that has a range of factors and influences that are associated with successful or unsuccessful outcomes. In considering bilingual language development this becomes even more multifaceted with further variables to consider. As a result, the scope of the research will focus on language abilities in terms of core, receptive and expressive abilities while also investigating a specific aspect of development, which has been previously reported to be specifically impaired in DS; phonological awareness (PA). Children of school age will be the focus of this project as, at this stage, it is expected that language acquisition will have begun to develop and be measurable both in terms of language comprehension and production. In addition, PA is known to develop during the early years of schooling in TD children (between 3 to 7-years-old) and is enhanced

by exposure to literacy instruction (Carroll et al., 2003; Smith & Tager-Flusberg, 1982). In addition, bilingual children will also have had exposure to English and Welsh either through the home, community and family environment or through schooling.

Previously, concerns have been raised as to the capacity of children with language impairments to learn two languages and researchers have documented that there is often a general belief that learning two languages is too great of a challenge for a child with a DD. The view that has been reported is that if learning one language is hard, two languages would then be *too* hard. Furthermore, practitioners (including speech and language therapists, teachers and doctors) who hold these beliefs have reportedly advised parents of children with DDs against exposing their children to a bilingual or multilingual environment (Drysdale et al., 2015; Pesco et al., 2016). To date, research has highlighted the general trajectories and profiles of language development in children with DS who are monolingual; however, only limited information exists on bilingual populations with DS. This research is consequently justified as a result of this deficit in our knowledge and will provide a novel insight into bilingualism in children with DS. It is anticipated that this research will make a substantial original contribution to the understanding of bilingualism in children with DS.

Significant implications may arise as a result of this research and will be discussed in this thesis in terms of clinical and educational policy and practice in Wales and beyond. In addition to this, the outcomes of the research may also assist families and clinicians in making appropriate language choices and evidence-informed recommendations in future. Furthermore, it is anticipated that this research will assist in overcoming some of the apparent misconceptions surrounding bilingualism (i.e. that bilingualism is detrimental to language development). This research will provide a significant contribution to our understanding and family practices regardless of whether bilingualism is found to be beneficial or detrimental to linguistic development. The focus of the research concerns Welsh-English bilingual populations which is an optimal and appropriate setting for this research, given the apparent support for bilingualism and drive for inclusivity in Wales.

Chapter 1: Bilingualism and the Welsh Language

1.1 Bilingualism: A Working Definition

In order to research bilingualism, it is necessary to understand and identify what is meant by the term bilingualism within the scope of this research. The issue of what or who is classified as a bilingual is a common theme in the literature, and the concept of a bilingual has also altered over time. Early researchers employed a strict classification of a bilingual as someone who is able to produce “complete meaningful utterances in the other language,” (Haugen, 1953, p.7). More recently it has been suggested that it is impossible to find a boundary between a bilingual and a monolingual. An alternative view, which is perhaps more realistic, is that bilingualism is a continuum (Mackey, 2000, p.26-27). A simplified and more flexible classification which is often adopted within research is the use of two or more languages. This is the generally accepted view with researchers currently, with the often-used phrase from Grosjean, (1995, p.259) “we will call ‘bilingual’ those people who use two, or more, languages... in their everyday lives.” This is the most standardized view of bilingualism, which is the standpoint taken when referring to bilingualism throughout this thesis.

As the current research concerns children, and more specifically children with intellectual disabilities, this definition will be appropriate as it may not be the case that complete meaningful utterances in a second language are what the children or their parents perceive as being bilingual. Children with DS may not become completely ‘fluent’ in an L1 or L2 to the same extent as a TD child, although they still may need two languages to function fully in their day-to-day lives, and therefore Grosjean’s definition appears to be the most appropriate for the population under study. Within the Welsh language context, this definition will also be an appropriate way to differentiate between monolinguals and bilinguals. As stated earlier, all children in formal education settings receive small amounts of input in Welsh even if they attend English-medium schools. In the context of this research and given the definition above, children attending English-medium schools who receive small Welsh language input would be considered as monolinguals as they do not use Welsh in their day-to-day lives.

The concept of an ‘uncontaminated’ monolingual was refuted by Bialystok (2001) where she argued that there was no such thing as a complete monolingual as “at the lowest levels of knowledge and awareness, exposure to fragments of other languages is unavoidable” (p.1). This quotation is undoubtedly relevant for English monolinguals in Wales who receive small amounts of Welsh language exposure within the public domain and through education due to this small degree of Welsh language teaching. For Welsh or bilingual families, in addition to exposure to Welsh through family, education and the public domain, there are also Welsh television and radio programs. The language of the vast majority of communities in Wales is English, and resultingly, Welsh-speaking families are automatically

exposed to, and learn English by default. It is accepted that monolinguals will not be defined as an individual who has had absolutely no exposure to a second language.

Language backgrounds are highly variable depending on a number of factors including the first age of exposure, consistency, quality and quantity of language input. A further distinction to be made at the outset of this research in the field of bilingualism is the concept of sequential (or consecutive) versus simultaneous bilingualism (Hamers & Blanc, 2000). Simultaneous bilingualism is often referred to as bilingual first language acquisition, where children are exposed to more than one language from birth or soon after. Sequential bilingualism, on the other hand, is where a second language is introduced after the first language has been acquired to some degree of proficiency. Within Wales, there may be a high proportion of early sequential bilinguals if children attend Welsh-medium education but are exposed to English at home. As a result, these children may not receive input to a second language until age 2-3 after the introduction of Welsh, if they attend Welsh-medium nurseries. Caution should be taken to ensure that the language backgrounds of any populations under study are carefully considered.

The definition of bilingualism will vary depending on who is providing or interpreting the definition, as different individuals within the same community may have different assumptions about what or who is bilingual. For example, fluent speakers of two or more languages may have different views towards bilingualism in comparison to researchers of bilingualism. Consequently, in researching bilingualism, caution is needed when using these terms with members of the general public to ensure a comparable understanding of bilingualism. There are also further terms used that may refer to the same concept, or similar concepts, such as multilingualism. The term 'bilingual' will be used to refer to an individual who speaks two languages, and 'multilingual' will be used to describe an individual who speaks more than two languages throughout this thesis. As mentioned earlier, the distinction is generally viewed as more of a continuum (Mackey, 2000:26-27), and this view can also be applied to multilingualism, with each additional language falling along another continuum of fluency.

It is now a commonly agreed upon standpoint that a bilingual is not simply the sum of two monolinguals (Grosjean, 1989), but that instead, a bilingual speaker has a unique combination of knowledge in two or more languages that results in an individual speaker with a unique understanding of those language systems. In addition, these systems may interact with each other. With this view, it is evident that there are differences within and between bilinguals and monolinguals in many ways, and it is reasonable to assume that bilinguals may be impacted by their languages in a different way than a monolingual. In the same instance, each bilingual will also be unique in terms of the languages they are exposed to, but also the unique language history across their lifetime.

A further consideration in relation to bilingualism research is the context of language exposure. It may be the case that a bilingual's two languages may be context-dependent, whereby one language is more commonly used in a given situation or circumstance. As a

result of this, their exposure to specific words may only ever occur in one language. Consequently, the vocabulary of that individual will vary across his or her languages, as exposure to a specific word may not be great enough to result in knowledge of that word in both a bilingual's languages. This is essential to consider when researching bilingualism, and more specifically in children as the number of times a child is exposed to a given word will, more than likely, be significantly lower than an adult's exposure to that word. When designing language tasks to assess language abilities, it is essential to consider a bilingual's two languages in order to ensure that a child's true language ability is being captured with the best degree of accuracy.

In summary, the definition that will be used for the following chapters will encompass a wide and fairly general definition of bilingualism, in that a bilingual will have communicative needs in two languages, and they will use two languages in their everyday lives. The output may not necessarily be as well developed in both languages, particularly with regards to populations such as those under study. This definition will encompass a wide range of language backgrounds and family language patterns (e.g. none/one/two parents speaking Welsh/English) which is a reflection of the current linguistic situation in Wales.

1.1.1 Bilingualism in Wales

There are two official languages in Wales (Welsh and English) as reported by the Assembly Commission (Official Languages Scheme, 2013), and both are used within the public and governmental domains. The Welsh language is part of the Celtic language family and would have originated in the British Isles. Welsh is still considered a minority language due to the number of Welsh speakers and it is also classified as an endangered language according to the UNESCO endangered languages atlas (Moseley, 2012). There is, however, a law in place which protects the language and gives it an official status, which is the Welsh Language Measure (Welsh Language Commissioner, 2011), which was passed and given royal assent in 2011 by the National Assembly for Wales. As a result of this measure, the Welsh language should be treated as favourably to the English language, and there are also subcomponents of the measure that establishes the role of the Welsh language, and also sets out current and future provisions for the language. The final component of the measure abolishes the previous Welsh language board. The Welsh language measure was put in place to replace the Welsh Language Act (1993), but the aims of which were in line with the new Welsh language measure, which is to promote and facilitate the use of the Welsh language.

A further aim of the newer Welsh language measure is to clarify the expectations of Welsh language services and ensures that there is consistency for Welsh speakers. As well as ensuring that the Welsh language is seen on the same level as English, it is also highlighted that a Welsh speaker should have the right to communicate fully in every part of their life through the Welsh language if that is what they desire. When it comes to bilingualism, as Welsh is a minority language, the language of the majority of societies as a whole are English.

As a result, there are only very few, if any, monolingual Welsh speakers. Therefore, all speakers of Welsh will generally be bilingual as it is considered a necessity to also speak English in most communities.

In terms of the number of speakers of Welsh, the most up to date official record of this was undertaken by the Office of National Statistics (henceforth ONS: Census 2011). Although there was found to be a slight decline in the number of Welsh speakers in the latest census in comparison to the previous census in 2001, there is still a stable number of Welsh speakers. In the latest census, of the people living in Wales who were over the age of 3, 19% of respondents reported being able to speak Welsh. This figure dropped slightly from 21% in 2001. In the latest census, there were a large proportion of people who reported to speak Welsh who were between the ages of three and fifteen (30%). In addition to this, the number of people in Wales who reported to be able to speak, read and write in Welsh also saw a small decrease of 1.7%, with the latest being 15% of the population. Table 1 displays an overview of the Welsh language skills captured in both the 2001 and 2011 census data from the Office for National Statistics.

Wales, 2001 and 2011, usual residents aged three and over						
Welsh language skills	Thousands, per cent					
	2001		2011		Change	
	Number	Per cent	Number	Per cent	Number	Percentage point
No skills in Welsh	2,008	71.6	2,168	73.3	160	1.7
Can speak, read and write Welsh	458	16.3	431	14.6	-27	-1.7
Can understand spoken Welsh only	138	4.9	158	5.3	19	0.4
Can speak but cannot read or write Welsh	79	2.8	80	2.7	1	-0.1
Other combination of skills in Welsh	84	3.0	73	2.5	-10	-0.5
Can speak and read but cannot write Welsh	38	1.4	46	1.5	7	0.1
Can speak Welsh ¹	576	20.5	562	19.0	-14	-1.5

Source: Office for National Statistics

Table 1. Summary of census information in 2001 and 2011 relating to Welsh language abilities in Wales (Office for National Statistics, 2011).

In terms of the location and distribution of speakers across Wales, some areas have a high concentration of Welsh speakers, whereas others have relatively low numbers. There are more communities of Welsh speakers towards the west coast and to the north of Wales. In addition to this, there are also some scattered areas with higher numbers of speakers than the neighbouring authorities. The local authority with the highest number of speakers is Gwynedd, which is again in the north of Wales, with 57.2% able to speak Welsh (Statistics for

Wales Bulletin, 2013). The areas with the lowest number of Welsh speakers are Blaenau Gwent, Merthyr Tydfil and Bridgend with 7.8%, 8.9% and 9.7% respectively, which are all in the south of Wales. In addition, the capital city of Wales is Cardiff, and this also has only a small number of Welsh speakers with 11.1% reportedly able to speak Welsh. Table 2 below presents the distribution of Welsh speakers and non-Welsh speakers by population number and by percentage.






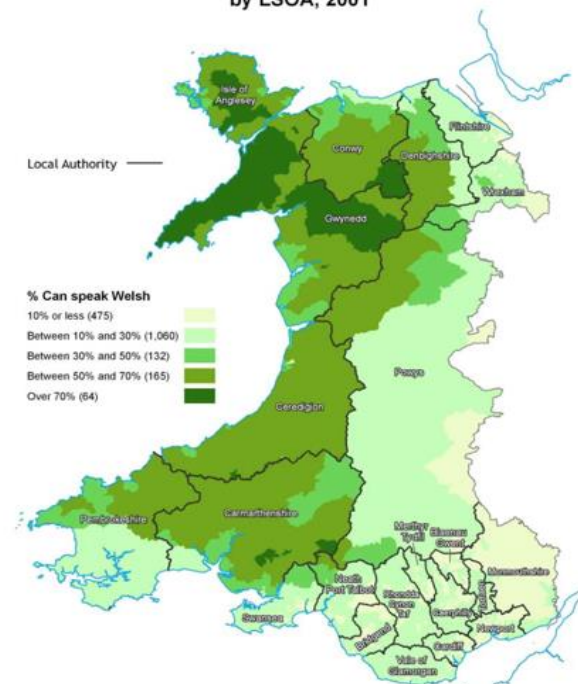
Local Authority 	Able to speak Welsh 	Not able to speak Welsh 	Percentage able to speak Welsh 	Total 
Wales	562,016	2,393,825	19.0	2,955,841
Isle of Anglesey	38,568	28,835	57.2	67,403
Gwynedd	77,000	40,789	65.4	117,789
Conwy	30,600	81,124	27.4	111,724
Denbighshire	22,236	68,291	24.6	90,527
Flintshire	19,343	127,597	13.2	146,940
Wrexham	16,659	112,766	12.9	129,425
Powys	23,990	105,093	18.6	129,083
Ceredigion	34,964	38,883	47.3	73,847
Pembrokeshire	22,786	95,606	19.2	118,392
Carmarthenshire	78,048	99,594	43.9	177,642
Swansea	26,332	204,823	11.4	231,155
Neath Port Talbot	20,698	114,580	15.3	135,278
Bridgend	13,103	121,442	9.7	134,545
The Vale of Glamorgan	13,189	108,829	10.8	122,018
Cardiff	36,735	295,538	11.1	332,273
Rhondda, Cynon, Taff	27,779	197,776	12.3	225,555
Merthyr Tydfil	5,028	51,595	8.9	56,623
Caerphilly	19,251	152,721	11.2	171,972
Blaenau Gwent	5,284	62,064	7.8	67,348
Torfaen	8,641	79,203	9.8	87,844
Monmouthshire	8,780	79,829	9.9	88,609
Newport	13,002	126,847	9.3	139,849

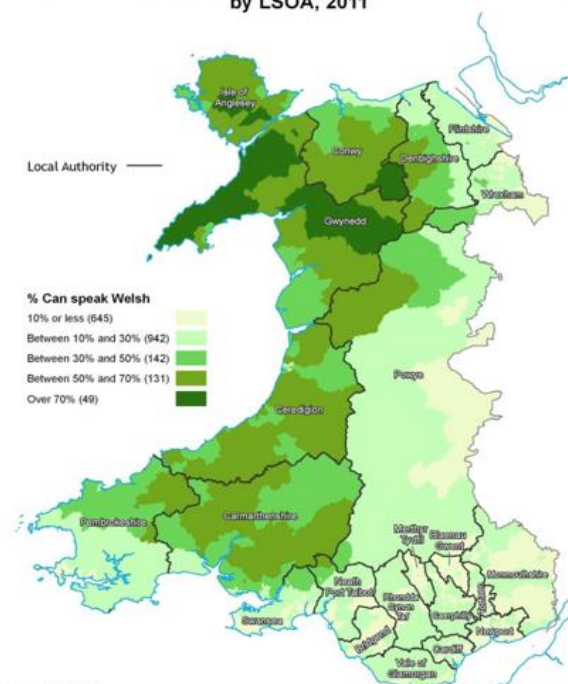
Table 2. Distribution of Welsh speakers by local authority (Office for National Statistics, 2011).

Typically, there are more speakers in the rural and northern areas of Wales, which includes the Isle of Anglesey. Figure 1 (Statistics for Wales Bulletin, 2013) displays the distribution of people who classify themselves as Welsh speakers, and also presents the distribution of Welsh speakers from the previous census in 2001, in order to enable a comparison over time. Although there has been a gradual decline in the overall numbers of Welsh speakers, the areas that have the biggest increase of Welsh speakers are generally located in the south of Wales and broadly around the Cardiff area, as well as in some regions in north Wales. The areas that have seen the biggest decreases in Welsh speakers are in the west of Wales in Ceredigion and Carmarthenshire. This is presented in Figure 2 below.

Proportion of people (aged 3 and over) able to speak Welsh, by LSOA, 2001



Proportion of people (aged 3 and over) able to speak Welsh, by LSOA, 2011



Source: 2011 Census

Figure 1. Speakers of Welsh by local authority according to census data from 2001 and 2011 (Office for National Statistics, 2011).

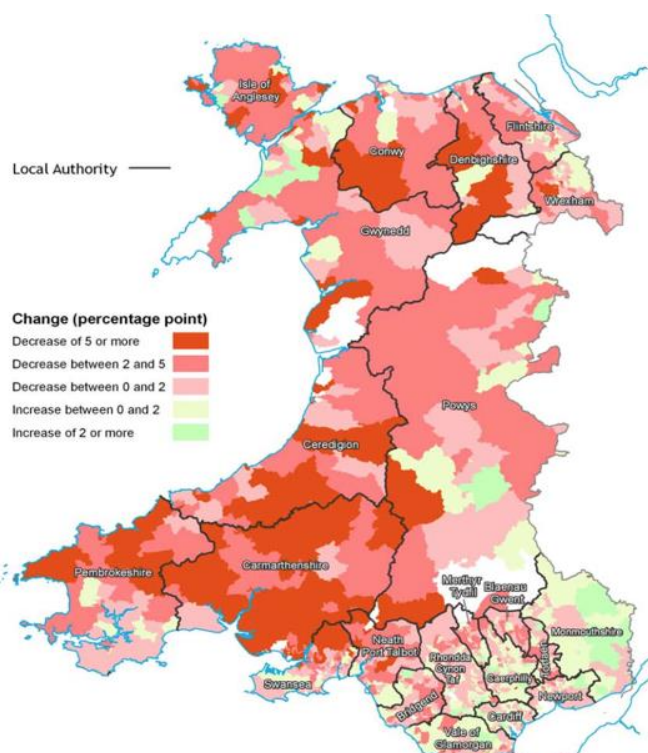


Figure 2. Changes in the number of Welsh speakers by each local authority between the 2001 and 2011 census (Office for National Statistics, 2011).

There have been more recent efforts to capture an accurate picture of the number and distribution of Welsh speakers, as well as their proficiency levels since the 2011 census. One of these is the Welsh Language Use Survey that was undertaken between 2013-2015 (Welsh Government: Welsh Language Use Survey, 2015). This survey was arranged by the Welsh Government and the Welsh Language Commissioner and collected information pertaining to the context and frequency that people in Wales spoke Welsh and with whom, as well as their reported language proficiency.

This survey gathered data regarding individuals over the age of three who lived in Wales and the survey investigated the number of 'fluent' Welsh speakers. The survey found that 11% of people who completed the survey (out of 310,600), classified themselves as fluent, which was a 1% decline compared to a similar study in 2004-2006. In contrast, the number of people who reported being able to speak Welsh but not fluently had increased. In the more up to date survey data, 23% of respondents stated that they could speak Welsh. In terms of the distribution of Welsh speakers, similar changes were found in the Welsh Language Use survey as the census, with the largest increases in speakers residing in South Wales, more specifically in Cardiff and Rhondda Cynon Taff. Similar patterns of declines in fluent speakers were also in the areas that generally have the highest concentration of speakers.

In considering the respondents of the Welsh Language Use Survey (2013-2015), there were a substantial number of people who said that they spoke Welsh who classified themselves as 'fluent', with 47% choosing this option (see Figure 3). The next level of fluency in this survey, 'a fair amount' accounted for 22% of Welsh speakers. Both of these figures are, however, lower than the previous 2004-2006 survey with 58% considering themselves as fluent and 21% stating that they spoke a fair amount of Welsh in the earlier survey. One factor that has not changed between the two Welsh language use surveys is the percentage of people who specified that they spoke Welsh daily.

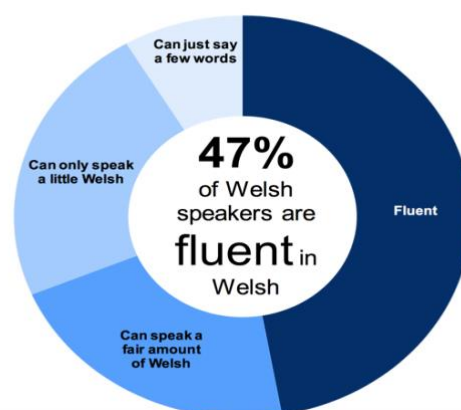


Figure 3. Fluency of Welsh speakers according to the Welsh language use survey from 2013-2015 (Welsh Government: Welsh Language Use Survey, 2015).

In terms of the overall population who report being fluent Welsh speakers, this figure is at 11%, with the vast majority of respondents of the Welsh Language Use survey reporting that they have no Welsh language skills (Figure 4).

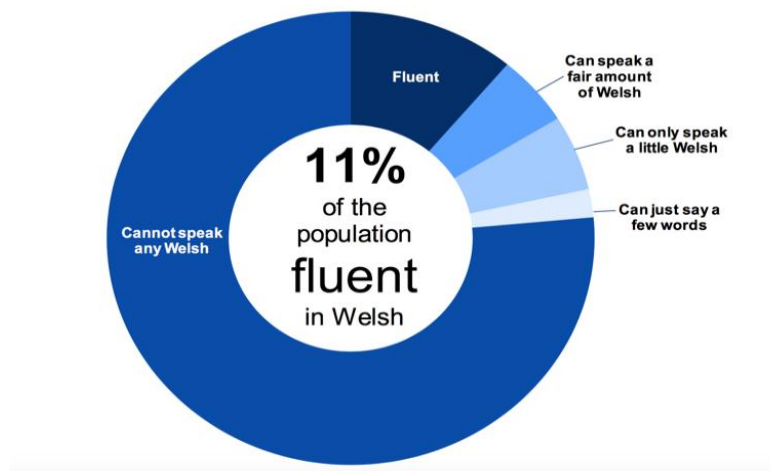


Figure 4. Number of speakers of Welsh including fluency according to the Welsh language use survey from 2013-2015 (Welsh Government: Welsh Language Use Survey, 2015).

Historically, the Welsh language has been in decline since the 1800s due to the legal status of Welsh during this time where English had a higher prestige. Since this, the number of monolingual speakers of Welsh has drastically declined, so that there are now only thought to be very few, if any, monolingual Welsh speakers. Since the Welsh Language Act (1993), the number of people who can speak Welsh did increase for the first time in a century, as displayed in Figure 5 according to the Welsh language use survey.

The Welsh language use survey is the most up to date record of the number, distribution and fluency of Welsh speakers, however, the main census is the most accurate and the key source of information regarding the Welsh language and it can capture more robust data regarding smaller areas such as each local authority in Wales. As a result, the data from the 2011 census is typically used to determine the effectiveness of language and educational policies. This data will be helpful when considering target samples for the present research project and identify the potential impact and implications of the outcomes of the research.

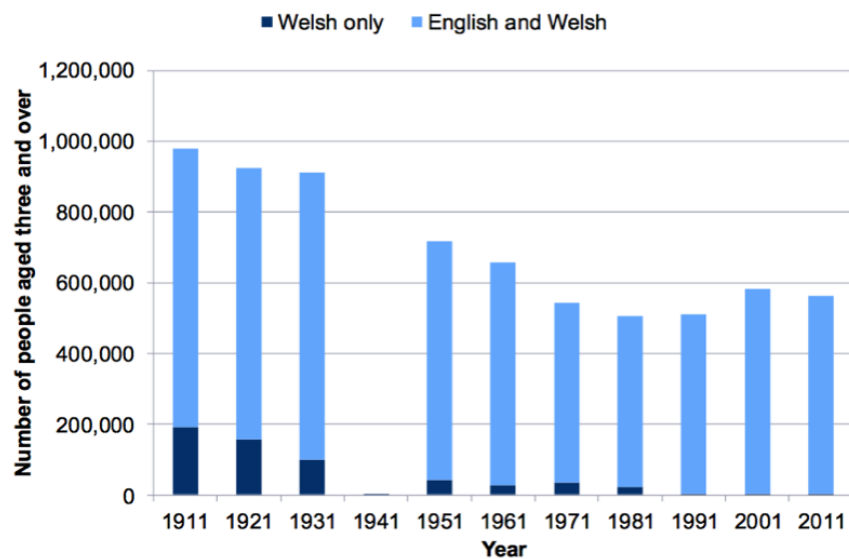


Figure 5. Number of monolingual and bilingual speakers on Welsh over the last century (Welsh Government: Welsh Language Use Survey, 2015).

1.1.2 Education Policy in Wales

The official status of Welsh has also transferred to educational provisions towards the Welsh language. In Wales, there is a legal requirement for schools in every local authority to have some degree of Welsh teaching for children between the ages of 3-16 in all public schools. One aim of the Welsh Government is to make Welsh-medium education accessible to all children from nursery through to secondary schools in order for Wales to achieve its goal to have one million speakers of Welsh by 2050, and will then be considered a 'truly bilingual Wales,' (Welsh Language Commissioner: Welsh Language Measure, 2011). Schools in Wales are either completely Welsh-speaking (besides English lessons), English speaking with Welsh lessons, or can also be a dual-stream with the option given to parents of which stream they would like their child to be placed in. In addition to this, some schools have varying combinations of Welsh use, however, ultimately all children in Wales will achieve some level of Welsh language skills.

In addition to the Welsh language being compulsory until the age of 16, there are also numerous schools and sixth form colleges that enable the continuation of Welsh-medium education and assessment at A-level. There are also some degree courses available at universities through the medium of Welsh, for example at Bangor University. Local authorities have a requirement by the national assembly (National Assembly for Wales Research Briefing, 2016) to constantly re-evaluate the Welsh-medium education strategy and ensure that adequate Welsh language provisions are available to parents who wish for their children to be taught through the Welsh language. Table 3 below (Welsh Government: School Census, 2016) displays the total number of Welsh-medium schools in each local authority in Wales for

January 2016. In order to compare this with the years prior to this, the table also displays the number of schools in previous years between 2012-2015.

The total number of schools in Wales during the time of this survey was 1574 (comprising of 13 nurseries, 1310 primary, 7 middle, 205 secondary and 39 special educational need (SEN) schools. The total number of Welsh-medium (including dual-stream and bilingual schools) in January 2016 was 482, giving an overall percentage of Welsh schools at 30.62%. Generally, the number of Welsh-speaking schools has declined slightly according to these figures since 2012, mainly in terms of primary schools, however, the number of children attending these schools has increased year after year, with the exception of pupils attending Welsh-medium secondary schools. There has also been an increase in the number of children receiving early years' instruction in Welsh in immersion programmes that are designed to give very young children a foundation of Welsh language skills.

There is one key provider of Welsh-medium education during the early years known as the Mudiad Ysgolion Meithrin (MYM) which translates as the Nursery Schools Movement. The MYM was established in 1971 to encourage parents to choose Welsh-medium education by providing playgroups and nurseries that promote the early development of the Welsh language. During the first 10 years of the organization, there were a total of 390 nurseries within this program and this led to an increase in Welsh speakers during the early years which was reflected in the subsequent census. This has continued to be the case for this age group in the three censuses which followed. MYM is now recognised as the prominent establishment for early years' education through the Welsh language. The quote below highlights the vision of the MYM.

	Primary (a)		Middle (b)		Secondary (b)	
	Schools	Pupils	Schools	Pupils	Schools	Pupils
Isle of Anglesey	46	5,571	0	0	4	2,959
Gwynedd	92	9,966	0	0	13	5,585
Conwy	23	2,288	0	0	2	1,292
Denbighshire	16	2,322	0	0	3	2,966
Flintshire	5	898	0	0	1	495
Wrexham	8	1,705	0	0	1	745
Powys	22	2,803	1	520	3	1,880
Ceredigion	41	3,540	2	1,404	4	2,211
Pembrokeshire	19	3,009	0	0	1	902
Carmarthenshire	71	9,805	0	0	5	5,502
Swansea	11	3,223	0	0	2	1,598
Neath Port Talbot	10	2,176	0	0	1	1,046
Bridgend	4	1,151	0	0	1	617
The Vale of Glamorgan	6	1,634	1	1,016	0	0
Rhondda Cynon Taf	16	4,786	1	519	3	2,512
Merthyr Tydfil	2	711	0	0	0	0
Caerphilly	11	2,927	0	0	1	1,499
Blaenau Gwent	1	266	0	0	0	0
Torfaen	3	920	0	0	1	965
Monmouthshire	2	366	0	0	0	0
Newport	3	686	0	0	0	0
Cardiff	16	5,348	0	0	3	2,625
Wales	428	66,101	5	3,459	49	35,399
Wales – January 2015	435	65,460	4	2,448	50	36,485
Wales – January 2014	444	64,366	2	1,577	52	37,400
Wales – January 2013	452	63,192	2	1,634	53	37,692
Wales – January 2012	461	62,446	.	.	56	41,262

Table 3. Distribution of Welsh speaking primary, middle and secondary schools in Wales by each local authority in January 2016 (Welsh Government: School Census, 2016).

“Our aim and privilege is to facilitate Welsh-medium care and education of a high standard to young children of all backgrounds in all parts of Wales. We will achieve this by extending our services as part of the national effort to tackle poverty and provide opportunities for every child in Wales to enable them to benefit from early years’ experiences through the medium of Welsh.”

- Mudiad Ysgolion Meithrin (2016, p.2)

In terms of Welsh-medium education for children with any form of SEN, the Welsh government has stated that each local authority should consider and evaluate the requirement for suitable provisions either through Welsh or English-medium in line with the child’s “linguistic needs as an integral principle of inclusive practice,” (Welsh Government: Inclusion and Pupil Support, 2017, p.9). In addition to this, the Welsh Language Act that was introduced in 1993 also contains some guidance in terms of children with SEN and the Welsh education system. In this act, it is stated that Welsh and English should be treated on a par with each other and that bilingualism should be provided “as far as reasonably practicable”, (Welsh Government: Special Educational Needs Code of Practice - Paragraph 1.7, 1993). It is

also noted that Welsh language schemes should be available to children with SEN assuming that it is appropriate and reasonably practicable, “according to the needs and wishes of the pupil and the parents.”

In addition to this statement, it is also noted that children with SEN’s progress will be hindered if they do not receive access to services in the language which is most appropriate for them and that they are most comfortable in. Therefore, if a child’s first language is Welsh, the Welsh Government suggest that they should receive access to Welsh-medium education and services in Welsh. Under the Education Act (National Assembly Government, 1996), a parent should have the right to a choice of language provisions, and local authorities have the duty to ensure that these provisions are in place and accessible to all. The Welsh government also stated in the Welsh-Medium Education Strategy (Welsh Government, 2014, p.12) that “no one should be denied opportunities for Welsh-medium education or learning Welsh as a language because of their race, ethnicity, disability, gender, sexual orientation, age or religion.” In line with this, if a child has a disability such as DS, that disability should not be a barrier to receiving Welsh-medium education.

The Welsh Language Board also reports that "specialist services should be provided for children with a SEN who speak Welsh or are educated through the medium of Welsh. These should include support from the sensory disability and physical disability services, speech therapists and the educational psychology service." In the Gwynedd County (which has the highest number of Welsh speakers as noted previously), the council website also states that the Welsh Education Scheme aims to ensure that children with a SEN receive equal linguistic provisions when it comes to bilingual education. Gwynedd Council state that “the aim of the Language Policy is to ensure that all pupils in the county are in possession of balanced, age-related bilingualism, to enable them to be full members of the bilingual society of which they are part.” (Gwynedd Council Website: Language Policy, 2014-2017).

Finally, in a recently published document, the Welsh Government’s review of the Welsh in Education Strategic Plans for 2017-2020 highlighted that one of the outcomes which it is expected that local authorities improve in terms of Welsh-medium education is the Welsh-medium provisions available for those with an Additional Learning Need (Welsh Government, 2017). The aims and regulations regarding SEN and the Welsh language appear clear, and these guidelines make it evident that access should be, and is encouraged through the Welsh language or a bilingual avenue. This will be explored in further depth later to identify if children with DS are encouraged to receive education in Welsh or bilingual by schools and whether the guidelines of availability of Welsh-medium provisions are available and accessible for these individuals.

1.1.3 The Welsh Language

In order to understand and assess the language development of children in Wales, and that of bilingual children acquiring Welsh and English, it is important to provide an overview of the

Welsh language. In particular, this review will briefly note differences and similarities between the Welsh and English languages which need to be considered in this research. More specifically, as one of the aims of this research is to explore phonological awareness due to the specific linguistic profile of children with DS (see section 3.3.5), it is important to highlight how the phonology and orthographies of the two languages under study compare and contrast with each other. In addition, this section will familiarize the reader of the Welsh language and Welsh sound system which may assist in identifying any cross-linguistic interactions. In this section, the Welsh language will be described in terms of its alphabet, vocabulary, phonology, orthography, morphosyntax, word stress and the mutation system.

Vocabulary and Alphabet

The first and foremost difference between Welsh and English is that the Welsh language has its own distinct vocabulary as with the majority of languages, with words often being unique and very different from other languages. At the same time, as a result of language contact with the two languages being used alongside each other in communities in Wales and the proximity of Wales to England, the Welsh language does have many loanwords/borrowings from English. For example, the word 'group' in Welsh is 'grŵp' /gru:p/ and the word for 'dance' is 'dawns' /dau:ns/. On the other hand, there are some English words which may have been influenced by the Welsh language such as 'penguin' which may have derived from the Welsh words 'pen', meaning 'head' and 'gwyn', meaning 'white' (French and Weekley, 1926; page 533).

In terms of the alphabets, there are 29 letters in the Welsh alphabet (compared with 26 in English), and seven of these are vowels (a, e, i, o, u, w & y). The Welsh alphabet is written using the Latin alphabet script and both Welsh and English share the majority of letters (Price, 1984). In addition, some letters are also marked with a circumflex in order to signify that the vowel is long. As mentioned, there are many loanwords from English used regularly in Welsh and the alphabet sometimes includes the letter 'j' which is not usually used in Welsh. Further loan words are often spelt slightly differently in Welsh in order to overcome the issue of not having the letters in the alphabet. An example of this is the word 'zoo', which is written as 'sŵ' in Welsh as there is no 'z' in the Welsh alphabet.

Welsh Phonology

The sound system of Welsh contains some phonemes that are non-existent in the English language. Additionally, there are also several regional variations found in the pronunciation of Welsh. Particularly distinguishable is the North and South Wales dialects and these will be discussed in terms of phoneme variations. In terms of consonants, there are 29 different phonemes found in the Welsh language in comparison to 24 in English (Borsley et al., 2007; Hannahs, 2013). There is an overlap with sounds which are also found in the English phonological system, however, there are some additional phonemes that are not found in English (for example the lateral fricative ɬ and the uvular fricative ɣ). The consonant

phonemes of Welsh are presented in Table 4 and are represented in terms of place and manner of articulation and voicing.

		Labial	Labial-Dental	Dental	Alveolar	Lateral	Palato-Alveolar	Palatal	Velar	Uvular	Glotal
Plosive	Voiceless	p			t				k		
	Voiced	b			d				g		
Affricate	Voiceless						tʃ				
	Voiced						dʒ				
Fricative	Voiceless		f	θ	s	ɬ	ʃ			χ	h
	Voiced		v	ð	z						
Nasal		m ɱ			n ŋ				ŋ ɳ		
Liquid	Voiceless				ɾ						
	Voiced				r	l					
Glide		w						j			

Table 4. Consonants found in the Welsh language in terms of aspiration, place and manner of articulation. *Note:* Adapted from Hannahs (2013).

There are between 11-14 single vowel phonemes in the Welsh language (in comparison to 12 single vowel phonemes in English), and these are displayed in Table 5, with reference to the tongue position in terms of front/backness, height and vowel length. The number of vowels is dependent on dialectal variation with speakers of Welsh, whereby those who have northern accents producing a larger variety of vowel sounds. Finally, the diphthongs of the Welsh language are displayed in Table 6, in terms of height, length and tongue position for the first and second components of the diphthong. There are between 8 and 15 of these (depending again on accent with northern accents typically producing more phonemes), whereas in English there are only 8.

	Front		Central		Back	
	Short	Long	Short	Long	Short	Long
Close	ɪ	i:	ɨ	ɨ:	ʊ	u:
Mid	ɛ	e	ə	(ə:)	ɔ	o:
Open			a	a:		

Table 5. Vowels found in the Welsh language in terms of height, length and tongue position. *Note:* Adapted from Hannahs (2013).

		Second Component		
		Front	Central	Back
First Component	Close	ʊi	ʊɨ	ɪu, ɨu
	Mid	ɛi, ɔi	ɛɨ, ɔɨ	əu, ɛu, ɔu
	Open	ai	aɨ, ɑ:ɨ	au

Table 6. Diphthongs found in the Welsh language in terms of height, length and tongue position for the first and second component of the diphthong. *Note:* Adapted from Hannahs (2013).

Orthography

In terms of orthography, the spelling of Welsh is very regular and pronunciation often follows simple one-to-one grapheme-phoneme matching. The sounds in the Welsh language often have a close relationship to the alphabet, and written forms of Welsh are generally reliable in inferring the spoken form (Spencer & Hanley, 2003). This is a substantial contrast to the orthography of English, as English is one of the least orthographically transparent languages in terms of consistency of grapheme to phoneme mappings, with silent letters and many possible variations in the pronunciation of some vowels. As noted previously, there are substantial variations in the pronunciations of Welsh depending on the regional dialect meaning that some vowel sounds will have more than one possible pronunciation, however, is it still far more orthographically transparent than English.

English phoneme to grapheme matching is also highly variable, as there are many irregular words that do not generally follow the patterns of English. For example, the letter 'i' in the words 'hint' and 'mint' are pronounced the same, however, the 'i' in 'pint' is pronounced differently. A further example of the inconsistencies of English is the letter 'h'. In words such as 'hospital' and 'humour', the pronunciation of the letter 'h' corresponds to the phoneme /h/ but in words such as 'hour' and 'honour', the 'h' is not produced. There are many possible explanations for these inconsistencies, for example, influences from Latin where the phoneme for 'h' was dropped. The pronunciation of English words and this phoneme in particular, is also determined by the speaker's regional accent.

Mutations

One additional element that is fairly complex in Welsh is that the initial consonant of many words changes as a result of the rule-governed mutation system, as with other Celtic languages. Mutations in the Welsh language are common, and these generally occur as the result of morphological or syntactic rules. The most frequent occurrence for a mutation in Welsh is when a word follows a number, a preposition or a possessive, and these fall into three categories of mutations, which are soft (meddal), nasal (trwynol) and aspirate (llaes; Borsley et al., 2007). Table 7 displays an overview of the most frequent consonants which mutate, and what sounds they mutate to within each mutation class.

The soft mutation is the most frequent mutation. An example of where the soft mutation occurs is the personal possessive pronouns '*dy*' meaning '*your*' and '*ei*' meaning '*his*'. This mutation also occurs after a number of prepositions including '*am*' meaning '*for/at*', '*wrth*' meaning '*by*' and '*o*' meaning '*from*'. This mutation is shown in the example of '*tad*' meaning '*father*' which mutates to '*dy dad*', '*your father*'. An example of the nasal mutation is when this occurs after the particle '*yn*' meaning '*in*' such as the town name '*Tonypanyd*' which mutates to '*yn Nhonypanyd*'. Another frequent nasal mutation is that following the personal pronoun '*fy*' meaning '*my*', whereby '*coes*', meaning leg, becomes '*fy nghoes*'. The aspirate mutation frequently occurs in several circumstances, for example following certain numbers (such as '*tri*' meaning '*three*', and '*chwe*' meaning '*six*'), as well as after the

conjunctive ‘*a*’ meaning ‘*and*’, and also following the prepositions ‘*â/gyda*’ meaning ‘*with*’. An example of this is ‘*te*’ meaning ‘*tea*’ and ‘*coffi*’, meaning ‘*coffee*’ which becomes ‘*te a choffi*’ (‘*tea and coffee*’) following the conjunctive.

Soft Mutation	Nasal Mutation	Aspirate Mutation
c (/k/) → g (/g/)	c (/k/) → ngh (/ŋ/)	t (/t/) → th (/θ/)
p (/p/) → b (/b/)	p (/p/) → mh (/m/)	c (/k/) → ch (/χ/)
t (/t/) → d (/d/)	t (/t/) → nh (/n/)	p (/p/) → ph (/f/)
g (/g/) → -	g (/g/) → ng (/ŋ/)	
b (/b/) → f (/v/)	b (/b/) → m (/m/)	
d (/d/) → dd (/ð/)	d (/d/) → n (/n/)	
rh (/r/) → r (/r/)		
m (/m/) → f (/v/)		
ll (/l/) → l (/l/)		

Table 7. Overview of the mutation systems in Welsh. *Note:* Adapted from Hannahs (2013).

Stress

Another distinction to be made between Welsh and English is the stress pattern. The stress will most often fall on the penultimate syllable in Welsh within polysyllabic words (with exception of borrowed words; Hannahs, 2013). In contrast, stress will usually fall on the first syllable in English, particularly for nouns, adjectives and compound nouns (however, there are of course many exceptions; Erdogan & Wei, 2019).

Welsh Morphosyntax

The word order of Welsh follows a distinctive pattern compared to English. Welsh, unlike the majority of other languages, has verb initial finite clauses, meaning that typically, Welsh follows a verb-subject-object (VSO) word order. In contrast, English usually follows a subject-verb-object (SVO) word order. There are, however, a number of auxiliary-initial clauses in Welsh. One example is sentences that use an Aux-Subject-Verb-Object word order which can be used to mark tense, as opposed to an inflection of the main verb (Borsley et al., 2007). These constructions follow a different word order where the subject follows the auxiliary verb, which is then followed by the main verb and the object (for an example, see Table 8). The standard forms are followed by the grammatical particle ‘*yn*’, which is subsequently followed by the appropriate verb. The main verb is not inflected in these cases.

Standard Form	Subject	Example
Rydw i	I	Rydw i yn cerdded
Rwyt ti	You	Rwyt ti yn cerdded
Rydych chi	You (formal)	Rydych chi yn cerdded
Mae e/o	He (south/north variation)	Mae e yn cerdded
Mae hi	She	Mae hi yn cerdded
Rydyn ni	We	Rydyn ni yn cerdded
Rydych chi	You (plural)	Rydych chi yn cerdded
Maen nhw	They	Maen nhw yn cerdded

Table 8. Example of auxiliary verb construction in Welsh present tense for ‘*cerdded*’, meaning ‘*walking*’.

These auxiliary verb constructions can also be used to indicate the future tense. For example, the future form of ‘*to be*’ is used along with the particle ‘*yn*’, followed by the main verb. An example of this is presented in Table 9 for the future tense of ‘*rhedeg*’ meaning ‘*running*’.

Standard Form	Subject	Example
Fe/Mi fydda i	I	Fe/Mi fydda i yn rhedeg
Fe/Mi fyddi di	You	Fe/Mi fyddai di yn rhedeg
Fe/Mi fyddwch chi	You (formal)	Fe/Mi fyddwch chi yn rhedeg
Fe/Mi fydd e/o	He (south/north variation)	Fe/Mi fydd e yn rhedeg
Fe/Mi fydd hi	She	Fe/Mi fydd hi yn rhedeg
Fe/Mi fyddwn ni	We	Fe/Mi fyddwn ni yn rhedeg
Fe/Mi fyddwch chi	You (plural)	Fe/Mi fyddwch chi yn rhedeg
Fe/Mi fyddan nhw	They	Fe/Mi fyddan nhw yn rhedeg

Table 9. Example of auxiliary verb construction in Welsh future tense.

In colloquial Welsh, these periphrastic forms are used more frequently than inflected verbs, however, in literary Welsh, inflected verb constructions are seen more frequently (Fife, 1986). This is true for the majority of languages, where spoken dialects have a preference for periphrastic forms, particularly for ‘*to be*’ verb constructions. In other constructions, the main verb is inflected to mark tense, number, person and mood. For example, in VSO constructions (also referred to as the simple or short forms), the main verb is inflected to indicate tense (Jones, 1969). In these cases, the affirmative forms, negative forms and question forms are created by adding the appropriate inflectional suffix to the verb stem to denote tense. An example of this construction to indicate past tense is provided in Table 10. In this example, the word stem for ‘*rhedeg*’ meaning ‘*running*’ is ‘*rhed.*’ The suffix for all regular verbs is affixed to the word stem depending on the subject.

Word ending	Subject	Example
-ais i	I	Rhedais i
-aist ti	You	Rhedaist ti
-och chi	You (formal)	Rhedoch chi
-odd e/o	He (south/north variation)	Rhedodd e
-odd hi	She	Rhedodd hi
-on ni	We	Rhedon ni
-och chi	You (plural)	Rhedoch chi
-on nhw	They	Rhedon nhw

Table 10. Example of verb inflection in Welsh.

One difference between English and Welsh morphosyntax concerns the plural system. The plural system of Welsh follows a different pattern to that of the English plural system. In contrast to English, in which nouns are commonly inflected with a suffix to create the plural form, plural nouns in Welsh can be shorter than the singular form. For example, the word for 'tree' is 'coeden', whereas the plural form, 'trees' is 'coed'. In contrast to this, there are some similarities between Welsh and English such as the frequency and distribution of consonant clusters. There are typically similar numbers of clusters found in the two languages, and they can also occur before or after a vowel within words in both languages.

On the whole, there are many similarities but also many differences between Welsh and English. There is considerable evidence that English has had some influence on the Welsh language (e.g. word borrowings) due to the vast majority (if not all) of Welsh speakers also being speakers of English. The understanding of the features of Welsh will be useful in the remainder of this thesis, particularly with reference to designing the phonological awareness stimuli (see section 4.1.2) and later interpretations of the results of the Welsh language assessments, and in exploring the possibility of any cross-language transfer effects. This information is particularly useful in considering that children with DS have specific difficulties with morphosyntax, and specifically verb inflections (see section 3.2.2).

1.1.4 Is Bilingualism Beneficial or Detrimental?

There has been much debate over the last few decades regarding the impact that a second language has on language development and cognitive functioning in TD children and adults. This debate will be discussed throughout the following section. Studies conducted in this area typically compare monolinguals and bilinguals on measures of language skills and cognition. It is worth highlighting that it is important to consider language competencies in all the languages of a bilingual or multilingual speaker (Paradis, Genesee & Crago, 2011). As stated earlier, Grosjean (1989) emphasized that a bilingual is not simply 'the sum of two monolinguals', as a bilingual will receive exposure to two languages with varying degrees of exposure to each language. This varying exposure to each language, combined with the other factors mentioned earlier (i.e. the context of exposure and age of acquisition, etc.), will result

in each bilingual having their own unique linguistic profile. In addition to this, the concept of a truly balanced bilingual appears to be thought of as the ideal, however, in reality, this is often not the case with one language typically being stronger than the other. It is also highlighted that measuring the language profile of a bilingual is a difficult task, as utilizing measures that are designed for use with monolinguals may not provide an accurate picture of a bilingual's true language abilities.

One possible influence on linguistic performance is the timing of exposure to a second language, as later exposure may result in a lower level of fluency in the second language. This was reviewed in a meta-analysis by Callan (2008) where eight studies were analysed in terms of language outcomes in bilinguals. Although the measures used varied from vocabulary to phonological development, overall, no delay was found in language learning in the papers that reported on simultaneous bilinguals. The remaining studies which reported on sequential learners found some group differences in favour of the monolinguals. This suggests that there are better language outcomes for simultaneous bilinguals, and research that assesses the linguistic abilities of bilinguals should endeavour to acknowledge the potential differences between sequential and simultaneous bilinguals. Therefore, it is also important to consider any potential differences that may exist between sequential and simultaneous bilinguals. The following sections will explore vocabulary, phonological and morphosyntactic development, metalinguistic awareness, executive function and cognitive decline in bilinguals and monolinguals.

Vocabulary

The initial debate surrounding the impact of bilingualism began when it was proposed that individuals who are exposed to two languages have a weaker grasp of language, specifically in terms of vocabulary sizes than monolinguals, as they have less time dedicated to developing these skills in each language. This idea led to the belief that parents should avoid exposing their children to multiple languages as this could lead to interference between the languages, particularly if they differ vastly in terms of linguistic characteristics (Lado, 1957). Recent research has, however, contradicted these claims for vocabulary sizes, as it has been reported that infants exposed to two languages acquire age-appropriate levels of language in both of their languages, meaning that their vocabulary sizes fall within the expected range after considering chronological age (Poulin-Dubois et al., 2011). In addition to this, research has also found that other early language development stages are met, for example, the age of first word not being significantly different for monolingual and bilingual infants (Genesee, Paradis & Crago, 2004).

In contrast, a study by Bialystok & Viswanathan (2009), reported that the vocabularies of bilinguals are not as diverse or as extensive as that of monolinguals, even when the bilinguals are considered to be fluent in both of their languages. More recent research has found that the vocabulary size of a bilingual in each of their languages is smaller than that of a monolingual, however, the conceptual vocabulary size of bilinguals is reported to be equal

or greater than that of monolinguals (Bialystok et al., 2010). In this large scale study by Bialystok and colleagues, it was reported that any initial deficits found in each of the languages of a bilingual do not lead to any significant differences in terms of academic achievements.

Phonological Development

In terms of phonological development, research suggests that there may be some cross-linguistic interaction between a bilingual's two languages. Cross-language transfer has been proposed for several aspects of language, however, phonological development has gained substantial attention in terms of bilingualism. It has been proposed that there are three ways in which bilingualism may impact language development in bilinguals as described by Paradis and Genesee (1996), which are acceleration, delay or interference and transfer. It is also proposed that any cross-linguistic influence will also be dependent on the languages being acquired, with similarities between the languages (e.g. being part of the same language family) resulting in transfer or acceleration (also described as positive transfer; Arabski, 2006), and dissimilar languages more likely to produce interference or delay in language acquisition.

For phonological development specifically, research has proposed that two phonological structures may result in a cross-linguistic transfer, which could be one way or reciprocal. Recent research suggests that phonological development does transfer at low levels, and may even be accelerated by bilingualism (Fabiano-Smith & Barlow 2010; Tamburelli, Sanoudaki, Jones & Sowinska, 2015). For example, Tamburelli and colleagues investigated consonant clusters in Polish-English bilingual children in comparison to a language-matched English monolingual control group. As Polish has more complex word-initial clusters and a greater range of clusters, it was hypothesised that this would lead to cross-linguistic transfer for the bilinguals. Results showed that the bilingual participants outperformed the monolinguals for some word-initial clusters, suggesting that the exposure to a more phonologically complex language (Polish) led to an acceleration of less complex structures in the other language (English). This example provides evidence that phonological development can be influenced by, and even be enhanced by bilingualism.

Morphosyntactic Development

A further aspect of development that has been explored in bilingual populations is morphosyntactic development. Morphosyntactic development can be a complex process and will also often depend on the languages being acquired. Some research studies have reported advantages in favour of bilingual children in terms of morphological awareness (Schwartz et al., 2016). Schwartz and colleagues (2016) investigated cross-linguistic transfer of morphological awareness in six-year-old Arabic-Hebrew bilinguals (with two groups of bilinguals with either Arabic or Hebrew as an L1). Results showed that each bilingual group performed significantly better than each of the monolingual groups who had been exposed to the same language as the bilingual group's L1 on tasks that measured inflectional

morphology in non-words. In addition, positive transfer was reported from Arabic to Hebrew for some morphological categories with the authors suggesting that this was due to some common “core linguistic features” of Arabic and Hebrew.

In contrast to this finding, studies have also reported that bilinguals are outperformed by monolinguals on measures of morphosyntactic development (Thordardottir, 2015). In a recent study by Thoradardottir, grammatical development was investigated in French-English bilingual and monolingual children who were aged three or five-years-old. Grammatical development was measured by MLU as well as the diversity and accuracy of morphemes used in spontaneous speech samples. Findings showed that children with equal experience in both English and French performed equally as well as monolinguals in both of their languages, however, bilingual children who had received unequal exposure in their languages performed significantly lower in their weaker language. The authors suggest that this finding highlights that for young bilingual children, the amount of input in each language is important for morphosyntactic development as grammatical ability in each language was influenced by language experience.

Metalinguistic Awareness

Bilingualism has also been reported to enhance various aspects of metalinguistic awareness (Bialystok & Viswanathan, 2009; Adesope et al., 2010; Barac & Bialystok, 2012). Metalinguistic awareness is defined as the ability to reflect on language separately to meaning, and consciously reflect on language in an objective manner. Encompassed in the term of metalinguistic ability are a number of abilities which include semantic, syntactic and phonological awareness. Researchers have found advantages for bilinguals in many of these areas and these will be discussed below. As bilingualism and phonological awareness will be explored as part of this research, advantages in phonological awareness will be discussed separately in more detail in section 2.1.2.

A large recent meta-analysis reported that the majority of studies that have investigated metalinguistic awareness in highly-proficient bilinguals report that bilingual groups often outperform monolingual control groups (Adesope et al. 2010). Advantages for bilinguals on measures of semantic awareness have been reported by Reder and colleagues (2013) in a study that involved several measures of metalinguistic awareness. In this study, French six and seven-year-old children learning German were compared to French monolinguals. Syntactic awareness and morphological compounds were found to be enhanced in the group of children who were second language learners of German, suggesting that even fairly minimal exposure to a second language was enough to lead to these advantages in this circumstance. No evidence of any bilingual benefit was found for phonological awareness or morphological affixes. The authors propose that the phonological similarities between French and German may be the reason why these enhancements were found for some aspects of metalinguistic awareness but not others.

Opposing findings were reported in an earlier study by Chiappe and Siegel (1999) which also investigated syntactic awareness. In this study, children of a similar age to those in Reder and colleague's study (six and seven-year-olds) were assessed on a range of tasks that assessed metalinguistic awareness. One of these tasks was a syntactic awareness measure where the participants were required to provide the missing word in a sentence. Results found that the English monolinguals displayed a greater performance than the bilingual children. It was highlighted that the children in this study had difficulties with English syntax and did not show any evidence of heightened syntactic awareness. Findings from this study showed that other aspects of metalinguistic awareness were similar between the groups of bilinguals and monolinguals. These proposed benefits have gained a substantial amount of attention in the past couple of decades and have created a substantial debate as to whether there is an increase in these metalinguistic abilities by being bilingual, and some have suggested that different skills and different tasks may be the reason behind these mixed results (Altman et al., 2018).

Executive Functioning

Executive functioning is an area that has gained the most amount of attention in the literature in relation to bilingualism. Executive functioning refers to a cluster of cognitive abilities including planning, focussing, inhibiting and self-monitoring. Proposed advantages in these abilities have been the focus of a number of research studies, some of which will be discussed in this section. One skill in particular within this cluster which is essential for any bilingual speaker is the ability to direct attention to relevant information and suppress irrelevant information (i.e. the alternative language that is not being used) during any task or activity, which is often referred to as executive control or cognitive control.

An example of this proposed executive functioning benefit was provided by Barac and Bialystok (2012). In this study, the authors examined executive functioning abilities in bilingual and monolingual six-year-old children, who were assessed on a range of executive functioning tasks. Findings revealed that the bilinguals outperformed the monolinguals on measures of grammatical judgement, executive control as well as having larger conceptual vocabulary sizes than the monolinguals. Similar findings were reported by Carlson and Meltzoff (2008) who found that bilinguals also outperformed monolinguals on tasks requiring controlled attention after controlling for socioeconomic status, verbal abilities and age. Interestingly, these bilinguals also outperformed a group of children who were classified as monolinguals but who were enrolled in a second language immersion kindergarten, suggesting that sufficient exposure and/or sufficient proficiency is required in a second language for executive function benefits to emerge.

A recent meta-analysis was conducted which included a range of studies that investigated the cognitive skills of bilinguals and monolinguals (Adesope et al., 2010). This study combined the findings from 63 studies (with a total participant number of 6,022). Findings of this analysis showed that there were no disadvantages for the bilingual

participants, however, some of the papers reviewed found evidence of a bilingual benefit. It was highlighted that the bilinguals in many of the studies included in this review had greater metalinguistic awareness, greater attentional control and enhanced working memory. Another review of the literature was conducted by Bialystok in 2001, which also reported cognitive benefits for bilinguals on measures of selective attention and cognitive flexibility.

A more recent meta-analysis of executive functioning in bilinguals was undertaken by Lehtonen and colleagues (2018). This review solely focussed on studies involving adults in order to evaluate the impact of bilingualism on six domains of executive functioning. This review aimed to synthesise the findings of both published and unpublished studies in order to ascertain if there is a publication bias in favour of research that reports a bilingual advantage. Although small effects of a bilingual advantage were found for inhibition, shifting and working memory, these effects disappeared after controlling for observed publication bias. The authors of the study conclude that the evidence documented in these 152 studies showed that there was no support for the proposal of a bilingual advantage for executive function, at least in bilingual adults. This study, therefore, questions the notion of a bilingual advantage for these cognitive domains and suggests that the limited published research in this field may be due to publication bias. The studies included a range of bilinguals with English as one of their languages and 30 different languages being the additional language, suggesting that their null findings were not simply due to the language pairings included. This view was earlier proposed by Paap and Greenberg (2013) who reported that there was no consistent evidence of a bilingual advantage in their three studies.

Other researchers have also expressed a need for caution when interpreting the results of studies that find a bilingual benefit, as further research has not successfully replicated the finding of a bilingual advantage (Gathercole & Baddeley, 2014; Valian, 2015). Within the Welsh context, a study of Welsh-English bilingual adults and English monolingual adults was conducted which comprised of 40 measures of executive control (Hindle et al., 2015). In this study, only nine of these measures favoured the bilingual participants and in contrast, the monolingual group outperformed the bilinguals for the majority of the executive control measures. As the monolinguals outperformed the bilinguals on the majority of measures, it was concluded that bilingualism did not result in an advantage on executive functioning.

Further research has been conducted within the context of the Welsh language, whereby researchers have sought to identify if there is any evidence of a bilingual advantage in executive functioning and metalinguistic awareness abilities in Welsh-English bilingual speakers (Gathercole et al., 2014). This large-scale study comprised of 650 participants ranging from children aged 3 through to older adults. The researchers assessed the cognitive and executive functioning abilities in monolinguals in comparison to fully fluent Welsh-English bilinguals who were either English or Welsh dominant but whose cultural and educational backgrounds were fairly homogenous. The authors concluded that there was no conclusive evidence to support the proposal of a bilingual advantage in terms of accuracy or reaction

times across a range of ages and a range of conditions. Alternative patterns were found, such as an increase in performance with age in children (as expected), and a decrease in performance in older adults (also as expected).

Mental Flexibility and Social Development

Reports also suggest that mental flexibility is enhanced by exposure to two languages. For example, Lazaruk (2007) reported enhanced creative thinking and mental flexibility in bilinguals who were students in a French immersion programme in comparison to monolinguals. This study reported that the students had high levels of language proficiency in English and French, and it is proposed that these high levels of fluency were due to the language immersion setting. It was suggested that these high levels of language proficiency subsequently led the bilinguals to have enhanced communicative sensitivity. Furthermore, Romaine, Bhatia and Ritchie (2004) suggest that there is a further benefit to bilingualism, as by having access to a second language, there may be access to different social groups that assist in forming identities within heritage communities, which is reported to result in social benefits. Further benefits have also been reported as a result of bilingualism, such as acceleration of conflict resolution at the age of four (Bialystok & Martin, 2004), and even at the age of two (Poulin-Dubois et al., 2011).

Cognitive Decline

Research also suggests that bilingualism might have an impact on cognition in later life. Studies report that the onset of cognitive decline is later in bilingual speakers (Bialystok, Craik & Freedman, 2007) and that the severity of cognitive decline and dementia is reduced in adults who are bilingual speakers (Hermanto, Moreno, & Bialystok, 2012; Bak et al., 2014). This is of particular relevance to the population under study in this thesis as early-onset dementia also has a high prevalence rate in those with DS. If bilingualism does act as a defence mechanism against the onset of this cognitive decline, it is interesting to speculate whether these advantages would also extend to adults with DS.

Studies have been conducted to examine the executive control abilities and cognitive functioning in older adults with early-stage Alzheimer's disease in Welsh-English bilingual speakers in comparison to English monolingual controls (Clare et al., 2016; Hindle et al., 2015). In these studies, no evidence of bilingualism acting as a protective mechanism was found. Performance on language testing was in line with the degree of exposure to each language (i.e. English monolinguals outperforming Welsh-English bilinguals on measures of English), however, no executive functioning benefit was reported in these older adults.

Clare et al. (2016) also did not find any significant differences in the age of Alzheimer onset between the Welsh-English bilinguals and monolinguals in their study, although the bilinguals did receive a diagnosis on average, three years later than the monolinguals. This might mean that bilingualism does not act as a complete protective mechanism against dementia, but may be able to delay its onset to some extent. Both these studies suggest that

there may be factors specific to the linguistic context in Wales which reduces or masks these proposed benefits of bilingualism found in other populations. The authors do stress the need for further research which considers the heterogeneous nature of bilingualism.

Theoretical Accounts

Several proposals have been suggested in order to justify the proposal of a bilingual benefit. The initial theory of a bilingual advantage was proposed by Green (1998) who suggested the “inhibitory control model”, which reports that the constant inhibition of one language leads to developing cognitive control which is then extended to a range of other tasks requiring a degree of control. This concept has gained further support and has been developed further by Bialystok and colleagues, who similarly suggest that this bilingual advantage emerges as a result of having to continuously inhibit competing information (i.e. the other language), particularly in the situation of naming objects. This then results in carry-over effects into other aspects that require attending to relevant stimuli and inhibiting irrelevant stimuli. The development of these cognitive enhancements is subsequently reported to lead to a protective mechanism against cognitive decline and postpone the onset of dementia (Bialystok, Craik & Luk, 2012).

Researchers have also speculated as to why these benefits are not always observed, particularly in the Welsh language context. Gathercole and colleagues propose that the linguistic situation in Wales specifically (as described in section 1.1) may have led to their findings. The authors suggest that the bilingual context in Wales may promote greater automaticity in linguistic processing between the two languages within fully fluent bilingual communities where the two languages are often used interchangeably. As a result, the fully fluent bilinguals in this study did not show superior levels of cognitive control as their two languages are both “online” at all times and these bilinguals are not required to monitor their linguistic surroundings to the same extent within these fully bilingual communities. This proposal would also explain why no differences were found between bilinguals and monolinguals on measures of cognitive decline. If cognitive control is not enhanced within these bilingual speakers, this would not lead to the development of protective factors against cognitive decline.

Summary

Although there is an ongoing debate regarding the possibility of a bilingual benefit, particularly within the Welsh language context, there certainly does not appear to be any cognitive disadvantages to bilingualism. If there are benefits for some individuals, it could also be proposed that these could transfer to children with language impairments, and enhance their skills in areas where they might have specific deficits. Within individuals who have DS, there may be cognitive advantages found in these individuals if they are bilingual, and any evidence of cognitive advantages may have more substantial significance as cognitive functioning is typically lower in these individuals.

Chapter 2: Language Development and Developmental Disabilities

2.1 Typical Language Development

The following sections will look at more specific areas of language acquisition and will address phonological development, vocabulary and phonological awareness in TD monolingual populations. Although language development is highly variable, there are often patterns and stages of development that children will experience during language acquisition. Early studies in the field of language perception in infants suggested that even at two days of age, infants are capable of discriminating between sounds which constitute of language or not (Ruben, 1997). This suggests that infants are born with the innate ability to recognise language. In terms of production, children will typically produce their first word around their first birthday (Clark, 2009), however, there are some earlier stages in the vocal development that are typically observed in infants which will be discussed in the following section.

The first pre-vocal development has been described as reflexive vocalizations (Stark, 1980), which occur during the first few weeks of an infant's life, whereby the infants' vocal cords are stimulated through breathing and sucking. During an infant's first 6-16 weeks, is the time in which laughter begins to emerge and cooing sounds also become apparent which often resembles the sounds of vowel productions. The stage following this is termed vocal play, during which many sounds begin to emerge including vowels and consonant-like productions, and following this is reduplicated babbling (also known as canonical babbling). During this stage (at 6-10 months), infants begin to combine consonant and vowel-like sounds, often in a CVCV structure which are reduplicated. At this stage, infants also show influences from the languages that they're exposed to, as they more often produce consonant and vowel-like sounds of the primary language of exposure (Whalen, Levitt & Goldstein, 2007). The final stage of phonological development before speech is known as non-reduplicated babbling, in which syllables begin to emerge with a further combination of different vowels and consonants. At this stage intonation and stress patterns also become part of an infant's productive abilities. Following these stages, the first word emerges around the 12-month milestone.

In addition to these stages of vocal development, there are further trends found in terms of time parameters, and progression for phonological development within young children. Firstly, the accurate acquisition and production of vowels typically precede consonant production, with a 36-month old infant generally having mastered all vowels and achieving high accuracy levels (97.39%, Dodd et al., 2003) but not for consonants. Consonant production continues to develop until around the child's sixth birthday (Doorn & McLeod, 2002) and the correct pronunciation of consonants may fluctuate with an infant producing a correct sound in one instance and incorrectly in another. In addition, sounds that occur more

frequently in a given language will typically be acquired quicker than sounds that are more uncommon or rarely used (Ingram, 1989; Edwards, Beckman & Munson, 2015). The acquisition of the phonology of a language is a complex process and as a result, children often employ simplification processes in which the pronunciation of certain sounds may be altered. Examples of these are presented in Table 11 below (adapted from Peña-Brooks & Hedge, 2000 and Williamson, 2016). These simplification processes presented in the table generally involve either a systemic process or a structural process. A systemic process involves the alteration of a speech sound whereby it is usually replaced with another sound as in the example presented of 'fronting' where a back consonant sound is replaced by a consonant where the place of articulation is at the front of the mouth. In contrast, structural processes describe simplification processes that alter the syllable structure of the word being produced. One example of this which is presented in the table is the early occurring process of reduplication. In this process, the child repeats the same syllable within a target word, for example, the word 'bottle' (/bɒtəl/) which might be simplified to /bɒbɒ/.

One final important consideration in phonological development concerns the debate of whether the errors or developmental patterns found in infant language are due to an inaccuracy in comprehension versus production. In considering production errors, the possibility of an auditory perception inaccuracy of language in infants should be considered, however, there is substantial evidence that suggests that infants are capable of perceiving adult language forms. Errors found are, therefore, attributed to production difficulties (Berko & Brown 1960; Ingram, 1989). For example, Berko and Brown studied a child who had difficulty producing the word 'fish'. When an adult repeated the pronunciation of the child in the sentence 'is that your fis?' he replied no, but when he was asked 'is that your fish?' he responded 'yes, my fis'. In addition, another child had pronunciation issues with words such as card/cart and jug and duck, however, the child was able to correctly identify the items when shown pictures of the objects.

Systemic Processes	Definition	Example	Typically Ceases
Fronting	Replacing back consonant with front consonant	<i>cup</i> /kʌp/ → /tʌp/ <i>fish</i> /fɪʃ/ → /fɪs/	3/4-years-old
Backing	Replacing a non-velar or non-glottal consonant with a velar or glottal consonant	<i>duck</i> /dʌk/ → /kʌk/ <i>bad</i> /bæd/ → /bæɡ/	Phonological delay
Stopping	Replacing continuant consonant with a stop consonant	<i>sun</i> /sʌn/ → /tʌn/ <i>love</i> /lʌv/ → /lʌb/	3/4/5-years-old
Frication	Replacing an approximant with a fricative	<i>you</i> /ju/ → /zu/ <i>red</i> /rɛd/ → /ðɛd/	4-years-old
Gliding	Replacing a continuant (especially a liquid sound) with a glide	<i>leaf</i> /li:f/ → /ji:f/ <i>red</i> /rɛd/ → /wɛd/	5-years-old
Labialization	Replacing tongue tip consonants with labial consonants	<i>dog</i> /dɒɡ/ → /bɒɡ/ <i>moth</i> /mɒθ/ → /mɒf/	5-years-old
Vowel harmony	Replacing unstressed vowel in multisyllabic word with the primary stressed vowel	<i>Peter</i> /'pitə/ → /piti/ <i>Agree</i> /ə'ɡri/ → /igri/	-
Consonant harmony	A target consonant assumes the place of articulation of a trigger consonant across an intervening vowel	<i>coat</i> /kəʊt/ → /kəʊk/ <i>top</i> /tɒp/ → /pɒp/	-
Voicing Change	Replacing voiceless consonants with voiced consonants and voiced consonants with voiceless consonants	<i>sun</i> /sʌn/ → /zʌn/ <i>nose</i> /nəʊz/ → /nəʊs/	6-years-old
Feature Synthesis	Combining the features of two segments to yield a single different segment	<i>spoon</i> /spun/ → [fun] <i>sleep</i> /slip/ → [tɪp]	-
Structural Processes	Definition	Example	Typically Ceases
Unstressed syllable deletion	Least stressed syllable omitted	<i>tomato</i> /tə'mɑ,təʊ/ → /mɑ,təʊ/ <i>elephant</i> /'ɛlə,fənt/ → /'ɛ,fənt/	4-years-old
Reduplication	Repetition of syllable from target word (total or partial)	<i>doggie</i> /dɒɡɪ/ → /dɒdɒ/ <i>bottle</i> /bɒtəl/ → /bɒbɒ/	2/3-years-old
Metathesis	Rearrangement of two consonants in a syllable.	<i>ask</i> /ɑ:sk/ → /ɑ:ks/ <i>star</i> /stɑ:/ → /sɑ:t/	-
Epenthesis	Insertion of a vowel within a cluster	<i>blue</i> /blu:/ → /bəlu:/ <i>film</i> /fɪlm/ → /fɪləm/	-
Consonant deletion	Consonant/consonant cluster omitted	<i>ball</i> /bɔ:l/ → /ɔ:l/ <i>cup</i> /kʌp/ → /kʌ/	3-years-old
Cluster reduction	Omitting consonants in a consonant cluster	<i>spot</i> /spɒt/ → /pɒt/ <i>bring</i> /brɪŋ/ → /ɪŋ/	3/4-years-old
Unstressed syllable deletion	Least stressed syllable omitted	<i>tomato</i> /tə'mɑ,təʊ/ → /mɑ,təʊ/ <i>elephant</i> /'ɛlə,fənt/ → /'ɛ,fənt/	4-years-old

Table 11. Phonological processes which are involved in the simplification process of typical child phonological development. *Note:* Adapted from Peña-Brooks and Hedge (2000) and Williamson (2016).

2.1.1 Expressive and Receptive Vocabulary

This section will explore receptive and expressive vocabulary in TD children. Vocabulary is often described as a single entity, however, expressive and receptive vocabulary abilities are often referred to separately as children who are in the process of acquiring language are often able to comprehend words and utterances that they cannot yet produce. This section will explore these two related but separate abilities in TD children. Vocabulary is a key foundation for later language development and is also closely associated with reading development (Moghadam et al., 2012), comprehension and phonological awareness (Walley, 1993, see section 2.1.2). Studies in the field of vocabulary knowledge examine the breadth of vocabulary (i.e. the number of words known) as opposed to the knowledge of pronunciation, spelling, or morphology. This section will be useful in comparing typical receptive and expressive vocabulary abilities compared to children with a DD and more specifically children with DS.

Typically developing children tend to produce their first word around their first birthday following the babbling stages described above in section 2.1. Following this, children produce more and more words and begin combining words around 24 months of age (American Academy of Paediatrics, 2017). There is also a documented vocabulary spurt between 15 and 24 months of age, whereby children suddenly acquire words at a much quicker rate than they had previously. Although vocabulary sizes are difficult to measure, Graves (1986) suggested that children aged five to six-years-old have a vocabulary size of between 2,500 to 26,000 words, evidencing the considerable variation between children. More recent and more accurate estimations of vocabulary sizes have provided narrower estimations. For example, Beck & McKeown (1991) reported that five to six-year-old children had vocabulary sizes of between 2,500 to 5,000 words. This study also suggested that TD children tend to acquire around 3,000 new words each year during primary education.

One factor that has been investigated in relation to its role in vocabulary development is memory. Researchers have explored whether differences in memory abilities are partly responsible for accounting for differences in vocabulary sizes between children. Early studies in this field suggested that semantic memory deficits were responsible for some of the difficulties that some children have in learning the meanings of new words (Swanson, 1986). This proposal was suggested to also explain some of the deficits seen in children with intellectual impairments as these children performed less well when required to learn words that were associated semantically, structurally or phonemically. The author proposed that children with intellectual impairments processed new words differently to TD children and that TD children used "knowledge or accumulation of facts about words which become increasingly accessible by means of well-trodden information processing routes" (p. 485).

In addition, studies have highlighted the vocabulary gap between children from different socioeconomic backgrounds, with those children from higher SES backgrounds outperforming children from lower SES backgrounds. This finding has been replicated a number of times suggesting that SES has a robust impact on vocabulary development. For example, Hart & Risley (1995) found that children at the age of three-years-old knew 600 more words on average if they were in a high SES group, compared to children from a low SES background. This vocabulary gap between children from high and low SES backgrounds also seems to continue and to widen as children progress through schooling. Biemiller & Slonim (2001) reported that children from high SES backgrounds in their fifth grade (aged 10-11) had an even greater advantage over those from low SES backgrounds with them knowing 4000 more words on average. Therefore, it will be important to include a measure of SES in the present research in order to account for any differences in vocabulary between groups.

2.1.2 Phonological Awareness

Phonological awareness (PA) is concerned with the detection and manipulation of sounds within a language at three levels; syllables, intrasyllabic (onsets/rhymes) and phonemes. Following on from the earlier section which discussed cognitive and metalinguistic benefits of bilingualism, PA is also considered a metalinguistic skill as it requires conscious awareness and reflection of linguistic properties. Most TD children begin to develop PA at three years of age (Lieberman et al., 1974), although some children have been found to have some awareness of the patterns of language even earlier than this by scoring above chance level on PA tasks at age two (Lonigan, Burgess, & Anthony, 2000).

Tasks that are used to measure and assess PA are detection and manipulation tasks which usually include listening tasks, syllable structures tasks, onset/rhyme tasks and phonemic tasks. The general progression of PA (which is relatively robust) is that the acquisition of larger units typically occurs prior to the acquisition of smaller units. In line with this, whole word awareness will typically precede awareness of syllables and phonemes (Caravolas & Bruck, 1993; Fox, & Routh, 1975; Treiman & Zukowski 1991). In addition to this, children are able to detect phonological distinctions before they are able to manipulate them themselves, and also learn to blend items before they can segment them (Anthony et al., 2003). The development of PA is not completely linear, with elements being re-adapted as other areas are newly being acquired.

As a metalinguistic skill, PA requires the ability to separate the meaning or communicative function of language from the linguistic structure. Phonological awareness has been closely linked to a number of aspects of language development and is often correlated with vocabulary sizes (Chaney 1992; Metsala & Walley, 1998; Carroll, 2001). For example, Metsala and Walley (1998) reported that children were found to have greater performance on phonemic awareness tasks if they also had a large vocabulary size. Additionally, assessments of vocabulary, syntax, morphology, and expressive language have all been reported as accurate predictors of PA (Dickinson et al., 2003; Chaney, 1992). Smith

and Tager-Flusberg (1982) also reported an association between language ability and PA as they found that these were highly correlated with each other. More specifically, they reported that rhyme judgement tasks are correlated with all measures of language in their study. In terms of language development, Walley (1993) reported that vocabulary growth is one of the precursors to processing phonological representations when combined with language play and the onset of reading. More recently, Carroll (2001) also reported that PA was highly correlated with language at different stages of development.

There has been a growing interest in metalinguistic abilities in children and PA has gained a lot of this focus due to the reliability and robust finding of the correlation between PA and the development of spelling and reading. Research finds that reading skills in an alphabetic literacy are closely related to, and are reliably predicted by PA (Bruck & Genesee, 1995). This suggests a reciprocal relationship between phonemic awareness and learning to read an alphabetic script. As a result, PA is often targeted alongside literacy instruction in educational institutions, for example clapping out syllables within words. Finally, if there are impairments found in PA, interventions in this domain have been found to not only be successful in increasing PA but also enhance the speed of reading acquisition (Bradley & Bryant, 1983). Longitudinal designs have also been employed to measure the association between PA and reading, and these also report that PA is an accurate predictor of later reading skills (Bradley & Bryant, 1985). Further research supports this association between PA and reading, for example, Kolinsky and colleagues (1987) suggested that reading instruction was the most essential component to the development of PA, in testing literate and illiterate adults on speech perception tasks.

It is not clear as of yet, which component (if any) of PA has the most responsibility in the development of reading, or which unit is the most accurate predictor of literacy abilities. Researchers have suggested that the ability to rhyme may be the most dominant skill required for the development of reading (Goswami 1991; Goswami & Mead, 1992). It is suggested that children can use their knowledge of rhymes to apply these to unfamiliar words which share the same rhyme unit. This may particularly be the case with irregular languages, such as English, as there is generally more regularity within larger chunks (i.e. onsets and rhymes) as opposed to small units such as phonemes (Treiman et al., 1995). Furthermore, the decoding of larger chunks, as opposed to many smaller units, may reduce the constraints on working memory, as this may be an issue for younger children.

In addition to this, the language which is being acquired may also play a role in the development of PA in terms of the speed of development. For example, it has been reported that those who are acquiring English, efficiently develop rhyme awareness due to the consistent regularity of these larger units (Treiman et al., 1995). In addition, those acquiring two languages may lead to differences in acquisition as suggested by Lukatela et al., (1995) who report that languages with transparent orthographies (such as their Serbo-Croatian sample) result in the earlier development of PA, as these languages promote word-decoding skills (associated with reading), which in turn facilitates PA. The proposal that orthographic

transparency influences the development of PA has also been extended to phoneme awareness, with some research suggesting that this skill, in particular, may be enhanced in children who are learning to read a transparent orthography. For example, de Souza and Leite (2014) investigated PA in Portuguese-English bilingual and English monolingual children between the ages of seven and eight. They reported that the bilinguals outperformed the monolinguals, and this was particularly the case for phonemic awareness.

Furthermore, the finding that orthographic transparency influences PA has led to researchers investigating if these effects extend to reading abilities. Enhanced reading abilities have been reported in children who were learning a language with a transparent orthography. For example, Goswami et al. (1997) found that children acquiring Greek were significantly more accurate at reading Greek compared to the reading abilities in English of children who learning English. There was a significant distinction between the two groups with the Greek children at an almost 60% advantage compared to learners of English. This advantage was, however, found to disappear with age and the children learning to read English began to catch-up within a year of formal reading instruction. Additionally, Cossu and colleagues (1988) reported that children who were learning to read Italian had more advanced phoneme awareness skills than matched children learning to read English. In addition to this, the Italian children also outperformed the English learners on measures of syllable segmentation. Furthermore, the children who were learning to read transparent orthographies become competent readers within a year of formal reading instruction, however, those learning English were far less competent which highlights the role that reading has on PA. Finally, Öney and Durgunoğlu (1997) similarly stated that learning to read a language that has a consistent orthography results in quicker development of reading and this also extends to spelling accuracies.

Further evidence of the crucial role of PA in reading and spelling has been provided in a longitudinal study. Caravolas et al. (2001) reported on the reading and spelling development of 153 children who were aged three at the beginning of the study. The children were assessed at various time points following the initiation of formal education. The study aimed to explore the relationship between, and the predictive role of reading, phoneme awareness, memory, cognitive ability, letter-sound and letter-name knowledge on later reading and spelling ability in English monolinguals. It was reported that for spelling ability, the variables that were most closely associated were phoneme segmentation and letter-sound knowledge. More complex orthographic structures were reported to be learnt following this formal instruction of targeted reading and spelling in combination with experience. Consequently, the authors stress the importance of phonological skills in forming the foundations of reading and spelling above that of memory or general cognitive abilities.

This section has highlighted that PA has an important and crucial role in the development of reading, spelling as well as a range of aspects relating to language development. The necessity of developing PA is clear and also has extensive potential

implications on aspects of educational attainment, which may in turn impact on other areas of development.

2.1.3 Phonological Awareness and Bilingualism

After considering PA in monolinguals, the following section will compare the developmental trajectory of PA in monolinguals and bilinguals in order to identify any differences in the emergence of this metalinguistic ability. An interesting finding is that, similarly to other metalinguistic abilities (see section 1.1.4), PA is also found to be enhanced by bilingualism. Although the research is limited, the evidence that exists in the field reports that bilingualism enhances, or accelerates the development of PA (Campbell & Sais, 1995; Bialystok et al., 2003; Chen et al., 2004; Canbay, 2011). In addition to bilingualism, it has been suggested that the degree of proficiency in a bilingual's languages also impacts on the development of PA, specifically in relation to phonemic awareness and phoneme segmentation (Verhoeven, 2007) whereby those with a higher degree of proficiency in two languages appear to have the most enhanced PA abilities.

One of the first pieces of research that investigated PA in bilinguals reported an acceleration, with the bilinguals outperformed the monolinguals on all measures relating to semantic and PA (Campbell & Sais, 1995). This research aimed to investigate if a bilingual upbringing for children of nursery age resulted in an enhanced ability to manipulate sub-lexical sounds. All children in the study had English as their primary language, however, the bilingual children were also exposed to Italian. The bilinguals were required to have a bilingual parent and to have been exposed to two languages from an early age. The bilingual group were slightly younger on average than the monolingual group, however, the bilingual children still outperformed the monolingual children. This study did not, however, investigate the children's proficiency in their L2, and as a result, the authors suggested that perhaps this acceleration was due to a feature of Italian exposure, as opposed to bilingualism itself. This was proposed due to the differences between Italian and English, such as the more regular syllabic and phonological structure of Italian and the regularity of the syllable stress. This is suggested to have led to these advantages as the children in this study were exposed to a simpler and "more reliable" language. This is an important consideration as the authors note that none of the children were proficient in their second language, and as a result of this, the authors argue that even limited bilingual exposure is adequate for enhancements on PA to be observed.

Further research has provided support for these findings. In alphabetic languages such as English, there are three levels of PA as noted earlier; syllables, onsets/rhymes and phonemes. There is, however, a fourth level of PA in tonal languages such as Mandarin and Chinese, as the addition of tone awareness is an extra facet of PA for learners of these languages. In research conducted by Chen et al. (2004), PA was investigated in bilingual and monolingual children who received Mandarin instruction in school, with the bilingual children also receiving Cantonese input as their home language. The researchers tested two age

groups, second-grade and fourth-grade children on a variety of PA measures including onset awareness, rhyme awareness and tone awareness. The results displayed an advantage for the bilingual children with this group outperforming the monolinguals on the onset and rhyme tasks, however, these benefits appeared to disappear with age as they were only apparent in the second-grade children and not the fourth-grade children. This research does, therefore, suggest that PA is developed at a quicker rate in bilinguals, but at a later stage in development, the monolinguals catch-up in their PA abilities. The authors of this study also speculated that as the children were enrolled in an immersion programme in this study, this may be a further facilitative factor to the development of PA. Further support for this suggestion has been provided by Rubin and Turner (1989), where they also found that 6-year-old children enrolled in French immersion programmes also had enhanced performance on measures of phoneme segmentation.

Further research has been conducted which focuses on the development of PA in Spanish/English bilinguals. In this research, Dickinson et al., (2004) conducted a large scale study with 123 children between the ages of two and five-years-old who were from low-income families. This study tracked the growth and development of PA in both of the children's languages periodically and each testing session was scheduled over two days. The Early Phonological Awareness Profile (EPAP; Dickinson & Chaney, 1997) was employed to assess PA at both testing sessions. The authors found that the highest predictor of later PA was the PA in the second language assessed at the first session. As a result, the authors conclude that there is a strong transfer of PA in one language to the other and vice versa. This transfer effect has also been reported in older children (Carlisle et al., 1999). As a result, if there are transfer effects found for PA between languages in bilinguals, this may be one factor that accelerates the development of this metalinguistic skill if each language is acting as a facilitating factor in the second language.

One further study that has investigated PA in relation to bilingualism was conducted by Bialystok et al. (2003). In this study, there were three sub-studies that assessed whether there is a bilingual advantage in terms of PA. Only one out of the three sub-studies showed any bilingual benefit, and this was for the Spanish/English bilinguals. No bilingual advantage was observed for Chinese/English or French/English bilinguals compared to monolingual control groups in the other studies for any measures of PA. The authors offer a suggestion as to why these results were found, which was that the language of literacy instruction may impact the skills relating to PA. It is proposed that an alphabetic orthographic language system may be more straightforward in the development of PA. In contrast, learning a second language that is phonologically or/and orthographically different from the first language, or if the language has a more opaque orthography, this may result in an interference or a lack of acceleration.

This relates to the proposal of the 'orthographic depth hypothesis' as described by Katz & Frost (1992, p.150) as "differences in orthographic depth" that "lead to processing differences for naming and lexical decisions" when reading. As a result, languages that have

a more transparent orthography encourage readers to utilise a decoding word recognition strategy. This is not the case for orthographically deep languages such as English, whereby a logographic approach may be employed (i.e. identification of whole words as opposed to graphemes). They also report that all languages will have an element of decoding and that often, a mixture of strategies may be employed in practice. Therefore, it may be the case that learning to read different languages may result in different strategies being employed, as discussed in the previous section in relation to PA development. When it comes to bilingualism, the orthography of the two languages being learnt consequently needs to be considered. This has also been highlighted by Lukatela et al. (1995, p.464), whereby they also suggest that "learning to read a phonologically shallow orthography may be characterised by a more rapid development of phonological awareness and a correspondingly rapid development of word-decoding skills".

The proposal that PA may be enhanced in bilinguals has received substantial attention, however, it seems that there is an ongoing debate as to whether languages with different orthographic depths results in acceleration or deceleration of PA. Recent research has validated the proposal that learning two languages that have different orthographic depths will be beneficial in terms of PA development (Spencer & Hanley, 2003). This is also the only known study that has explored PA in Welsh-English bilinguals. In this research, the aim was to explicitly compare phonological skills and early reading abilities of children learning English and Welsh. This exploration provided a novel insight into PA in Welsh-English bilingual children, where the authors report that the bilinguals had enhanced abilities in terms of phoneme awareness in comparison to matched monolingual controls of the same age and who lived in similar geographical locations. In addition to this, the bilinguals were also significantly better at reading words and non-words at age five and six, compared to the children who only spoke English. The authors suggest that it was the orthographic transparency that exists in Welsh that enhanced PA as opposed to the fact that the children were bilingual, although they do note this as a possibility. Consequently, it is not clear whether the results were found are due to the orthographic transparency of the Welsh language specifically or bilingualism.

As previous research has found that PA is enhanced in Welsh-English bilinguals, further investigations into the cause and the factors that play a role in this advantage are required. In addition, Welsh is an alphabetic language that has high orthographic transparency, as it has a closer one to one matching of phonemes to graphemes in comparison to English. As suggested by Bialystok et al. (2003), a second language which has an alphabetic orthographic language system may facilitate a bilingual advantage of PA. Investigations of PA within the Welsh language context will extend understanding of this further due to the orthographic transparency of Welsh. It may be the case that transfer effects will increase PA in English (as non-orthographically transparent language) and vice versa.

In Wales, there are many children who become Welsh/English bilinguals by the means of immersion in nurseries and schools such as the MYM programme of early years' education

as discussed earlier (see section 1.1.2). Immersion programmes generally result in high levels of proficiency in two languages, which appears to be a facilitating factor in the development of PA (Chen et al. 2004; Verhoeven, 2007). As a result, these factors may play a role in enhancing the development of PA in Welsh-English bilinguals. Furthermore, the complexity of the languages in terms of consonant clusters is reported to play a role in PA development in bilinguals (Caravolas & Bruck, 1993). In their research, two groups of children were recruited who were learning two languages (Czech and English), and they were assessed on a battery of PA measures. The results indicate that the emergence of PA in terms of the recognition of complex and simple onsets is dependent on the phonological input and the orthography of the language being acquired. This was concluded as the Czech learners had a greater awareness of complex onsets, and the English learners had higher abilities in terms of simple onset awareness.

This is a particularly important finding for the current research given the mutation system that exists in Welsh (see section 1.1.3). Higher awareness of onset and initial phonemes may exist for Welsh-English bilinguals with the need to attend to initial phonemes. This would also help to explain the findings reported by Spencer and Hanley (2003) with Welsh-English bilinguals. On the other hand, the opposite might be true for those acquiring Welsh whereby onsets are less informative due to the inconsistency in initial phonemes due to mutation. This was reported in a study by Vihman and colleagues (2007) who found that Welsh monolingual infants did not show familiarity effects at 11 months whereas the English monolinguals did. The authors propose that word recognition may not develop to the same extent due to the fact that initial consonants in Welsh frequently change depending on grammatical conditions. This may be an interesting consideration later in this thesis.

Further studies investigating if bilingualism influences the development of PA have found mixed results. For example, Loizou and Stuart (2003) explored PA in four different groups of children which were; English-Greek Bilinguals, Greek-English bilinguals, Greek monolinguals and English monolinguals. The authors expected to find that both groups of bilinguals would outperform both groups of monolinguals, however, it was reported that only the English-Greek bilinguals children performed significantly higher than the other groups. It was proposed that this finding supports the concept of a bilingual advantage in terms of PA, but only for bilinguals whereby their second language is simpler in terms of phonology than the second language (i.e. the bilingual enhancement effect). This suggestion may also explain the lack of a bilingual advantage in other studies, for example, Chiappe and Siegel (1999) who examined Punjabi-English bilinguals and Bialystok and colleagues (2005) who included Mandarin-English bilinguals. Finally, in a recent study conducted by Kuo et al. (2016), a lack of a bilingual advantage in terms of PA was also reported in both English-Japanese and Japanese-English bilinguals in comparison to monolinguals, which disputes the proposal of a bilingual benefit for PA altogether.

In conclusion, there is a significant body of research reporting that bilingualism enhances the development of PA, particularly in orthographically transparent languages such

as Welsh, although it is also important to highlight the number of studies that have not documented the same results. It may be important to consider the studies that have not found evidence of a bilingual advantage in order to ascertain in what circumstances and what contexts these are usually reported. Typically developing children aged five and six appear to show the greatest phonological awareness benefits (Marinova-Todd et al., 2010) which will be important in informing the target sample in the current study and the interpretation of the findings.

2.2 Language Development in Bilinguals with Developmental Disabilities

In order to explore the language abilities of children with DS who are bilingual, it is also justifiable and valuable to consider the literature that exists concerning bilingual children with other language impairments. The findings from these studies will inform research on children with DS as the findings of these studies may transfer to different populations. Skills such as grammar and vocabulary typically cause difficulties in children with language impairments (Leitão, Hogben & Fletcher, 1997; Fletcher & Buckley, 2002; Miller, Leonard & Finneran, 2008). As a result, it may be even more important to identify the role of bilingualism on the development of these areas specifically for those with a DD, and consequently areas of specific weakness will be highlighted throughout the following sections. Although children with DS have a specific language phenotype, meaning that findings of research concerning children with other DDs cannot simply be generalised to this population, the findings reported in this chapter will guide the present research and provide useful information as to the potential outcomes of research concerning bilingual children with DS. Children with DS will not be included in this review as this is the focus of Chapter 3.

Additionally, Chapter 7 will explore bilingual children with a dual DS and ASD diagnosis, meaning that exploration of the literature relating to bilingual children with ASD is even more relevant and will be particularly informative to the design of this research and the interpretation of these findings. Furthermore, research conducted with bilingual children with other DDs will inform the methodology of the present research (see Chapter 4), such as the suitability of various assessments and employing appropriate control groups. This section will explore the language abilities of bilinguals with DDs including Autism Spectrum Disorders (ASD), Williams syndrome (WS) and Developmental Language Disorder (DLD; formerly known as specific language impairment; SLI). The vast majority of available research to date in the field of bilingualism in DDs has focused on bilingual children with ASD. This section will also briefly explore the support and advice given to parents or caregivers of a child with a DD concerning bilingualism.

One of the most recent and comprehensive reviews of bilingualism for various DDs has been conducted by Kay-Raining Bird et al. (2016). This paper also highlights the current gaps in the literature and suggests certain criteria that should be met in any future investigation of DDs and bilingualism. These include information relating to the languages spoken, age of exposure, duration and consistency of exposure, frequency of current

exposure, language proficiency, social status of language, day-care setting language, school program attendance, and the language of instruction (LOI). These recommendations were made to enable comparisons between the research that exists to date and any future research in the field. This review also provides recommendations for future research (particularly in relation to DS which is lacking), which includes different populations, longitudinal designs and information regarding sequential bilinguals.

2.2.1 Autism Spectrum Disorders and Bilingualism

The disorder that has gained the most attention in the field of bilingualism literature to date is Autism Spectrum Disorder (ASD). ASD is a lifelong developmental disability, in which individuals vary in impairments that usually comprise of social interactions, verbal and non-verbal (NV) communication and restrictive behaviours. Deficits in language are often a core and defining feature of ASD and are one of the factors within the triad of impairments that were previously used to make a diagnosis of ASD. The current criteria for an ASD diagnosis include “persistent deficits in social communication and social interaction across multiple contexts” alongside “restricted, repetitive patterns of behaviour, interests, or activities” (DSM-5; American Psychiatric Association, 2013). There must also be clinically significant impairments that are not better explained by an intellectual disability or a global developmental delay. The causes leading to ASD are not yet clear, however, it is believed to be a combination of environmental and genetic factors.

Delayed language onset is often one of the first signs identified by parents as an indication that an infant may have ASD. The lack of language is also reported to be one of the biggest concerns for parents when their child does not reach one of the big milestones; their first word, within the typical time period of 12-18 months (De Giacomo & Fombonne, 1998). Communication difficulties are often associated more with ASD than with other DDs. Furthermore, individuals with ASD often also have impairments related to social development. In terms of verbal and non-verbal communication, it has also been reported that children with ASD make significantly less communicative acts both verbally and non-verbally (Shumway & Wetherby, 2009). It is also important to note the distinction between children who have either High Functioning Autism (HFA) and Low Functioning Autism (LFA). Although there is no distinction made between the two in the DSM-5’s diagnostic criteria, clinicians and researchers often categorize children according to these labels as the clinical presentation, support and the prognosis is different for each (de Giambattista et al., 2019). The term LFA is usually used to describe children with ASD who have an IQ score of below 70, and HFA for those above 70. The linguistic profiles of each may also differ according to this distinction.

As reported above, many children with ASD experience language delays with over half of all individuals with ASD having language impairments of some form (Aarons & Gittens, 1999). It is not the case that all children with ASD will have language delays or impairments and some individuals with ASD have high competencies in language (especially females with

high-functioning ASD; Sturrock et al., 2019). A quarter of all individuals with ASD will remain NV throughout their lives (Lord et al., 2004) with many others being classified as ‘minimally verbal’ (Tager-Flusberg & Kasari, 2013). It is important to establish the aspects of language development that impairments usually impacted in children with ASD. The distinction between children with ASD who display these language delays and impairments is often referred to as ASD with or without language impairment (ALI and ALN, respectively). Although again these distinctions are not made in the diagnosis of ASD, the findings of research that report on the language abilities of children with ASD often consider this factor.

In children who do develop speech, researchers have explored the age that children produce their first word (Howlin, 2003). It was reported that children with ASD achieve this language milestone later on in their development, where their first word generally occurs at an average of 36 months (TD \approx 12-18 months). This has very important implications as research suggests that the age that “useful speech” is acquired is a good indicator of independence in later life. Further to this is the concept of a language regression. Many parents of children with ASD report that their children began with typical language development e.g. babbling, and first word within typical time frames, but then regress to an earlier stage of development. This is specific to infants with ASD, as infants with other DDs do not generally display this language regression (Lord et al., 2004). There is ample evidence of language delays being a very dominant feature of ASD, although not all children who have ASD will have language impairments (in line with the newer DSM-5 criteria reported above). The children who do have these difficulties display very varied levels of language abilities from minor language delays to the complete absence of language.

Research in the field of ASD and bilingualism has vastly increased in the literature in the past two decades (Hambly & Fombonne, 2009; Ohashi et al., 2012) however, there is still an unclear picture regarding the linguistic implications for language acquisition for children who have a diagnosis of ASD and are exposed to two (or more) languages. Of the research that does exist, there is no evidence to support the suggestion that bilingualism confuses or interferes with language development (Valicenti-McDermott et al., 2013; Drysdale et al. 2015). These studies will be discussed in further depth throughout this section. As previously mentioned, ASD is a very varied disorder and the language impairments that are displayed in those with ASD are often very different from children with other DDs. As Dyches et al. (2004) point out, it is not always appropriate to compare studies that look at bilingualism with other disabilities, however, identifying how bilingualism impacts the linguistic development of children with ASD will be informative in understanding the impact of bilingualism for children with DS.

One of the first recorded studies that investigated bilingualism and ASD was a single case study design that was conducted by Seung and colleagues (2006). In this research, a bilingual language intervention was initiated by the introduction of English into a three-year old’s Korean speech therapy sessions. The child was gradually exposed to more English, even though Korean was his primary language. After 24 months, significant gains were observed in

his receptive and expressive language abilities both in English and Korean. The authors concluded that SLTs should work in a culturally appropriate way with professionals and bilingual families in order to observe the optimal outcomes.

Some studies have attempted to establish if bilingualism in preschool children results in any increased developmental delays. One of these was a large scale study conducted by Peña et al. (2011), with a total of 1029 infants. The authors reported that there were no increased risks of language impairments, for those in bilingual environments compared to the average occurrence of developmental delays for infants in a monolingual environment. Language milestones have also been used in retrospective studies to identify if these are achieved at the same time for bilingual children with ASD. Hambly and Fombonne (2009) recruited infants aged between 18 months and 6-years-old in order to compare the age of significant language milestones between monolinguals and bilinguals with ASD. The age of first words and phrases were not significantly different between groups, however, this study did not differentiate between children with simultaneous versus sequential bilingual exposure.

In a similar design to Hambly and Fombonne's research that investigated language milestones in those with ASD, Ohashi et al. (2012) also found no significant differences between bilinguals and monolinguals on measures of language milestones, nor for receptive and expressive skills, communication impairments or functional communication abilities. Importantly, this research also controlled for individual differences. Additional literature in this field has used group designs in order to distinguish if there are group differences between monolinguals and bilinguals. For example, Petersen, Marinova-Todd and Mirenda (2012) compared 14 bilinguals and 14 monolinguals with ASD on measures of conceptual vocabulary sizes. After controlling for IQ differences, there were no significant between-group differences for vocabulary. The bilinguals did, however, have a higher overall conceptual vocabulary size. The authors concluded that those with ASD have the capacity to acquire two languages successfully.

These findings have also been replicated by Hambly and Fombonne (2012) who compared the vocabulary sizes of children between the ages of three and six-years-old who had ASD. Forty-five were bilingual, and 30 were monolingual. This study also compared sequential versus simultaneous bilingual exposure, using parent report measures along with vocabulary scores. Findings showed no significant differences between the three groups on measures of expressive and receptive vocabulary sizes. Interestingly, there was no significant difference between the vocabulary sizes of simultaneous and sequential bilinguals. There were, however, significantly higher scores for the simultaneous bilinguals on the Vineland Adaptive Behavior Scale (measuring social skills; Sparrow, Cicchetti & Balla, 2005), suggesting that simultaneous bilinguals with ASD may even benefit from bilingualism in terms of daily functioning and social skills. The authors concluded there were no additional delays as a result of bilingual exposure in infants with ASD.

Further to the reports that there are no additional delays for children with ASD who are exposed to a second language, Valicenti-McDermott et al. (2013) also looked at vocabulary

sizes in monolingual versus bilingual children with ASD and found concurring results. The amount of babbling, word combinations and vocalizations were also equal between the groups suggesting that bilingualism does not impact any of these early language developments for young children with ASD. One significant finding of this research was that the bilingual infants 'cooed' more than the monolingual group, which may imply that there is something different happening in the early bilingual development of these infants. Again, the crucial implication of this research is that bilingualism is not a hindrance for those with ASD. This study did, however, use mostly parent-report measures, which may not be as reliable as empirical studies and authors stress that the results require replication. Additionally, this was a study that was conducted over 7 years and focused on infants below three years of age only.

One final study that also supports bilingualism within children with ASD, focused on an older child who had a diagnosis of HFA. In this research, the researchers recruited a family who was bilingual and had chosen not to follow the advice given to them by practitioners to only use English (Ochs & Solomon, 2010). Although no comparisons were made between other children, the authors suggest that those with ASD, specifically HFA could successfully become bilingual. Furthermore, it was suggested that bilingual exposure was better than artificially restricting the family's input to English only, particularly as English was not the family's primary language. They conclude with the statement that "given that the family is the primary institution for nurturing social and emotional bonds, the importance of the mother tongue and default language of the home for promoting autistic sociality cannot be overstated," (P.4). Wharton et al. (2000) also highlights this and report that parents are more engaging and affective in their primary language, and therefore better outcomes will exist for children with ASD if families do not restrict language input.

Further aspects of linguistic development have been assessed in bilingual children with ASD. For example, Gonzalez-Barrero and Nadig (2019), investigate morphological abilities and vocabulary sizes via standardized language assessments in bilingual children with ASD (n = 13) alongside NVMA matched monolinguals with ASD (n = 13). Results showed no significant differences in the children's dominant language for morphological abilities or receptive vocabulary after considering chronological age and parental socioeconomic status. In addition, it was reported that both groups of children performed within the expected range after considering their developmental ages. The bilinguals in this study were considered 'proficient bilinguals', and the authors conclude that bilingualism does not negatively impact morphological and vocabulary development in these school-age children.

Finally, Meir and Novogrodsky (2019) investigated morphosyntactic skills, theory of mind and working memory in monolinguals and bilinguals with HFA alongside TD monolinguals and bilinguals. This study included 85 children between the ages of four and nine-years-old and demonstrated that morphosyntactic abilities were required for the development of subject and object pronoun use in children with HFA and that importantly, these morphosyntactic abilities were similar for both monolinguals and bilinguals with HFA. This suggests that bilingualism does not negatively impact these important aspects of

development for children with ASD either, and extends this finding to novel aspects of linguistic development. This study does, however, highlight that both monolinguals and bilinguals with HFA have marked difficulties with morphosyntax, and specific difficulties with third-person subject and object pronoun use.

Although some practitioners are reported to still believe that a bilingual household is still too much of a challenge for children with ASD (Kay-Raining Bird et al., 2012), research contradicts these claims and even suggests that the removal of a native language could be more detrimental (Altan & Hoff, 2018). This will be discussed further in section 2.3, however, one comprehensive review of the literature in this field was conducted by Drysdale et al. (2015) in which 8 studies were included with a total of 182 participants. This systematic review concluded that there are no additional language impairments or delays found for children placed in a bilingual environment for children with ASD. There are however limitations to consider, such as the retrospective methods often employed (parent reports), the limited sample sizes and the fact that these studies only provide a snapshot of development. There is also currently a need for longitudinal studies in this field.

Finally, a recent review by Lund, Kohlmeier and Durán (2017) specifically explored bilingualism in children with ASD. They reported that there was no evidence to suggest that the language abilities of bilingual children were different from that of monolingual children in the seven articles reviewed. The research that exists to date frequently reports that those with ASD can, and do become bilingual. This does not, however, mean that bilingualism will be suitable for every child with ASD, as highlighted by Genesee (2006: p51), where he proposes that it is essential to consider the individual circumstances in each case and decide if bilingual exposure is appropriate. This research will be useful in guiding bilingualism research in DS, and it may also be possible to compare profiles of language development. In addition, there is somewhat of an overlap between DS and ASD with an estimated 5-15% of those with DS also receiving a diagnosis of ASD (DiGuseppi et al., 2010). Therefore, this is a further factor to consider when assessing bilingualism in children with both DS and ASD, which will be addressed in Chapter 7.

2.2.2 Other Developmental Disabilities and Bilingualism

Although researchers in the field of bilingualism have primarily researched bilingual language development in children with ASD, recent research has also emerged which extends this field to children with other DDs. For example, Paradis (2007) assessed the language abilities of English-French bilingual children with a Developmental Language Disorder (DLD; previously Specific Language Impairment, SLI), which is a language disorder which presents significant long-term impairments in speech and language that has no obvious cause (e.g. no hearing impairment or disability). In this study, a range of assessments were administered in order to assess the use of grammatical morphemes in bilinguals when compared to English and French monolinguals with DLD. Given that inappropriate morpheme use forms part of the clinical diagnosis for DLD, the researchers were interested in identifying if the bilinguals would show

further difficulties with grammatical morpheme use. No differences were found between the bilinguals with DLD and the monolinguals in either language. The authors concluded that this provides evidence that the bilinguals in this study did not exhibit any additional difficulties within this key domain for individuals with DLD, and that bilingualism was not an aggravating factor on these grammatical impairments.

Researchers with similar aims have explored further elements of language in bilinguals with DLD, including object clitic pronouns (Paradis et al., 2005) and tense morphemes (Paradis & Crago 2000; Paradis et al. 2003). These aspects also pose challenges for individuals with DLD and are frequently used as a clinical marker for this disorder. These studies have varied in using matched monolinguals with DLD and younger TD children. Findings of these studies revealed that children with DLD displayed marked difficulties in these areas compared to the TD controls, however, the bilinguals performed comparably to the monolinguals with DLD. These studies suggest that although these children were displaying the expected linguistic difficulties, bilinguals did not show any evidence of any further impairments. In these studies that were conducted in Canada, only French-English bilinguals were considered. Consequently, it may be the case that learning two languages which have a high majority language status assisted in developing equivalent skills in their two languages.

This view is, however, challenged by more recent research which suggests an opposing stance. Orgassa and Weerman (2008) investigated the accuracy of gender-marking in the L2 of sequential Turkish-Dutch bilinguals with SLI in comparison to a number of control groups. These groups included monolinguals with SLI, TD monolinguals and bilinguals, and also younger TD groups. Although similar error patterns were found for children with SLI, the bilinguals with SLI were significantly less accurate compared to both the monolinguals with SLI and the TD bilingual group. The authors suggest that this provides evidence of a cumulative effect (i.e. the combination of bilingualism and SLI leading to processing difficulties in Dutch gender). Although it is important to note that the children were being assessed in their second, weaker language, the bilingual SLI group also performed lower than the TD group who were also being assessed in their L2.

Furthermore, one recent study investigated the lexical, grammatical and narrative skills of simultaneous Italian-German bilingual children with DLD in comparison to TD bilingual children (Marini et al., 2019). After controlling for a number of variables including the context of acquisition, age of exposure and degree of exposure to each language, the authors reported that the children with DLD had reduced lexical skills and lower phonological short-term memory. Interestingly, these impairments were evident in both of the children's languages. It was also highlighted that the impairments in phonological memory were related to the children's abilities in naming items and grammatical abilities, suggesting that the role of phonological short-term memory may underlie these impairments in children with DLDs who are either bilingual or monolingual.

Less is known about the capacity of individuals with more rare disabilities to become bilingual due to the small numbers of individuals who have these rarer disorders that also

lead to impairments with language. In such conditions, case studies have provided early insights into the language abilities of bilingual children with Williams syndrome (WS; Perovic & Locket, 2015) and Prader-Willi syndrome (García-Alcaraz, 2018), which are both rare genetic conditions that result in mild to severe intellectual difficulties. Many children with WS appear to have relatively preserved speech (given their cognitive functioning) and often have intact language skills. In contrast, children with Prader-Willi syndrome often have substantial language delays and impairments. Case studies that investigate bilingualism in these populations report that the individuals under study were developing their languages at the expected pace considering their genetic disorders (Perovic & Locket, 2015; García-Alcaraz, 2018). Although no comparisons were made in Perovic and Locket's early report of a 5-year old with WS, García-Alcaraz specifically explored the narrative abilities of a bilingual with Prader-Willi syndrome in comparison to younger TD bilingual adults. The findings suggested that bilingualism did not exacerbate the difficulties observed within each disability on linguistic measures and also on cognitive development (Perovic & Locket, 2015). Although case studies are not generalizable, they are capable of providing valuable insights into the language profiles of rare developmental disabilities.

In a review of the literature, Kay-Raining Bird et al. (2016) explored the language profiles of bilingual children with a range of DDs including ASD, DLD and DS. In this analysis, it was found that children who were simultaneous bilinguals were always reported to have similar language abilities to matched monolinguals for the same DD when either the dominant language or both languages were considered. In contrast, those who were sequential bilinguals may have lower abilities than monolinguals with the same condition, particularly if they have only received limited exposure to the second language, although this was reported primarily for SLI (as reported above). On the whole, the review suggests that the addition of a second language does not lead to any additional difficulties for these children, and consequently, they should be included within a range of educational programs and bilingual provisions where appropriate.

Finally, a recent meta-analysis investigated research that directly assessed bilingual language development in children with a range of language impairments (ASD $n = 10$, intellectual disabilities $n = 2$ and communication disorders $n = 39$; Uljarević et al. 2016). Systematic methods were undertaken to include a range of empirical studies that investigated this topic. Of the studies which met the inclusion criteria, the majority reported that the bilingual groups were not at any disadvantage in terms of their language skills. Only a handful reported negative impacts of multiple language exposure, however, these studies did not employ a monolingual control group of children with the same language impairment. Consequently, these studies which reported small negative effects of bilingualism may not be as methodologically valid as those who did include control groups. In contrast, one recent study which was included in this meta-analysis reported a positive effect on social and communicative abilities for a sample of children with ASD (Hambly & Fombonne., 2012). This meta-analysis consequently concurs with the vast majority of the research reported above

which reports that bilingualism is not detrimental to those with developmental disabilities. The authors do note that caution is warranted as the available research in this field is small, particularly for research which is of a methodologically high standard.

2.3 Professional Recommendations Regarding bilingualism

Recent research has provided an insight into the topic of bilingualism within individuals with DDs and has focused on the specific difficulties that individuals within these populations may face after receiving a diagnosis of a particular disorder. Parents may subsequently be in a position of having to make important decisions regarding educational provisions and language exposure. One of the most comprehensive reviews that has investigated policies and practices related to bilingualism in DDs was undertaken by Pesco et al. (2016). Multiple locations and organizations were included in this review, which were Canada, the US, the UK and the Netherlands. The primary finding of this report was that the majority of the locations had policies that explicitly referenced the inclusion of students with a DD, however, in practice, there was a fair amount of variability in the opportunities that existed in terms of inclusion in bilingual services. An example of this was that some locations did not have special education state schools for children with DDs, while others had a high number of students attending these types of schools, especially in the Netherlands.

In regards to the language of instruction (LoI), some countries only had one majority language (e.g. Manchester- English) and as a result, there was no option other than to become a functional bilingual in this language if a child's home language was a minority language. Some other locations had 'welcome classes', and an intensive support system for those where the LoI was not English. Others had other support systems such as immersion programmes. In terms of support for minority languages, some countries offered community initiatives to support these languages, however, these often seemed to be very limited. Overall there appears to be very mixed support for minority languages. All countries supported L2 learning during primary and secondary education, suggesting that policies reflected the idea that an L2 can be learnt without detrimental effects to the L1, although these varied from immersion programmes to optional schooling programmes of modern languages.

The inclusive schooling policies suggested that bilingual opportunities are also available to children with a DD. Some of the special education policies explicitly stated that children with a DD had access to bilingual opportunities. For example, immersion or bilingual programmes in Canada mentioned that 88% of the students with a DD were included in the overall education process. One policy in British Columbia supported children in learning a second language, however, had an 'opt-out' option from second language classes for children who had a DD if parents felt that their children might be "unable to demonstrate learning in relation to the expected learning outcomes of the second language course" (p.26). In other countries, there were no language-in-education policies that directly addressed second language learning in children with a DD. The authors concluded that there is some variability in the amount of support that is given. On the whole, the policies reflect the proposal that those with a DD should be included in second language learning settings, although it is not

clear the extent to which these are adhered to. The language(s) of the community and the LoI will ultimately also have a strong indication of the amount of support that is available. No policy addressed the issue of the severity of the disorder, and there was no mention of tailoring education to individual needs in this respect.

In terms of ASD specifically, it has been reported that clinicians' recommendations are not in line with bilingualism research when advising parents regarding language exposure (Kay-Raining Bird et al. 2012; Kremer-Sadlik, 2005). Clinicians are one of the first points of contact for parents who are concerned that their child may be on the autism spectrum or is having language and communication difficulties. Therefore, the recommendations that they provide is likely to be very influential in parent's actions in regards to language exposure for their child. In these studies, some parents who had a child with ASD reported that they felt that two languages would be too confusing for their child, and may delay language development in both the first and second language as a result of being advised of this by professionals (Kay-Raining Bird et al., 2012; Yu, 2013). There has been an increasing focus on the importance of early detection and interventions for ASD (Eaves & Ho, 2004), and so it is crucial that practitioners are up to date with recent research in order to appropriately advise families and employ evidence-informed recommendations.

In order to identify if parents have been advised appropriately, one study explored ASD and bilingualism in an ethnographic approach (Kremer-Sadlik, 2005). In this research, it was reported that parents of children with ASD who were not native English speakers were advised to speak *only* English to their child, regardless of their own English language proficiency. Clinicians suggested that multiple languages would confuse children with ASD and that simplified language input would facilitate language learning. Restricted language input was reported to be difficult for the families, and Kremer-Sadlik concluded that in order to become members of the community, the parents felt that their children need to be given the opportunity to learn their mother tongue alongside English. The implications of restricting language use were also highlighted. For example, parents may not be as engaging in a non-native language with their children and may not be adequate language models in languages in which they are not as fluent as their native language. This would certainly have significant implications on children with ASD who often have language delays, to begin with. The advice given to parents does not appear to take these implications into account when these recommendations are provided to families. This is also supported by De Houwer (1999) who argues that bilingualism is not a choice, but a necessity in order to participate fully and communicate with families, and this remains the same for children who have a DD. This goes alongside Wharton et al.'s (2000) findings that parents who communicate in their native language are more engaging and affective, which in turn promotes better relationships and better language outcomes for their children.

One conflicting finding has been reported by Yu (2013), where Chinese-English mothers of children with ASD expressed that the academic outcomes and language success were more important than language that their child acquired. The findings of the research

displayed that parents believed that multiple language input would confuse their child, and this was a view that was reinforced by the practitioners that the mothers had contact with. In this research, the parents reported that they believed that English was more important to meet the demands of society, especially as education services tend to be solely through English in this situation. In this study, the parents concurred with the advice of practitioners (i.e. that a monolingual input would result in better outcomes.) As reported elsewhere, only a small number of parents (20%) were supported by practitioners to raise their child bilingually. The majority received advice against bilingual exposure. This research utilized an interview design that provided in-depth and elaborate responses, however, these only provided a snap-shot of information from the time of the interview, and the parent's views may have altered over time. The authors also raise caution as their sample was non-representative as all the mothers were highly affluent and highly educated.

Recent research by Marinova-Todd and colleagues (2016), specifically investigated professional practices and opinions regarding services available to bilingual children with a DD. The aim was to obtain an insight into the opinions of school and education-based professionals and the reported practices that they had undertaken for children with a DD regarding bilingualism. Marinova-Todd et al. aimed to investigate whether research findings in the field had informed practice in a number of countries. The survey was designed in collaboration with the six sites involved and comprised of information pertaining to demographic data, statements regarding sequential and simultaneous bilinguals, optional second language learners and bilingual services. Participants were asked to consider the questions in relation to different types of children and the study was adapted to the cultural requirements depending on the location. Participants were mainly female and were primarily teachers and speech-language therapists. The scope of the research did not target parents of children with a DD, and instead targeted professionals who had direct contact with these children.

Results were combined to form three groups; TD children, children with mild disabilities and children with severe disabilities. Overall, children who were TD were perceived differently to children with a DD. There was generally a strong agreement that children who were TD and those with mild disabilities could acquire two languages, but that for a child with severe difficulties that their capacity to learn a second language was more limited. In terms of the availability of services, there were mixed findings depending on where the respondents were located, however, on the whole, the more severe the language impairment, the less of an opportunity there seemed to be to access bilingual services. Reports of what participants felt *should* be available differed, as most believed that language services should be more accessible. These results suggest a good awareness of recent research in this field, however, there is still some discrepancy between the research and what practitioners are recommending to families, and particularly the support that is available to accommodate these requirements. Generally, there seems to be a lack of resources available to assist with minority language development in children with a DD and so future bilingual

assessment and intervention practices are needed. Generally, practitioners seem to be more supportive of bilingualism for children with a DD than previously reported.

One final study that investigated the advice received by parents of children with ASD was conducted recently by Hampton et al. (2017). In this research, qualitative interviews were utilized with parents of children with ASD, in comparison to parents of children who were TD. Some general concerns relating to bilingualism, in general, were mentioned by both groups, as well as some shared societal and cultural factors that were perceived as having importance on their decisions to employ a bilingual language experience for their children. There were, however, some additional factors that only concerned the parents of a child with ASD. For example, some perceived that there would be an interaction between specific difficulties with ASD and bilingualism, such as impacting not only language development but also cognitive and behavioural development. The authors also emphasize the importance of parent and child bonds which may form to a greater degree by exposure to a family's native language.

In summary, there appear to be mixed messages received by parents regarding bilingual exposure for a child with a DD. In the literature to date, there seems to be a tendency to focus on this issue within children who have a diagnosis of ASD, however, there also seem to be concerns regarding bilingualism within parents of children with other DDs.

2.3.1 Disability and Bilingualism in Education

Closely related to professional recommendations regarding bilingualism is the recommendations and support provided from educational provisions. Parents have considerable contact and support from a child's education setting in addition to the support received by various other professionals. In the UK, children with a Special Educational Need (SEN) may attend a mainstream (MS) school, or a SEN base within a MS school, or may attend a SEN school if there is a suitable placement that is able to cater to the needs of that child. In Wales, out of all the schools in the country, 22.1% of children have some form of SEN. Table 12 (Welsh Government: School Census Result, 2012) below displays the distribution of children who attend the various school options between the years 2008-2012. As the Welsh Government data shows, there has been a small increase in the number of children in SEN schools overall, particularly for those over the age of 16.

In terms of the number of children who have a statement of SEN, the total number of children has remained stable at around 2.7% up until 2016. Table 13 (Welsh Government: School Census Result, 2016) displays the distribution of all children who have a SEN statement between 2012-2016. A recent report by de Valenzuela et al. (2016) investigated the opportunities and service provisions that are available to bilingual children with a DD across four countries (England, Canada, the USA and the Netherlands). This international project analysed 79 semi-structured interviews from a range of professionals including teachers, speech and language therapists and policymakers. Perceptions relating to a wide range of intellectual and DD were explored in order to provide an overview of the opportunities to access bilingual services. As bilingualism does not appear to have any detrimental effects on

language development for children with a DD, this study aimed to identify if the policies and recommendations were being implemented.

		2008	2009	2010	2011	2012
Nursery						
	Pupils aged: Under 5	1,787	1,791	1,672	1,572	1,530
	5 and over	0	0	0	0	0
	All ages	1,787	1,791	1,672	1,572	1,530
Primary						
	Pupils aged: Under 5	62,915	63,985	65,421	67,721	69,766
	5 to 10	198,510	194,189	191,907	191,359	192,293
	11 to 15	170	129	107	101	76
	16 and over	12	11	10	8	9
	All ages	261,607	258,314	257,445	259,189	262,144
Secondary						
	Pupils aged: Under 5	0	0	0	0	0
	5 to 10	29	15	6	5	19
	11 to 15	178,597	176,299	173,914	171,142	168,204
	16 and over	28,310	29,107	29,987	30,083	29,792
	All ages	206,936	205,421	203,907	201,230	198,015
Special						
	Pupils aged: Under 5	201	201	198	217	213
	5 to 10	1,060	1,018	1,032	1,008	1,062
	11 to 15	2,108	2,164	2,116	2,117	2,111
	16 and over	671	732	771	839	868
	All ages	4,040	4,115	4,117	4,181	4,254
Nursery, Primary, Secondary and Special						
	Pupils aged: Under 5	64,903	65,977	67,291	69,510	71,509
	5 to 10	199,599	195,222	192,945	192,372	193,374
	11 to 15	180,875	178,592	176,137	173,360	170,391
	16 and over	28,993	29,850	30,768	30,930	30,669
	All ages	474,370	469,641	467,141	466,172	465,943

(a) At January each year.

(b) Age as at 31 August in the previous year.

Table 12. Number of pupils with a statement of special educational needs by major need from 2012-2016 in Wales (Welsh Government: School Census Result, 2016).

		2011/12	2012/13	2013/14	2014/15	2015/16
Total		103038	103791	105303	104957	105143
Total	Moderate learning difficulties	34097	29249	24808	20631	17983
	General learning difficulties	16868	21034	24890	27963	29629
	Severe learning difficulties	2456	2463	2339	2166	2080
	Profound & multiple learning difficulties	729	775	775	785	766
	Dyslexia	9304	9161	8978	8590	7733
	Dyscalculia	420	482	623	561	435
	Dyspraxia	497	537	574	566	556
	Attention Deficit Hyperactivity Disorder	801	986	1167	1243	1368
	Autistic Spectrum Disorders	3968	4227	4592	4915	5327
	Physical and medical difficulties	4267	4332	4437	4549	4692
	Hearing impairment	1932	2081	2149	2182	2215
	Visual impairment	823	845	862	867	861
	Multi-sensory impairment	64	66	57	48	49
	Speech, language and communication difficulties	13244	13940	14680	15336	16259
	Behavioural, emotional and social difficulties	13568	13613	14372	14555	15190
	Specific learning difficulties
	Physical disabilities
	Medical difficulties
	Emotional and behavioural difficulties
	Other
	Not stated

Table 13. Number of pupils with a statement of special educational needs by major need from 2012-2016 in Wales (Welsh Government: School Census Result, 2016).

Thematic analysis was used as the coding method for all of the locations and all researchers responsible for coding the data used the same coding scheme. This was also verified for reliability between the researchers. The overarching theme of the analysis was the inclusion/exclusion of children with a DD to bilingual language programmes and the emerging themes were *'We include all kids, special needs drives it, time scheduling conflicts, IEP/Statement drives it, it's up to the parents and service availability.'* All sites reported that students who were in inclusive settings also had access to special needs services, however, some recognised that inclusion in all aspects is ultimately dependant on each individual

student. In addition to this, it was clear that the diagnosis of each individual and the severity of their disorder was an influential consideration as to the access to language programmes that are part of general education practices. Some stated that the DD was more important than bilingualism and as a result, language opportunities seemed to receive less attention than other services. Respondents also made it clear that they felt that learning two languages would be more difficult than one, and would put extra pressure on them. As a result, bilingualism was not regarded as a priority for children with DDs.

Further respondents stated that practicalities did not make it possible for a child with a DD to be fully involved with a language programme, for instance, time and scheduling issues. There are often many additional support programmes that children with a DD have access to and it seems that these other programmes may take priority over language programmes. The authors suggest the possibility of combining these types of programmes to overcome these challenges. Related to this is the issue of service availability. It seems to be the case that the language programmes simply do not exist for all target languages, as well as a lack of funding within SEN or having the staff available to meet the needs of each child given the wide range of languages that the children may speak. Additionally, it is reported that the Individualized Education Plan (IEP) or Statement drives the opportunity to access language services. An IEP or a Statement is a legal requirement for each child with a SEN, in which a programme is designed to provide strategies and information relating to the curriculum that will be followed for the child with a disability or learning difficulty. In the report by de Valenzuela et al. (2016), there appeared to be some variability in what was included in each child's planning report, with some being more precise about the issue of language programme inclusion and others not. Some made it clear that they felt that the language programme should be included in the IEP/Statement as best practice.

Finally, a seemingly large factor regarding the access to these services was that much of the decision making was down to the parents. Some noted that if a parent specified that they wanted their child to have access to language programmes, that the parents themselves should be language advocates so that they had more access to other languages, in turn enabling them to become bilingual. An emerging issue here is that often parents are explicitly asked to make these decisions, automatically raising the suggestion that they should not be expected to participate. To conclude, there seems to be a discrepancy between the policies that exist on this topic and the practice that takes place. It seems that having a child with a DD results in policy adaptations or, in some cases, policies not being adhered to at all.

One final resource available to parents of a child with DS in the UK is the Down Syndrome Association's Educational Resources (2014). These resources are directed at primary and secondary educational institutions and focus on various areas of development. The first unit gives educational professionals an overview of the general profile of children with DS in terms of speech, hearing, memory, motor, attention and vision. This is a practical and educational based resource that offers practical advice to overcome some areas of weakness. This unit also highlights what inclusion is and how this should be approached. The

resource states that MS education should always be the preferred method of schooling and that the child should always progress through the school with their peers by tailoring the resources wherever possible. It is noted that SEN schools should only be an option if other avenues have been explored first, including transfer to a different MS school. This is because a MS environment allows for more peer-to-peer interaction and appropriate models for children with DS. The final unit of relevance is the developing language skills resource. In this document, the general patterns of strengths and weaknesses of language are identified, for example, the disparity between receptive and expressive vocabulary skills. The resource also highlights that language is an essential component to a child's overall development in terms of social, emotional and cognitive development, and as a result, improving language capabilities will subsequently assist in other areas of development. This resource does not, however, provide any support or recommendations regarding bilingualism.

Chapter 3: Down Syndrome

3.1 Introduction

For this project, the focus specifically concerns the language development of bilinguals with Down syndrome (DS). The following section will provide an overview of DS and clarify the progression of development for these individuals. Following this, research that exists to date concerning language development, including grammatical skills, phonological development and phonological awareness will be explored. This chapter will then present the research that has been conducted to date concerning the language development of bilingual children with DS, before presenting the aims of the present research.

Down syndrome is an intellectual disability and is the most common chromosomal disability with an estimated prevalence rate of around 1 in 700-1000 live births (Morris & Alberman, 2009; Parker et al., 2010). In the majority of cases, DS occurs when there is an extra copy of the 21st chromosome (trisomy 21), and the genetic make-up that results from the addition of this chromosome expresses itself in the physical, cognitive and developmental characteristics of DS. There are two additional forms of DS, one of which is termed mosaic DS which results in similar developmental trajectory as trisomy 21, however, those with this subtype usually display less severe impairments. This type of DS is less prevalent and only occurs in around 3.85% of DS cases (Devlin & Morrison, 2004). This subtype is distinct from the usual DS as not all cells contain the extra chromosome, only some of the genetic material has the additional copy, hence the term 'mosaic'. This results in a slightly different profile to that of trisomy 21, whereby children with mosaic DS show higher intellectual development (Fishler, Koch & Donnell, 1976; Papavassiliou et al., 2015). There is a third less frequent subtype of DS termed translocation which only occurs in around 1.45% of cases. This occurs when one of the chromosomes attaches itself to a different chromosome (Devlin & Morrison, 2004). In terms of the prevalence of DS, there is a debate in the literature concerning the occurrence of the disability as there has been a substantial increase in the accuracy of available screening programmes. This has resulted in an earlier and higher detection rate which it is argued leads to higher rates of terminations (Maxwell et al., 2015). In contrast, the age at which individuals have children has also been increasing. As there is also a higher rate of DS as the age of the mother increases (Munne et al., 1995), the number of children being born with DS may be changing.

Those with DS are reported to follow a distinct profile of development whereby there is a pattern of strengths and weaknesses seen. It is however reported that there is a fairly large range in abilities with a considerable discrepancy in the phenotype across each individual with DS. There has been considerable research that explores the early developmental trajectory of children with DS, and these have focused on the similarities and

differences that are seen between TD children and those with DS. It has also been reported that the linguistic abilities of those with DS are disproportionately impaired in comparison to other aspects of development and cognitive abilities (Chapman et al., 1998). There are, however, important variables that are specific to those with DS that should be considered in assessing language development. Specifically, evaluations of cognition and language may be complicated by unidentified hearing and sleep disorders, which are often a feature in young children and adults with DS (Roizen & Patterson, 2003).

Recently, there has also been an increase in understanding of the association between DS and ASD as a dual diagnosis. As knowledge and understanding of ASD have been increasing, the identification of children and adults with the disorder has also increased (Blumberg et al., 2013; Matson & Kozlowski, 2011). Delayed language onset is a key marker of ASD (De Giacomo & Fombonne, 1998), but is also prominent in those with DS. Typically, children with DS have strengths in their social abilities, however, this is not the case for individuals with ASD. The number of children that have been identified as having a dual-diagnosis of DS and ASD has increased, with estimated prevalence rates between 5-7% and 15% of children with DS (DiGuseppi et al., 2010). Therefore, there is a higher rate of ASD found in children with DS than in the general population, with prevalence in the general population being around 2.24% (Zablotsky et al., 2015). These children may consequently have a unique profile of development and this will be the focus of Chapter 7.

3.1.1 Physical and Cognitive Development

One of the most prominent characteristics of DS is a moderate to severe intellectual disability, however, other atypical developments include physical characteristics and health implications, which again vary from individual to individual. In addition to these, a vast majority of individuals with DS also experience some form of hearing difficulties, with estimations suggesting that 80% of those with DS also have a hearing disorder (Sacks & Wood, 2011; Balkany et al., 1979). Hearing disorders in DS can be conductive, sensorineural or both, however, one significant co-occurrence in those with DS is otitis media with effusion (more commonly referred to as glue ear; Roberts & Medley, 1995). In addition to hearing disorders, ear infections are also reported relatively frequently, and these will both have implications on language development, due to the possibility of limited auditory input. Identification and treatment of these are generally an important aspect of medical management in DS.

Hearing loss and ear infections may have an impact on the perception of speech in these individuals, however, estimates report that these only accounts for 4-7% of the impairments found in the receptive language abilities in individuals with DS (Chapman et al., 1998). Therefore, it is suggested that other factors impact the deficits found in expressive and receptive language ability other than simply due to hearing impairments. More recent studies have found evidence that suggests that the impact of hearing loss on language is more instrumental to development than previous reports. For example, Laws and Hall (2014)

investigated the impact of hearing loss on later language development and found that the children with early hearing loss performed significantly worse on the language measures than the children with DS who did not suffer from hearing loss, after accounting for non-verbal mental age (NVMA) and chronological age (CA). Therefore, the clear identification of any hearing loss should not be underestimated in the evaluation of language development.

One further consideration in dealing with expressive language development in DS is that many individuals have a low muscle tone in various muscles that are fundamental to the production of speech. The muscles in the tongue, lips and cheeks in individuals with DS are commonly affected which results in less firm precision of the production of speech sounds (Kumin, 1996). Barnes et al. (2005) also reported that there are difficulties associated with oral-motor skills which result from structural abnormalities that occur in some cases of DS which may also impact the development of language. In addition, there are structural differences in the brain development of individuals with DS (e.g. underdeveloped lobes, and smaller overall brain size). This is argued to be a possible contributing factor to the functional differences in these individual's cognitive development (Pinter et al., 2001) which will be discussed in further depth in the following section. The combination of these physical factors provides some insight into the expressive and receptive language impairments that are found in individuals with DS.

In terms of the cognitive development of those with DS, there is believed to be a different trajectory compared to that of TD individuals. One area that this has been extended to is memory abilities in DS, which has been explored extensively in the literature to date, in terms of short and long-term memory capacity. Kay-Raining Bird & Chapman (1994) specifically investigated short-term memory recall for various stimuli in a range of auditory and visual memory tasks. Participants with DS were matched to TD children on mental age (MA), and it was reported that individuals with DS recalled fewer items than the TD control group. This finding was evident for the auditory memory tasks as well as visual memory tasks, and therefore, this research suggests that both visual and auditory memory recall is impaired in individuals with DS.

One further piece of research that investigated the memory abilities of individuals with DS was undertaken by Seung and Chapman (2000) in a digit span study. This research aimed to determine if there is a deficit in the phonological loop process (i.e. articulatory rehearsal or passive storage), which might help explain the deficits found in language abilities. Thirty-five individuals with DS were matched to TD controls on language production skills, and it was reported that although speech production rates were equal across the two groups, the DS group had a lower digit span score. This suggests that short-term memory impairments in those with DS are even greater than their language impairments, which is an important aspect to consider when researching the language abilities of individuals with DS. In contrast to the apparent memory deficits that are found, research suggests children with DS are found to perform better at visual-spatial tasks (Abbeduto et al., 2007; Jarrold et al., 2008). As a result,

memory abilities may be improved by providing visual cues in order to appropriately assess skills in other areas of development.

Further cognitive impairments have been found in the phonological memory abilities of individuals with DS. One type of task that is used to measure phonological memory is non-word repetition tasks (NWRT), and these are reported to be good predictors of language comprehension and reading abilities (Laws, 1998). Children with DS typically struggle with these types of tasks (Cains & Jarrold, 2005), suggesting that phonological memory impairments may result in difficulties with language comprehension and expression. In addition to this, Laws and Bishop (2004) also report that children with DS have weak phonological skills, which is specifically related to phonological memory. It is suggested that this may account for some impairments relating to grammatical difficulties. In this longitudinal study that was conducted, the age of the first word tended to occur later than typical development, however, the 'language spurt' and combination of word development seemed to lag behind even further for children with DS compared to TD children. It is also reported that there are considerable deficits in the productive domain of language within children with DS, specifically in terms of grammar and phonology. The authors suggest that the fundamental component which underlies these skills is short-term memory. It was concluded that this may consequently explain the performance of individuals with DS on early linguistic development, as memory abilities may not develop adequately for language acquisition to occur at the appropriate pace.

Furthermore, individuals with DS are also reported to have difficulties in maintaining attention on a particular task. This is a further factor to consider when accounting for the poor cognitive development of these individuals. This was reported by Brown et al., (2003) who examined the sustained attention abilities of infants with DS and it was reported that there were lower engagement levels shown by infants with DS. Time spent attending to appropriate stimuli was lower when compared to a control group, matched for MA. This factor should be considered when designing research paradigms that measure language in order to evaluate and understand the role that this may have on language development.

Finally, one further cognitive component of DS that has been well documented in the literature is the proposal of early onset of cognitive decline or early-onset dementia (Pack, 2014; Lautarescu et al., 2017). In typical adults, dementia is the result of cognitive decline due to old age, and this can compromise language and communication abilities (e.g. not being able to remember words for objects, etc.). This is also the case in adults with DS, however, this cognitive decline has been found to occur significantly earlier (Lautarescu et al., 2017). As a result, it has been suggested that individuals with DS have a high risk of developing early-onset dementia. Lautarescu and colleagues conducted a systematic review to specifically identify any longitudinal studies that explored this issue. They reported that there was a high prevalence rate of dementia in this population, but also that the symptoms were not always identical for each individual. Therefore, assessment methods should be employed that enable accurate early diagnoses for individuals with DS. Preliminary intervention studies have also

been conducted with adults with DS who have early-onset dementia, and these have been found to alleviate symptoms or prolong the period of cognitive deterioration for these individuals (Fonseca et al., 2015).

In conclusion, this section has highlighted a range of areas that are generally challenging for individuals with DS. It is useful and appropriate to provide an understanding of these as it is important to consider these elements in designing tasks that target these populations. This will be particularly relevant when it comes to memory abilities as these may be inter-related to language abilities. At the same time, tasks should employ designs that do not put too many constraints on memory capacities that may underestimate language abilities.

3.2 Down Syndrome and Language Development

The following section will explore the language development of individuals with DS and will consider linguistic development in several domains of language. These will include vocabulary, syntax, morphology, phonology and phonological awareness as these are all areas in which children and adults with DS tend to have specific difficulties with. In assessing language development in children with DS, studies often employ control groups of TD children in order to ascertain areas of specific weakness compared to typical development and to identify if the developmental trajectory is similar to TD children, or whether different patterns are observed. In order to compare children with DS to TD children, studies often employ matching techniques based on the developmental ages of the participants. In doing so, measures of mental age (MA) or nonverbal mental age (NVMA) are often employed. In these studies, this matching technique usually leads to the TD children being chronologically younger to children with DS but similar in terms of developmental ability. This is important to consider in the following section where studies of language development in children with DS are presented.

Firstly, one robust finding concerning the language development of individuals with DS is that the impairments found are often more substantial than anticipated given the cognitive abilities found. This means that even after considering the developmental ages of children with DS, even greater impairments are often observed for language. In addition to this, there is an ongoing debate in the literature concerning the linguistic development of individuals with DS, as some argue that the developmental trajectory is the same as TD children but simply substantially delayed. In contrast to this is the hypothesis that there is an atypical or 'deviant' developmental pathway, for example in terms of the speech errors that are made (Cleland et al., 2010), grammatical development (Hesketh & Chapman, 1998) and pronoun comprehension (Sanoudaki & Varlokosta, 2014). These atypical errors or weaknesses often occur alongside typical errors and delays seen in TD children.

A further debate that arises as a result of findings such as these is whether language is a modular or a singular processing system. Researchers have argued that because language impairments in children with DDs and particularly children with DS are not impaired to the

same extent across the various domains of language, this suggests that there are several 'modules' of language (Fodor, 1983; Perovic, 2006). This view proposes that language is not a single entity but that there are computational modules within language and there is a dissociation between the modules of language. This dissociation explains why some modules of language can be specifically impaired whereas other areas only show impairments and delays in line with cognitive functioning (e.g. receptive language). This proposal is important to consider in the current research as identifying whether a modular approach is supported in the present research will extend this debate to the modularity of language in bilingual children (i.e. are these modules language-specific or language independent).

Due to the nature of DS, there is a vast range of individual differences found in all areas of development and this extends to language development. Although there is a range in the severity of language impairments in DS, these generally seem to follow specific patterns and there are distinct characteristics within language development, as mentioned earlier. It is widely reported that expressive skills are considerably more delayed than receptive abilities (Chapman, 1997). When considering expressive language specifically, some aspects appear to be more difficult for individuals with DS than others, as mentioned earlier such as expressive morphosyntax, phonology and PA.

In comparing children with DS to children with Williams syndrome (WS; which is a genetic disability also caused by a chromosomal abnormality), individuals with WS have increased linguistic capabilities than would be expected given their MA. Researchers have explored the cognitive profiles of DS and WS and reported that both these groups of children tend to share some similarities in intellectual development, but that there is a distinct profile of characteristics for each syndrome (Klein & Mervis, 1999). During the early stages of language development, children with DS are found to lag behind children with WS in terms of their expressive vocabulary. Further research has found that the language profiles of both these disabilities follow a different developmental trajectory than TD children, as opposed to language delays (Perovic, 2006; Perovic, Modyanova & Wexler, 2013). Although the research suggests that there are impairments in the linguistic profile of both of these groups, the expression of impairments follow different patterns with children with DS having more difficulties relating to morphology, whereas those with WS tend to have the most difficulties with semantics. Perovic, Modyanova and Wexler (2013) suggest that there may be separate issues in cognitive processing that underlie the linguistic capabilities of each disorder. Furthermore, researchers have argued that there are distinct 'profiles' of development specific for each disorder, with overlapping strengths and weaknesses. Although there may be similarities in some of the cognitive processes that underlie language in these two disorders (e.g. impairments in the cognitive processing concerning working memory), it has been argued that there may be different processing mechanisms which explains the differences observed in cognitive and linguistic profiles (Bates, 2004).

Studies have found differences in the way that language is processed in the brains of individuals with DS and other DDs, including that of children with a developmental language

disorder (DLD). Although the brain structure of children with DS is physically different from children with DLD, imaging studies have found that the volume of certain areas are significantly smaller in individuals with DLD. For example, Brocas area (which is very well documented for its apparent fundamental role in the acquisition and production of language) was found to have a smaller overall volume in those with DS (Sahin et al., 2009). These studies suggest that there are similarities in the linguistic profiles of these various disorders, which may be due in part to impairments in working memory. It is also highlighted, however, that there is a distinct profile of language development apparent in DS that is not identical to these two other disabilities or that of typical development.

In contrast to the verbal communication weaknesses found, children with DS are reported to have relative strengths in terms of NV communication and often begin to develop gestural communication before producing language (Chan & Iacono, 2001). These strengths may enable a child with DS to express themselves non-verbally if their expressive skills are lacking, and Chan and Iacono (2001) highlighted the importance of recognising these communicative gestures to avoid missing essential opportunities for language development. Interestingly, Caselli et al. (1998) showed that children with DS had a significantly greater variety of gesture usage than TD children. Researchers have also explored early NV communication skills in order to identify if there are any differences in these patterns of communication between children with DS and TD children (Mundy et al., 1995). For example, Mundy et al. (1995) conducted a study to identify NV communication in infants with DS and TD infants after matching them on chronological and developmental age. Findings showed that gesture usage for infants with DS followed similar patterns of acquisition to that of TD infants in terms of the frequency of usage. One distinction was found, however, whereby the communicative functions of gestures were found to differ between groups with a possible requesting deficit in the group of infants with DS.

Finally, children with DS are reported to have strong abilities in terms of imitating and modelling based on social cues when compared to TD children, and children with DS are found to use more imitation in play (Wright et al., 2006). This should be considered when establishing language abilities and may help design research paradigms that focus on early language development. Any task should be tailored to the target sample's strengths to enable a clearer picture of abilities, without disregarding the communicative functions of gesture usage. Further research has focused on specific areas of linguistic development in DS and these will provide further insight into the developmental profiles of these individuals. These will be the focus of the following sections.

3.2.1 Expressive and Receptive Vocabulary

This section will explore the development of expressive and receptive vocabulary in children with DS, and will identify the disparities found between the development of these areas for children with DS and children who are TD. Expressive vocabulary refers to the production of words and is often measured in terms of lexical quantity and complexity. Receptive

vocabulary on the other hand is concerned with skills in comprehending or understanding words. These two skills will be examined in children and infants with DS during the early stages of language acquisition and the progression of these abilities over time.

The aims of early studies in the field of language in DS were to generate an overview of the vocabulary sizes of these individuals and some researchers have employed longitudinal designs to establish the foundations of early language development. One of these studies reported that the vocabulary size of young children with DS is substantially smaller than 3-year-old TD children when groups are matched on MA (Strominger et al., 1984). This is typically the time when TD children experience the vocabulary spurt, suggesting that children with DS do not experience this stage in development, or at least not at the same point in development as TD infants. In TD children, the vocabulary spurt is shortly followed by a development of grammar and vocabulary composition (Kauschke & Hofmeister, 2002). It could be proposed that if the vocabulary spurt stage is absent in children with DS, this may partly explain some of the structural deficits found in the language of children with DS if they also lack the grammar and vocabulary composition stage.

A further study which investigated the production of first word and two-word phrases in children with DS was conducted by Oliver and Buckley (1994). This was a parental report study that tracked the development of the infants under study by following them and their families to collect data at various stages in the children's development. This study reported that the vocabulary of infants with DS was delayed by around 18 months, however, the infants in the study followed the similar patterns of vocabulary development to the TD infants. In contrast, Strominger et al. (1984) reported that the majority of the children with DS did not exhibit the 'language explosion' which TD children did when they were around 2 years of age. This again suggests that the language development of children with DS during infancy may not be simply delayed, but is fundamentally different. It was also highlighted that there was a great deal of individual variability found. In a similar study that also utilised parental report measures, Miller et al. (1995) extended these findings to children at a later stage of development. Findings of this research also reported delays in expressive vocabulary skills of children with DS compared to TD children and found that these delays also extended to syntactic development. The delays found were again not consistent with the children's cognitive abilities with greater impairments found than would be expected.

Another influential early piece of research was conducted in 1985 by Cardoso-Martins et al. (1985). This study sought to compare vocabulary acquisition in children with DS compared with TD children. The authors found that TD 9-month-old infants had similar cognitive abilities and similar levels of production of object names as 17-19-month-old infants with DS. This is an outdated study and the participants were not selected as matched controls, however, it does provide a valuable insight into the expressive vocabulary skills of these children. The authors also report that the production of object names was slower to develop than would be expected as language development continued later, and continued to progress throughout childhood for the children with DS. At the same time, Galeote and colleagues

(2008) similarly report that the lexical skills of children with DS develop at a much slower pace to that of TD children.

More recently, various researchers have explored the relationship between expressive and receptive vocabulary (Oliver & Buckley 1994; Kumin 1996). These studies have often reported that expressive vocabulary abilities are more impaired than receptive vocabulary abilities (Roberts et al., 2007). Receptive vocabularies are reported to be commensurate with cognitive abilities, whereas expressive vocabulary seems to be more impaired with smaller expressive vocabulary sizes (Chapman & Hesketh, 2007; Martin et al., 2009). These additional difficulties with expressive vocabulary seem to continue throughout adolescence and adulthood and the gap between expressive and receptive skills seems to increase with the progression of expressive language being slower than language comprehension and not linear with non-verbal cognitive abilities.

In a large-scale parental report study which investigated the vocabulary sizes of individuals with DS, it was reported that the vast majority obtained spoken language with 90% of three-year-olds able to produce one or more words by this age (Berglund et al., 2001). As noted previously, individuals with DS often have memory impairments, which may result in difficulty with recalling language, particularly in complex or lengthy sentences. The addition of this factor, along with hearing difficulties makes language acquisition a long and difficult process for some children with DS. In summary, it is evident that there is a specific weakness in expressive vocabulary abilities in children with DS.

3.2.2 Syntactic Development

Syntax relates to principles of language that concern the structure of language, such as word order, and this area of language is found to be specifically impaired in individuals with DS. For example, research has reported deficits in receptive and expressive syntactic development for children with DS. This was reported in a study by Abbeduto et al., (2003) who found that children with DS performed significantly lower on syntax comprehension trials. This study compared the comprehension of syntax in children with DS in a MA matched design using TD controls (aged between three and six-years-old) and MA matched children with fragile X syndrome. As the children with DS performed significantly lower than both control groups, the authors concluded that syntax poses a significant challenge for individuals with DS.

In contrast, one study investigating the syntactic production abilities of individuals with DS was conducted by Thordardottir et al., (2002). In this research, adolescents with DS were recruited and these were matched with TD controls on MLU (TD children aged between two and four-years-old). The aim of this research was to investigate the usage of complex syntax in individuals with DS compared to the controls. There were no significant differences between groups in the variability of sentences used or the number of complex sentences produced and both groups used various syntactic forms, e.g. conjoined and subordinate clauses. The authors also reported that syntactic development continued in individuals with DS at a later stage of their lives, and in a similar trajectory to that of TD controls, similar to

that reported by Chapman et al. (1998). A further recent study aimed to investigate the syntactic comprehension abilities of individuals with DS (Weitecy & Penka, 2017). The researchers sought to establish if receptive syntax continued to develop into adulthood, or whether development ceased at a certain point in. The authors found that language comprehension was independent of chronological age (CA) and that receptive syntax did appear to develop for a longer period of time than typical syntactic development. Therefore, it was suggested that receptive language abilities may differ in this respect from TD individuals, and interventions may be effective with older children with DS also.

A further recent development in the literature concerning receptive syntax abilities in individuals with DS has explored pronoun comprehension. There has been some debate in the literature concerning whether language deficits in DS are simply delayed compared to TD children, or if there is a specific syntactic deficit (Fowler, 1990). Sanoudaki and Varlokosta (2015) investigated this with the suggestion that there may be a 'deviance' from typical development when it comes to reflexive and personal pronoun comprehension. This research explored the development of pronoun comprehension by examining the abilities of individuals with DS at various ages in order to identify potential patterns of this syntactic deviance. As mentioned previously, language comprehension appears to be an area of relative strength for individuals with DS, however, prior research suggests that there are difficulties comprehending reflexive pronouns. TD children are also reported to have some difficulties with pronouns up until the age of six, however, in contrast to those with DS; the difficulties in TD individuals seem to be primarily with personal pronouns (Varlokosta, 2001). The aim of the research by Sanoudaki and Varlokosta was to examine the deviance hypothesis further in pronoun comprehension for individuals with DS by exploring its relationship with age. Twenty-eight individuals were recruited for the research (14 with DS and 14 TD), and each individual in a group was matched together on the DVIQ which is a test of verbal IQ with the TD children aged between four and five-years-old. In addition, eighteen typical adults were also tested on the task to ensure the validity of the experimental task.

The findings of the research by Sanoudaki and Varlokosta complemented previous reports (Perovic, 2002; Perovic, 2006) that individuals with DS do have a different pattern of development of pronoun comprehension. They reported that individuals with DS have more difficulties with the comprehension of reflexive pronouns in comparison to personal pronouns. Typically developing children usually display greater difficulties in the interpretation of personal pronouns, not reflexives (which is often referred to as the delay in principle B). These difficulties also extended during adolescence and into adulthood for individuals with DS. The analysis of personal pronouns found that there was a significant correlation with age in the DS group, suggesting that performance did progress with age, however, this structure has previously been found to be a relative strength (but of difficulty to TD children). It is also highlighted that this research should be taken as a pilot study, and the results interpreted with caution as the removal of the oldest and youngest participants eliminates the significant finding. This research does, however, provide a useful insight into

the development of pronouns and provides strength to the deviance hypothesis. It is also a useful consideration when targeting interventions with individuals with DS, as it appears that there can be significant benefits with targeted interventions in various aspects of linguistic development (Sepúlveda et al., 2013).

Children with DS also display impairments in their morphosyntactic development with frequent grammatical errors often being made during language acquisition. More specifically, marked difficulties with expressive morphosyntax have also been noted as being particularly challenging (Chapman et al., 1998; Laws & Bishop 2003). For example, Chapman and colleagues (1998) recruited 47 children with DS (aged between 5-20-years-old) alongside 47 TD children who were statistically matched for NVMA. In this study, the authors investigated the MLU of these children in narrative and conversational language samples in order to ascertain if there was any evidence of an impairment in grammatical morphology. Findings showed that the children with DS produced more simplistic utterances with more omissions of word tokens and types than the TD children. The authors concluded that there was evidence of a specific impairment in the children with DS in terms of their morphological development, but that there was no evidence of a “syntactic ceiling.” This finding was reported as the older children with DS continued to increase their MLU with age.

Research has also investigated the morphosyntactic abilities of individuals with DS compared to those of children with DLD. Children with DLD and DS have been alongside each other as they sometimes display similar patterns of strengths and weaknesses in their language development. For example, there are often marked impairments in the grammar and the phonology of children with these disorders (Eadie et al., 2002). DLD is not believed to be a solely genetic impairment as DLD is thought to be a combination of environmental and biological factors, however, there does also appear to be similarities in the cognitive processes that lead to successful facilitation of language in both disorders. For example, impairments in the phonological short-term memory are frequently observed in children with DS and children with DLD. On the other hand, there are differences found in the types of impairments within each disorder, such as that individuals with DS have additional difficulties with syntactic and grammatical tasks specifically.

Laws and Bishop (2003) investigated a number of language domains in individuals with DS in comparison to controls with DLD. Findings of this study showed that those with DS tended to omit regular tense-related morphemes more frequently than the children with DLD and that grammar, on the whole, was more impaired than vocabulary. Furthermore, Fowler (1990) also expressed that there are substantial and unexplained delays in the development of language structure specifically. Regardless of the assessments undertaken, it is reported that children with DS continue to have difficulties with grammar as there appears to be a ceiling of development which is markedly lower than MA equivalent expectations. Consequently, this further highlights the claim that those with DS have a particular profile of development that is not only different from TD children but also differs from children with other language impairments.

3.2.4 Phonological Development

A further area of linguistic development to note is phonological development, which concerns the way that phonemes are structured in language which leads to the formation of syllables and words, which are used to convey a communicative function through speech. As mentioned in the section relating to typical phonological development, one of the first signs of language development is the production of vowel and consonant-like sounds, i.e. babbling. Researchers have compared these early signs of phonological development in TD infants by examining the babbling stages compared to children with DS. Dodd (1972) examined this in infants with DS at both 9 and 13 months of age. Findings showed that the occurrence of babbling began at similar time-points to TD children and that babbling occurred in the infant with DS within the typical development parameters. The Bayley scales of infant development were also compared between the groups, and although overall scores varied between TD and DS groups, scores on the number and variety of consonants and vowels, and number and length of utterances did not differ between the groups either.

Complementing this is another early study that was conducted by Smith and Oller (1981). In this research that compared children with DS and TD children, it was found that there was a very similar language development profile in terms of the place of articulation in babbling production. The consonants were predominantly articulated towards the back of the mouth up to the first six months, and after this, front consonants were produced more in both groups between the ages of seven-nine months. One conflicting finding to this suggests that there are delays in the prelinguistic development of children with DS (Lynch et al., 1995). In this study, it is reported that the onset of canonical babbling is delayed by around two months in comparison to TD children (occurring at around 9 months of age) and that the frequency was less stable. On the whole, the research suggests that babbling seems similar to that of typical development, particularly if employing target stages of development. The early phonological development of infants with DS appears to begin in the same way as TD infants.

Although the early phonological development appears similar for TD versus children with DS, there is a general consensus that 'meaningful speech' is delayed in individuals with DS, although as aforementioned receptive abilities are not impaired to the same extent. Once language development has reached word level, it appears that phonological delays or impairments are still existent. Generally, the occurrence and frequency of babbling are associated with better speech and language abilities in later development (Stoel-Gammon, 1998) suggesting that babbling is a platform for further language development. This relationship does not seem as straight forward in children with DS.

Further studies have compared the phonological production of children with DS to TD children and also found that groups were comparable on measures of phonological development (Smith & Stoel-Gammon, 1983). This study compared the emergent pattern of different speech sounds and found that the development was the same for both groups in terms of consonants with stops, nasals and glides emerging first. Later in development were

the production of fricatives, affricates and liquids. In this longitudinal research of speech sounds, they also looked at the errors made in production between groups. The errors were subdivided into four types, but also combined to look at overall error frequency. At first testing, the children with DS made errors 61% of the time and the TD children made errors 63% of the time. The TD children were re-tested after 12 months, and it was found that this was reduced to 25% error frequency. The children with DS were re-tested after a longer period of time (36 months) and were still found to have a 40% error frequency. This research was one of the first to suggest that the errors made although were similar and occurred at the same frequency in early development, the errors were slower to correct in the children with DS. This coincides with the later research by Dodd and Thompson (2001) which also suggests that the error patterns in children with DS are more varied.

Intelligibility is an aspect of language which is often reported to be one of the greatest challenges for individuals with DS. Cleland et al. (2010) reported on the distinct language markers in children with DS which results in significant difficulties in understanding the speech of these children. This article also highlighted that there may be a high co-occurrence of undiagnosed dyspraxia and the researchers suggest that more research is required to address this factor. This research also addressed some further explanations for decreased intelligibility of speech, such as a smaller oral cavity, motor functioning difficulties and the co-occurrence of hearing disorders. The researchers extend factors to phonological delay and the fact that children with DS appear to make more distorted or unusual phonological processes compared to TD children who make similar levels of phonological errors. The researchers aimed to identify if children with DS have a disorder of phonological acquisition as opposed to speech delay. The findings of this research were that the children presented with atypical and typical errors and that they had underspecified phonological representations. In addition to this, the more common phonological process found was cluster reduction, although other structural simplification processes were found to be fairly common in the sample. Therefore, there may be a specific weakness in the phonological acquisition stage for those with DS.

This has been extended in research by Roberts et al. (2005) who also reported a high occurrence of phonological processes in DS. This study looked specifically at the phonological patterns and accuracy of the production of consonants in children with DS (aged 4-13 years old) in comparison to TD controls (aged 2-6 years old) and children with fragile x syndrome (aged between 3-14 years old). In this study, boys were all matched for NVMA within the three groups. The focus of this research was to look closely at the phonological production of these children as these speech characteristics are said to be important for their implications on assessment and intervention practices. Speech errors were reported to have a high frequency in the children with DS, as a specific delay in phonological correction seemed to occur. More specifically, previous research reported that there is a specific difficulty in consonant accuracy for these individuals, a finding that was replicated in this study, however, stops and nasals seemed to be produced correctly. The correction of phonological errors was

slower, and there were also some abnormal phonological processes found in children with DS that do not generally occur during typical development.

The analysis comprised of measuring the accuracy of consonant production, phonological processes and approximation of phonological word structures. The findings for the consonant accuracy found that overall, the DS participants performed lower than the TD and fragile x groups of consonant accuracy for early, middle and late consonants, with the DS group having 54% accuracy, the TD group 87% accuracy and fragile x group 82% accuracy. In regards to phonological processes, the DS group had a higher occurrence of phonological processes than the two other groups. In addition to this, the research identified specific patterns of phonological development for children with DS and fragile x syndrome. Overall this research suggests that the children with DS have a specific pattern of phonological development that indicates that there is a specific weakness in their phonemic development. The authors do also report large individual differences and this should be taken into consideration when interpreting the results, and this may also explain why the patterns of phonological development are not identical to previous reports. Although these errors are also found in TD infants, the errors continued into childhood at higher rates than would be expected. It is suggested that this may result in lower speech intelligibility, and consequently more difficulties for others to comprehend the speech of children with DS. This is reported to be of considerable concern for parents of children with DS, with reports of children with DS having substantial difficulties with the articulation of speech sounds (Kumin, 1994). On the whole, productive abilities in terms of phonology certainly seem to be particularly problematic for individuals with DS.

3.2.5 Phonological Awareness

Phonological awareness concerns the ability to detect and manipulate speech sounds within a given language. In the majority of languages, it is comprised of three levels; syllables, onsets/rhymes and phonemes. Following from the previous section which discussed phonological development, phonological awareness is a higher level metalinguistic ability, which is often closely related to reading and spelling development in TD children (Friend & Olson, 2011; Lonigan et al., 2000). As noted previously in section 2.1.2, in TD children, PA begins to emerge around the age of three and continues to develop throughout early childhood. Phonological development has been noted as a specific area of weakness for individuals with DS. At the same time, PA development has been associated with reading and spelling development in this population also (Cupples & Iacono, 2002; Stoel-Gammon, 2001). Some researchers have argued of a specific deficit (or lack of all together in some reports) concerning the development of PA in individuals with DS.

One of the first empirical research studies that explored PA in children with DS was conducted by Cossu et al. (1993) where 10 children with DS were tested on a range of PA tasks. The children were aged between 8 and 15 and were reported to perform very poorly on all tasks of PA in comparison to TD controls matched on reading ability (aged 7). This

subsequently led to the belief that children with DS have a complete lack of PA and employed a different reading strategy by associating the written form of a word with the spoken word as opposed to using a phonics-based decoding strategy. Byrne (1997) also reported that children with DS had difficulties in adopting an alphabetic reading strategy, as they appeared to rely on logographic strategies further along in reading development in comparison to TD children who were learning to read.

Further research explored the relationship between PA and literacy development in children with DS (Evans, 1994). In this small scale study, six children with DS were recruited during early reading instruction. Repetition tasks were conducted, with words and non-words, with the addition of PA measures (rhyme, alliteration, blending and segmenting). The findings of this research were that the children with DS had particular difficulty with the PA tasks. The authors concluded that PA is not necessary for the development of reading per se in DS, as those tested were developing reading skills in the apparent absence of PA. This suggestion received further support by Bradley and Bryant (1983) who found that many children with DS are able to read effectively despite their lack of abilities in PA. Previous findings also supported the concept of a logographic reading strategy in DS as opposed to an alphabetic approach as children with DS are also reported to make more semantic errors than TD children (Buckley et al., 1986).

This early work resulted in the assumption that PA is not a prerequisite for the acquisition of reading in children with DS, and that those with DS were completely incapable of developing PA. Since this, however, these claims have been unsupported by further research, and the claims that were made as the result of Cossu et al's research (1993) have been dismissed by apparent flaws in their methodologies and conclusions. For example, some researchers have questioned the conclusion that children with DS have a lack of PA, such as Byrne (1993) as he pointed out that the children in Cossu et al.'s study did not score zero on the PA tasks, and he suggests that the scores that they did obtain might show that they were sufficient for the development of reading in that population. This was also the case for tasks that did not have an element of chance, whereby the children with DS scored well above zero. Therefore, PA in DS has been viewed as more relative to reading ability as opposed to an 'all or nothing' ability.

Following these criticisms of Cossu et al (1993)'s research, Fletcher and Buckley (2002) subsequently adapted PA tasks in order to identify if the abilities of children with DS were being underestimated by previous measures that were used to assess phonological development. The measures were adapted by making visual stimulus available alongside auditory input as this allows a permanent stimulus that would not decay by short term memory deficits. In addition to this, words that only contained 2-4 phonemes were used as individuals with DS are known to have small digit spans which usually fall between this range. This research hoped to identify if the phonological abilities of individuals with DS had previously been underestimated and if there was a relationship between PA and reading ability in individuals with DS. Participants (n=17) were recruited as part of a larger longitudinal

study that aimed at identifying the reading development of children with DS and all attended a MS primary school with an average reading age of a 7-year-old. At this age, PA tasks are believed to be successfully completed. Children were tested in a two-hour testing session on a range of standardized and non-standardized tests, as well as four tests of PA.

The PA tasks were all answered by pointing to a correct image as opposed to giving a verbal response and were always a choice of two pictures, therefore, there was a 50% chance level of a correct response for each trial. The results showed that the group as a whole had a fair degree of literacy ability, although reading comprehension was slightly slower than the development of other aspects. The children tested also performed far poorer on the non-word reading tasks, suggesting that they were not using an alphabetic reading strategy. In terms of PA, the children performed the best on the tasks assessing blending and alliteration, Only one child scored at chance level for all tasks, with the remaining participants scoring above chance on one, two or all three tasks. This suggests that the children in this sample were capable of completing the PA tasks to various degrees. There was also a significant correlation between reading and spelling ability and awareness of phonemes, however, when it came to the non-word reading tasks, the children with high scores on phoneme awareness still struggled with this task. The authors interpret this to mean that “awareness of phonemes may be necessary but not sufficient for acquiring a decoding strategy.” The authors note that the tasks used may have been more accurate in determining the real PA abilities of children with DS as the tasks used are not masked by cognitive limitations, as aforementioned.

In addition, the authors highlight that there is a large variability found in children with DS, and further research is required. Overall, the researchers report that although the DS participants did display poor PA abilities, they were not completely absent as had been previously reported. The authors argued that PA tasks that are designed for use with TD children rely too much on cognitive abilities (e.g. short-term memory skills and attention). These skills are also known for being areas of weakness for children with DS and as a result, prior research that assessed PA in DS had not taken these into account when employing tasks that were designed for use with TD children. The results do, therefore, suggest that children with DS do acquire measurable levels of PA, and highlight that previous research may have been insensitive to the populations under study by the testing measures employed. This research will have important considerations for future research that examines PA within atypical populations and highlights the need to adapt resources and research tools to the children under study.

Further research that supports the finding that children with DS have measurable levels of PA was conducted by Kennedy and Flynn (2003). This research sought to identify the PA abilities in children with DS, in association with their reading abilities. As noted earlier, literacy development is closely related to PA abilities in TD children, however, it is not clear how phonological development occurs in children with DS and if there is the same association with reading development. There were nine participants who were required to be over the age of 5 and had at least six months of exposure to literacy instruction. A variety of testing

measures were employed to determine speech perception, production, memory, expressive language, reading and PA. Testing was split into four sessions and the PA tasks were carefully designed so that they did not depend on short-term memory wherever possible. Participants were requested to point to pictures for their responses in order to overcome expressive language and intelligibility difficulties.

The authors noted some patterns in the results such as difficulties with the rhyme, non-word and spoken tasks with many children not scoring above chance level. Overall, the children who scored best on the literacy tasks also had the highest levels of PA, which suggests that PA did play a role in the acquisition of reading in those children with DS. The relationship between the two is not suggested to be bi-directional, but that literacy instruction directly enhances PA. In addition to this, the research shows that adapted versions of PA tasks are required to obtain more appropriate measures of PA in children with DS that do not rely on verbal responses, or too many cognitive demands (i.e. short-term memory). There are some limitations to consider, such as the small sample size and as a result, the results should be interpreted with caution. In addition to this, a control group would have been beneficial in order to fully interpret the children's performance. In addition, the majority of the assessment procedures had a high change level (1 in 3), and, therefore, more research is required in order to substantiate these findings, possibly with multiple measures of each skill as the authors suggest.

Further research has sought to identify if there is an association between PA and reading abilities in children with DS (Cupples & Iacono, 2000). This research follows on from the findings of Kennedy and Flynn, as the authors aimed to examine the possible association between reading and PA in those with DS. The authors describe the dual-route models of reading, whereby children either learn to read by converting graphemes into phonemes versus whole word reading where a sequence of graphemes is recognised as a familiar word (the lexical route). Previous research suggests that children with DS may opt for the lexical route as opposed to a decoding route due to their difficulty in reading non-words and their difficulty in phoneme segmentation tasks. In Cupples and Iacono's study, 22 children with DS (between the ages of 6;7 and 10;3) initially completed tests of receptive language, cognitive functioning, oral reading, and PA. Reading and PA abilities were reassessed approximately 9 months later. Better oral reading was associated with superior phoneme segmentation skills on reassessment.

Furthermore, there was some evidence that early segmentation ability predicted later non-word reading, but not the reverse. As this study employed a within-group design, the children were not being compared to a control group and the authors suggest that this is a more appropriate way of analysing PA in these populations. It is also important to note that this finding was not significant for all measures of PA. As a result, it still is not clear how PA and reading relate to each other in children with DS, as it appears that some skills may be more closely related to reading abilities than simply all aspects of PA. Overall, the results replicate the findings of Kennedy and Flynn by finding an association between PA and early

oral reading ability in children with DS. The authors conclude that PA does play a central role in reading development in DS. Cupples and Iacono (2002) also provide evidence in a further study that PA can be developed in children with DS if provided with appropriate training.

One final investigation of the issue of PA in DS and its role in reading was conducted by Lemons and Fuchs (2010). In this article, the authors reviewed 20 papers that explored PA in individuals with DS and its role and correlation with reading ability. The overall findings suggest that although children with DS do perform worse on measures of PA than TD children, their abilities are still related to their ability to read and there are correlations found between measures of PA and reading. The studies that were selected in this analysis employed a range of matching techniques (chronological age, reading ability, MA) and also employed a range of PA measures comprising of phoneme, rhyme and syllable tasks. In addition to being related to reading development, this may have an impact on language development as often reading and language develop simultaneously. For example, a group of TD children were assessed on PA and language and it was reported that rhyme judgement tasks were correlated with all general measures of language (Smith & Tager-Flusberg, 1982). Similarly, Bowey (1996) also reported that PA was very closely related to the vocabulary sizes of TD 5-year-old children. This may also be the case for children with DS, as PA and language are both areas of difficulty for this population, and developments in one area may be correlated in development in the other. It has also been proposed that interventions that target PA should be implemented within speech and language therapy (SLT) as a means of improving PA, speech production and reading (Gillon, 2000). This was specifically for children with a DLD, however, it may also be useful to extend this to children with other DDs.

In addition, research has attempted to identify the effectiveness of intervention programmes in relation to PA and letter knowledge in children with DS (van Bysterveldt, Gillon & Moran, 2006). As PA in children with DS has been identified as an underdeveloped skill, the researchers sought to identify if parent-led interventions that targeted PA would result in successful outcomes. The present study employed print referencing techniques and the sample was chronologically age-matched to a TD control group who did not receive any type of intervention. Phonological awareness tasks were designed and implemented and groups and were both tested twice (pre and post-intervention). Four-year-old children were recruited for the research and were tested on a range of standardized language assessments and the experimental assessments that were designed to measure PA and letter knowledge. In the experimental tasks, no verbal responses were required in order to control for expressive language deficits or intelligibility impairments for the children with DS.

Following the intervention, the children were re-tested on all measures and there was a significant increase in performance for the children with DS on measures of letter-sound knowledge, print concepts and initial phoneme identity. There was, however, a great deal of variation in performance for the individuals with DS as some made vast improvements whereas others were only marginal. The results were also measured against the average gains that the control group made, and overall the authors report that the group who had PA

interventions, did improve to a greater degree as a result. The authors suggest that the reason why some of the participants may not have benefited from the intervention could be due to their low scores on other measures of language development. It is suggested that the ability to benefit from the intervention required a certain level of language ability. If these results are accurate, the implications are promising as this would suggest that parents are capable of influencing the development of PA and subsequently reading ability.

Similarly to the intervention study conducted by Van Bysterveldt et al (2006), Sepúlveda et al. (2013) also reported that interventions seem to be successful with the majority of language features within DS, specifically syntax, morphology, and semantics. One area that did not seem to develop as well with targeted interventions was pragmatics, however, overall intervention programmes seem to be successful as a clinical and educational tool for individuals with DS. One final piece of evidence that supports the utility of interventions was conducted recently by Lemons et al. (2015). In this study, the author also indicated that children with DS are able to benefit from early PA interventions. It is concluded that this type of instruction should be provided to children with DS in the future to enhance their apparent deficits in PA.

As noted in an earlier section, hearing disorders are often concurrent in individuals with DS. The resulting impact that this may have on PA should also be considered. A second consideration that is required when assessing individuals with DS is that there are often deficits found in short-term auditory memory which will also impact any research that explores PA. It seems appropriate to conclude that although children with DS are reported to have significant difficulties with PA in all the studies above, more recent research suggests that PA is not completely lacking in these individuals particularly if the methods employed overcome working memory and expressive language deficits. Interventions also appear to have successful outcomes in the research that exists to date in this area.

3.2.6 Conclusion

The research presented in this section shows that individuals with DS have language impairments which are greater than would be expected given general cognitive abilities. Consequently, it appears that there is something specifically related to linguistic processing which leads to these observed deficits in the various domains reported on above. Furthermore, some elements of language appear to be impaired to an even greater extent compared to other areas. As a result, this supports the proposal that language is modular as opposed to being a singular processing system given the strengths and weaknesses observed in individuals with DS (Perovic, 2006). Those with DS have a specific language phenotype comprising of substantial difficulties with various elements of language which are not replicated in TD children and adults, or individuals with other language impairments. By identifying how this language profile can then be extended to bilinguals with DS will provide a novel insight into language development in DS.

3.3 Down Syndrome and Bilingualism

The field of bilingualism and DS is very limited in the literature to date. Early research in the area has employed case study designs with later group studies tending to focus on simultaneous bilinguals. There is also somewhat of an overlap of participants in some research studies whereby a lot of the data that currently exist in the field have used the same populations in Canada. As a result, the majority of the research in the field concerns only English/French bilinguals. The following section will provide an overview of the research that exists to date concerning the abilities of children with DS to become bilingual.

3.3.1 Early Case Studies

The first study that reported on bilingualism in an individual with DS was conducted in 1993 by Vallar and Papagno (1993). This case study concerned a multilingual adult with DS who was 23-years-old at the time of data collection and who a successful trilingual speaker dominant in Italian. One of the aims of the research was to explore the role of phonological short-term memory on language acquisition. It was reported that the individual had relatively enhanced verbal short-term memory as measured using a digit span test, however, she did have some weaknesses in terms of verbal and NV reasoning and long-term verbal memory. The authors concluded that short-term verbal memory played a role in her successful language acquisition, as she displayed strong verbal abilities.

A second case study that followed the findings of Vallar and Papagno was conducted by Woll and Grove (1996). In this study, a pair of twins who were born with mosaic DS were studied. Interestingly, the twins were born to parents who were deaf and used BSL as their primary form of communication. The twins also received English language exposure through other family members and also received their educational instruction through English. The twins under study were in a unique situation with the opportunity to obtain fluency in two languages that used different modalities. It was hypothesised at the beginning that BSL would be the strongest language as research suggests that children with DS make more progress with signing than with spoken languages, and as they had frequent input of BSL as it was the only home language. It had been previously suggested that the visual-motor modality would be easier to access, recall and produce, and this natural exposure to two modalities enabled a unique insight into the development of these two language modalities within DS. The research began when the twins were aged 10 and comprised of oral recordings from their home and at their school. In addition, they were tested on a range of cognitive and language assessments. One twin, Sally, was reported to outperform her sister in the majority of aspects concerning her language ability, however, both had a high fluency in both BSL and English.

Findings suggested that both twins were full and 'functionally bilingual', however, they still had a preference for verbal communication through English and were considered to be English dominant. When the twins were in isolation they both chose to speak in English even though the twins were exposed to BSL first and this was the original home language. Difficulties with morphosyntax were evident in both spoken English as well as BSL, however,

different structures posed different challenges in each modality. For example, the twins had well-developed abilities with plurals in English, although these structures in BSL were not as well-developed. These findings suggest that hearing impairments which are often apparent in those with DS are not wholly responsible for the deficits observed in spoken language in domains such as phonology and grammar as these impairments were evident in the twin's BSL also. The authors concluded that the impairments related to language development which are found in DS are not modality-specific and that the linguistic system of BSL was no easier to learn for the twins than spoken language.

3.3.2 Vocabulary and Lexical Abilities

The first piece of empirical research conducted in a group design was undertaken by Kay-Raining Bird et al. (2005). This is one of the most influential pieces of research in the field of bilingualism and DS to date given the sample size compared with previous research (with the inclusion of 20 children with DS). This study was undertaken in order to gain an understanding of the language abilities of children with DS who were bilingual, as there was a substantial lack of understanding regarding these individuals' capacity to acquire more than one language. Expressive language difficulties are often a marked impairment for children with DS and as a result, some might automatically assume that two languages would pose too big of a challenge for an individual with such a language profile. Fifty-one children participated in this research and formed four groups (bilingual DS, N = 8; monolingual DS, N = 12; monolingual TD N = 18; and bilingual TD N = 11). Participants were matched by group for NVMA, both within the two groups of children with DS and between the groups of children with DS and the TD groups.

The inclusion criteria stated that children with DS had to be within the very early stages of language development with a minimum of 100 productive words, and a MLU of <3.5. Parents were also given a language background questionnaire to gain an understanding as to the child's language history as well as completing the Communicative Development Inventory in English, the Preschool language scale, a vocabulary comprehension test and the authors took a language sample. The results of this research reported that for general language abilities, there were no significant differences between the bilinguals with DS and the monolinguals with DS. Deficits were found for the two groups with DS (as would be expected), particularly concerning morphosyntax. This supports previous claims in the literature that morphosyntax is one of the areas where the most substantial impairments are found for individuals with DS.

The findings suggest that there is no detrimental impact of bilingualism for children with DS as there were no significant differences between groups in terms of vocabulary sizes, however, there was variability in abilities across both groups. It is important to note that the bilingual DS group had a higher CA and NVMA, although this was not statistically significant. The authors concluded that some children with DS were capable of becoming successful bilinguals, although there are large individual differences that should also be considered. The

authors also state that this is the first study to look at bilingual abilities in those with DS. Therefore, future research is required to substantiate these claims with larger sample sizes and some children with DS may be more successful than others in becoming bilingual.

One further piece of research in the field of bilingualism in DS was conducted by Trudeau et al. (2011). This research focused on the lexical development of French/English bilinguals with a diagnosis of DS and data was collected by means of a parental report measures. A total of 18 participants were recruited and parents were asked to document vocabulary sizes. This research employed a longitudinal design to monitor development over a period of time for the majority of the participants. There was again found to be a substantial variability on second language performance both at the beginning and at follow-up testing sessions, however, the development of a second language was not reported to hinder the development of the first language as similar abilities were found between monolinguals and bilinguals with DS. In addition, all children who were assessed at follow-up had increased their vocabulary size in English. The rate of language acquisition for the second language was found to be of a linear relationship to L2 language exposure. The authors concluded that their findings coincide with the findings of Kay-Raining Bird et al. (2005)'s study, which was that children with DS are able to acquire two languages and function as bilinguals, however, this will be dependent on the frequency and consistency of exposure to a second language.

One final piece of longitudinal research has been conducted into bilingualism in DS and is reported on in an unpublished PhD thesis (Valdivia, 2005). In this small-scale study, four children in California (3 males, 1 female) were followed for a period of 6 months with observations and interviews taking place. In this qualitative socio-cultural project, case reports were presented for the children by means of observations within the home, school and community as well as interviews with family members. The research aimed to gain an understanding of the children's language abilities and the author reported that all four children displayed 'communicative competence' in their two languages. It is noted that the children were in the very early stages of language development (some even being pre-verbal). The four bilingual children with DS are reported to have appropriate language abilities that were comparative to age-matched monolingual children with DS, although the author notes the requirement for further research in this field.

3.3.3 Morphosyntax

Morphosyntax is a further area that is reported to be specifically impaired in those with DS, as aforementioned. A study to investigate morphosyntax in bilinguals with DS was conducted by Cleave et al. (2014) and this experimental research used a matched design study with bilingual and monolingual individuals with DS alongside a TD control group (aged between 3-6-years-old). As morphosyntax is impaired in individuals with DS, this research aimed to explore this ability within bilinguals with DS, with a specific focus on syntactic bootstrapping. Syntactic bootstrapping is a concept that proposes that children acquire meaning by applying knowledge of syntax to decode sentences and recognising syntactic categories (Gleitman et

al., 2005). This dynamic fast mapping ability was targeted in the study by Cleave and colleagues as it is stated that this skill is a good measure of morphosyntactic ability. It is a skill that is also reliant on skills that are often lacking in individuals with DS, such as attention to language form and the lexical representation simultaneously. Findings showed that the TD group outperformed the DS groups as was expected, given the impairments reported in this domain for those with DS. In comparing the bilinguals and monolinguals with DS, it was reported that there were similar results, with only a 1% margin between the abilities found in the two groups. The authors interpret this as bilingualism having no detrimental impact on this ability. There was not, however, any evidence of a bilingual advantage as the authors hypothesised, given previous findings with TD bilingual populations. This research also complements that of Kay-Raining Bird et al. (2005) with the viewpoint that bilingualism does not further impair language development. The authors also recognise the need for future research that employs experimental designs to gain a greater insight into bilingualism and DS, and highlight that different patterns of bilingualism should also be explored (e.g. simultaneous versus sequential bilingualism).

In a follow-up study, four children with DS were assessed on measures of semantics and morphosyntax. This study used an individually matched design with two matched children for each of the four children with DS, one who was a TD bilingual, and the other a monolingual child with DS (Feltmate & Kay-Raining Bird, 2008). Children in each triad were matched on NVMA and second language exposure (for the bilinguals). The focus of this research again concerned morphosyntax due to the observed impairments in this population. There were 12 children in total and participation inclusion criteria stipulated that they had to be in the early stages of language development (less than 100 productive words and MLU <3.5.). The Bead memory and pattern analysis was used in order to determine NVMA for matching purposes and a questionnaire was completed by the parents concerning language exposure and demographic information. A language sample was also taken which constituted of 20 minutes of speech while the children were given toys to play with.

The results were checked for reliability and scores of less than 75% agreeability were excluded from subsequent analysis. Findings showed mixed results with no clear positive or negative effects found for bilingualism. It was concluded that the bilingual children with DS were developing functional second language skills and these included semantic and syntactic abilities. As is often reported with this population, there were large individual differences in expressive language abilities and some participants consistently outperformed others. Various factors were suggested to play a role in this, for example, parent education, CA, auditory-verbal memory and hearing abilities. The authors concluded that there were no consistent differences between the groups, suggesting that bilingualism did not appear to be a hindrance to these individuals in terms of their morphosyntactic language development.

3.3.4 Cognitive Abilities

Researchers have also explored the cognitive impact of bilingualism within children with DS (Edgin et al., 2011). There is a well-established debate within bilingualism research that bilinguals may have some advantages as a by-product of being bilingual in TD individuals (see Section 1.1.4; Cummins, 1979; Bialystok & Martin 2004; Adesope et al. 2010). Subsequent to this, it has been proposed that these benefits may also be observed in children with DDs, including those with DS. In Edgin et al.'s research, 41 participants were recruited, including bilinguals and monolinguals. Exposure to a second language of one hour or more per day only was required to be considered bilingual. Measures were taken to ensure that the groups were equally matched in terms of NVMA, CA, social background and gender. Findings revealed no significant differences for language ability, nor any measure relating to cognitive or adaptive functioning between the bilinguals and monolinguals. This research only measured the majority language (English) and it is also not clear if all participants were English dominant, nor was it noted if the children under study were simultaneous or sequential bilinguals. These factors would have implications on the findings of the study. The authors conclude that bilingualism again did not hinder language development, however, it did not appear that there was any evidence of bilingualism having an advantage over monolinguals for this population. It was also highlighted that second language exposure may not always be appropriate for every individual with DS and it is essential to consider each individual on a case-by-case basis.

3.3.5 Phonological Awareness

As noted earlier, one area that individuals with DS find particularly challenging is PA. Initial reports suggested that PA was completely lacking in those with DS (Cossu et al., 1993), however, adapted measures have found that children with DS do have measurable levels of PA (Kennedy & Flynn, 2003) which are more appropriate for use with children who have limited memory capacities and expressive communication difficulties. In turn, PA may also relate to reading and spelling development in both TD children and children with DS. In TD bilinguals, it is reported that there is a bilingual benefit in this metalinguistic ability (see section 1.1.4; Bialystok et al. 2003; Chen et al., 2004; Campbell & Sais, 1995). Consequently, the question arises as to whether individuals with DS will also show benefits in PA as the result of exposure to a second language. If this is the case, this would have a substantial impact and a range of implications on the use of bilingualism for those with DS. In addition to this, any benefits found in terms of PA development may also transfer to benefits in reading and spelling development, with substantial implications for education.

To date, there is only one piece of research that has explicitly assessed PA in bilingual within DS (Burgoyne et al., 2016). In this single case study, the authors assessed language, PA and reading ability in an individual with DS who was a competent bilingual in English and Russian. The aim of the research was to identify the extent to which a second language influenced PA development and the resulting association on reading. In some children with

DS, commensurate levels of reading have been obtained compared to their CA (Hulme et al., 2012) however, in some individuals reading ability seems to surpass expectations given levels of PA (Lemons & Fuchs, 2010). Phoneme awareness and letter knowledge, however, seems to be the most accurate predictor of reading ability. The researchers hypothesised that bilingualism may have a positive effect on PA and reading ability in this individual with DS.

The child under study was compared to other children with DS who only spoke one language, and also compared to TD bilinguals (aged 6 to 7-years-old). The bilingual with DS was a sequential bilingual who was exposed to Russian as her L1 from birth, and English as her L2 at the beginning of formal education. She was tested on a range of standardised tests of cognitive and linguistic ability (digit and word recall), as well as bespoke PA tasks. In addition, adapted standardised tasks which enabled testing of her Russian abilities were also employed. These were administered over three sessions, however, all comparison groups were tested at around her second session. The language assessments employed were the BPVS (Dunn et al., 1997) and CELF-IV (Semel, Wiig & Secord, 1998), which were used to assess expressive and receptive language and a section of these were also translated directly into Russian. Reading and PA were tested in both languages and these were matched on word class, word frequencies, number of syllables and phonemes, as well as consonant-vowel structures.

Overall the bilingual with DS displayed generally well-developed language skills in her two languages, and an appropriate level of reading ability given her disability, however, she had relatively poor performance on PA. Compared to the monolingual group with DS, she scored better than average on reading and speech, although her scores on the PA tasks were not significantly different. The authors concluded that she did not have any enhanced PA by being bilingual. Interestingly, the patterns of PA were very strong in one language but very poor in the second language. The authors concluded that the case study provides evidence that children with DS can become competent and fully functional bilinguals, particularly in terms of speech and reading. It is important, however, to consider the fact that the individual's parents were very well educated, which may have assisted in her speech and reading development. In addition to this, she was selected for this study after having been incorporated in a reading intervention program, which may have explained the unusual pattern of PA across languages. It is also highlighted that this research finds no reason to suggest that a child with DS should not be taught in just one language.

In addition to this, one finding which emerged as an unexpected finding was reported by Vallar and Papagno (1993), when they incidentally discovered that their case study of a 23-year-old trilingual with DS also had unusually high PA skills for an individual with DS. The individual with DS who spoke Italian, English and French had stronger expressive abilities as opposed to non-verbal cognitive abilities which are usually found to be the opposite in the majority of children and adults with DS. It was hypothesised that the individual's multilingualism may have resulted in her particular skill profile, and more specifically the unusually high PA skills that she displayed. The reports of higher than expected PA abilities

were not explored by the authors, however, there may be several possible explanations. For example, the high lexical similarity between Italian and French, the transparency of the orthographies, or indeed bilingualism itself. This area warrants further investigation.

Finally, there is one known study of PA in a group of bilinguals with ASD, reported on in an unpublished thesis (Pereda, 2013). Given that there are apparent metalinguistic and cognitive benefits within TD bilingual populations, the research aimed to identify if these benefits would translate to bilingual children with ASD. Fourteen children were recruited, six of which were Chinese-English bilinguals and were assessed on measures of PA and attentional control. Although correlations displayed consistent performance between attentional control, NV and verbal abilities, there were no significant differences between the bilinguals and monolinguals. The lack of a bilingual benefit in this population may be evidence that the advantages found in TD children do not extend to ASD, or may have been due to the limited effect size due to the small sample. Importantly, however, is the fact that there were no disadvantages reported for this population by being exposed to a second language. To date, there are no group studies which have evaluated PA in bilinguals with DS.

3.4 Conclusion

This field of research has begun to develop over the past two decades, however, there is still very limited information available to guide clinicians and parents as to the outcomes of bilingualism on language development. The majority of research that exists to date tend to be case studies or have limited sample sizes due to the difficulties involved in recruiting such a select sample, and there is generally a large amount of individual variability in DS. Furthermore, there is no study to date that employs intervention studies or longitudinal design. Consequently, intervention studies may be useful in this field of study also. In addition, research in TD bilinguals has highlighted the potential benefits of bilingualism as discussed in section 1.1.4. It is interesting to speculate whether these benefits are also evident in children with DS who are bilingual. Ultimately, in order for any individual to become a successful bilingual, there needs to be a supportive environment which enables the development of both languages which will include adequate input in terms of quality and quantity in the two languages, and this will also certainly be the case for individuals with DS.

The field of language development within children with DS has primarily focused on the language profiles of children who only speak one language, however, given that there is a considerable number of bilingual speakers across the world, with estimated ranging from 50-70% (Baker & Jones, 1998), bilingual language development within DS is an area requiring investigation. Research in this field is scarce, and consequently, the fundamental goal of this research is to develop our understanding of this area, particularly in relation to bilingualism in the UK. This is the first piece of empirical research to investigate this issue within the UK and will specifically address bilingualism in Wales within the Welsh language context. The research will provide a comprehensive overview of the language abilities of bilingual and

monolingual children with DS, and will also target specific areas of linguistic development such as PA which is known to be an area of difficulty for these children, but a strength for TD bilingual children. Consequently, the development of PA in bilinguals with DS may even provide some benefits to this area of linguistic development.

The research will have important implications which are threefold. The first is within the clinical practice domain, as it seems that practitioners working with bilingual families may be unaware of the expected language outcomes for this population. This research aims to guide professionals by obtaining information regarding bilingualism in DS that may be useful for future clinical practice. In addition, by identifying strengths and weaknesses in bilinguals with DS, this may also assist practitioners in formulating appropriate interventions tailored to each individual, and each family's circumstances. Interventions that have been employed in children with DS have proved to be successful in various aspects of language development including syntax, morphology, and semantics (Sepúlveda et al., 2013). As a result, intervention programmes could be a successful clinical and educational tool for those working with individuals with DS, which would be tailored in line with the findings of this research.

Secondly, the research may have the potential to impact educational policies in Wales. Although bilingual policies exist in that Welsh should not be seen less favourably to English (as noted in section 1.1.1), it is unclear how to approach the Welsh language and modern language classes within children with DS. This research may inform future educational policies and may assist schools in providing the most appropriate educational provisions. Finally, parents may also benefit from the knowledge that will be obtained from the research and may assist in guiding their expectations and enable them to make more informed decisions in terms of language exposure and language in education. Ultimately, this research aims to provide a comprehensive overview of the language profiles that exist for Welsh-English bilinguals with DS.

3.4.1 Aims and Outline of this Research

There are four main aims of the current research. Firstly, this research aims to examine whether there are any differences in language development for children with Down syndrome who are bilingual compared to monolinguals with DS on a range of linguistic skills. This aim will be met by comparing groups of children with DS who either speak Welsh and English or only English after carefully controlling for NVMA, socioeconomic status (SES) and working memory (WM). The second aim is to identify the patterns of language acquisition for bilinguals and monolinguals with DS by identifying strengths and weaknesses in both groups in order to establish if the groups follow similar trajectories of development or not. Additionally, this research will specifically investigate the development of PA. Given that PA is typically an area of weakness within DS and that PA is an area of strength within bilinguals, this research hopes to ascertain whether this benefit is extended to bilingual children with DS. If there is any evidence of a bilingual benefit within this domain for those with DS, this

will have important implications for future interventions, with further implications for education policies and practice. Finally, the language abilities of bilingual children with a dual DS-ASD diagnosis will be explored in Chapter 7 as, to date, this population has not been explored in relation to bilingualism.

Down syndrome is the most common chromosomal disability with a range of impairments including physical, cognitive and linguistic elements. Language impairments are often an overarching concern for parents of a child with this disability, particularly as they tend to be greater than would be expected given relative cognitive abilities. Generally, these children are found to have a greater understanding of language than spoken or productive abilities, however, there is also a great deal of individual differences. Research in the field of language development within children with Down syndrome has primarily focused on the language development of children in one language only, however, there are a considerable number of bilingual speakers across the world, including in Wales which is the only officially bilingual country in the UK. The patterns of language development within Welsh-English bilinguals with DS is an area requiring investigation.

Of the limited research that does exist, the findings to date suggest that children with DS are not at any disadvantage if they are exposed to a second language. Previous research that has compared the language abilities of bilinguals and monolinguals with DS by comparing groups on measures such as vocabulary sizes and morphosyntax have found that the language abilities of the two groups of children are comparable. Therefore, the addition of a second language may not be a detriment to language acquisition. This is in contrast to what some may automatically assume given that language delays and impairments are often apparent within children with DS. This has not been researched in the UK and therefore, this research seeks to address this gap in our knowledge. In addition, this research will specifically target an area that is known to be lacking in DS, which is phonological awareness. In typically developing bilinguals, however, this is a skill that seems to be enhanced. Subsequently, bilingualism in DS may offer some benefits to this area of linguistic development. Phonological awareness has also been closely associated with later reading and spelling abilities, therefore, this may have further implications on other areas of development.

The current research aims to expand our knowledge by investigating the language abilities of children with DS who are bilingual by building a language profile of bilingual children with DS. The project will specifically focus on Welsh-English bilinguals, given that Welsh has an official language status. As there is support for bilingualism through the language inclusion policies, this seems an appropriate population to investigate this issue. As there has been an increase in bilingual speakers across the world, it is essential to consider how bilingualism or multilingualism impacts the development of children with DS. Given that children with language impairments are often reported to be advised to avoid bilingual exposure, it is crucial to identify if this is a suitable recommendation in order to guide future clinical and educational practice. Therefore, this research aims to provide a comprehensive

overview of language development within this population. The research questions are as follows:

- I. What are the language abilities of children with Down syndrome who are bilingual?
- II. How does language development compare for children with Down syndrome who are bilingual versus monolingual?
- III. How does phonological awareness develop in bilinguals and monolinguals with Down syndrome?
- IV. Are there any factors that predict language outcomes in bilinguals with Down syndrome?
- V. What are the language abilities of children with a dual diagnosis of Down syndrome and Autism Spectrum Disorder who are bilingual?

In line with findings to date for bilingual children with other DDs and preliminary reports on bilingual language development for children with DS, it is anticipated that there will be no significant differences between the two groups of children with DS on measures of receptive and expressive speech, with comparable performance on assessments of language development in English. In terms of the PA assessments, it is anticipated that the bilinguals will perform at least as well as the monolinguals on these tasks with the possibility that they may outperform the monolinguals, given that TD bilingual children are reported to show enhancements on this ability. More specifically, phoneme awareness has been found to be enhanced the most, particularly in Welsh-English bilinguals. Therefore, if there is any evidence of a bilingual advantage, it is speculated that this will be most evident within the phoneme level tasks. It is anticipated that this research will provide a novel insight into bilingual language development in Down syndrome, which has the potential to impact families, educational policies and clinical practice. This is the first empirical group study of PA in bilinguals with DS and consequently, the findings of this research will lead to a greater understanding within this population. This research will also result in the potential to determine which methods of teaching should be used in order to facilitate the development of PA in relation to reading and spelling outcomes in bilinguals with DS.

Chapter 4: Methodology

The aim of the current research is to investigate bilingual language development in children with Down syndrome in the UK and more specifically within the Welsh language context with Welsh-English bilinguals. This chapter will provide an overview of the method employed in obtaining quantitative data relating to bilingual language development within this population. This chapter will outline the method that was employed for the research, with a more detailed method section presented in subsequent chapters in line with the specific research questions being answered and the samples included in each part of the research. In the present chapter, the methodological processes that were selected for undertaking this research will be described and justified. This includes describing the materials that were designed, adapted and used for the research which are described in detail in section 4.1.2, alongside reports of the practical procedures employed, such as participant recruitment and engagement. Following this, relevant demographic information regarding the participants is presented, which includes their language backgrounds, information pertaining to them and their families and the matching processes undertaken (section 4.1.3). The ethical principles and procedures concerning the research are also presented and justified.

4.1 Design

This research employed an experimental quantitative design whereby four groups of children were recruited from different regions across Wales with one additional participant located in England. The participants in all groups were assessed on a range of standardised tasks which measured general non-verbal cognitive development, as well as standardised Welsh and English language assessments. In addition, children were tested on specially designed phonological awareness (PA) and working memory (WM) measures. Participants were recruited to one of the following groups; Down Syndrome Monolingual (DSM), Down Syndrome Bilingual (DSB), Typically Developing Monolingual (TDM), Typically Developing Bilingual (TDB). The shared inclusion criteria for all four groups was confirmation from a parent or guardian that the hearing status of the child was normal or corrected with no more than mild hearing loss. This design allowed for children with mild hearing loss to be included in order to obtain representative groups that were a reflection of the realistic characteristics of individuals with DS, as many children in this population frequently have mild hearing loss and the use of hearing aids is common.

Children recruited to the two DS groups were required to have a confirmed diagnosis of DS by a parent or guardian and to be between the ages of 5-16 at the time of data collection. Parents of these two groups were also asked if their child had any additional disabilities or health concerns that may impact language development and were excluded if they were reported to have an additional diagnosis that may impact linguistic development

(e.g. dual diagnosis of ASD and DS)¹. For inclusion in the TD groups, parents were asked to confirm that their child had no suspected or diagnosed developmental disability and no reported or suspected language disorder. Children were matched on developmental age or non-verbal mental age (NVMA). This resulted in the typically developing (TD) children being chronologically younger than the DS groups, with the children in the TD groups aged between 3-7-years-old.

In terms of inclusion in the bilingual groups, children were required to have substantial input in two languages, specifically in English and Welsh. As noted previously in Chapter 1, bilinguals were required to have communicative needs in both languages. This generally meant that bilingual children received exposure to Welsh either at school, home or both. All participants within the bilingual groups were required to have been exposed to Welsh by the age of three and to be consistently exposed to both English and Welsh, as reported in the questionnaire (described below). In terms of language dominance, the children could be dominant in either Welsh, English or considered balanced bilinguals. Inclusion in the monolingual groups required reported language exposure to English as the majority input language, while allowing limited unavoidable exposure to Welsh through schooling (due to the legal requirement to have some Welsh language teaching in all schools in Wales). Furthermore, as all the monolingual children (besides one) resided in Wales, they all possibly received small amounts of exposure to Welsh through bilingual signage, resources and media present in the public domain. English language input was required to be at or above 85% of lifetime and current exposure for the two monolingual groups.

Data was collected from a range of locations across Wales, with one additional (monolingual) participant being recruited from England. This allowed a broad assessment of the range of language abilities that exist across the country and a sample that represented a large area of Wales. Data was collected from the following counties: Gwynedd, Flintshire, Rhondda Cynon Taff, Vale of Glamorgan, Newport, Ceredigion, Anglesey, Denbighshire, Neath and Cardiff, as well as the one participant from Essex in England. Further information regarding the participants is presented in section 4.1.3 below. The children were assessed on a range of language and cognitive assessments, which are explained in detail in section 4.1.2. The bilingual children were also assessed on their Welsh language ability. All children were tested in a one-to-one setting with the same researcher.

Testing sessions were divided as required according to the needs of each child, although separate sessions for the Welsh and English assessments were conducted in different sittings which were on different days wherever possible. During the Welsh sessions, only Welsh was spoken to the children and during the English sessions, only English was spoken to as to reduce code-switching as much as possible and to put the children in a

¹ Four children were excluded from main analyses due to additional diagnoses of ASD, however, these are reported as separate analyses in chapter 7.

‘monolingual mode’ (Grosjean, 2012), to the extent that this was possible. The first session for each child was always conducted in English with the administration of the English assessments. This was decided to enable direct comparison with the monolingual groups as the English tasks were administered in an identical order for bilingual and monolingual groups. This also eliminated any practice effects that would result from having two similar PA measures for the English assessment as any apparent practice effects would be exclusively restricted to the Welsh assessment of PA. It would not be possible to control completely for practice effects as it is necessary to obtain language assessments in a child’s two languages. The possibility of practice effects is inevitable when assessing two languages as it is not possible to directly measure the same ability in both languages without any differential transfer effects with equivalent tests.

Full ethical approval was obtained from the College of Arts and Humanities Ethics Committee at Bangor University prior to all data collection (see appendix item 2). Ethical considerations were treated very carefully due to the nature of the target populations and their families in relation to; confidentiality and anonymity, appropriate data storage and handling, time considerations, the suitability of measures used, location and environment of testing and child protection (with the primary investigator obtaining a DBS prior to any contact with families). In addition, discussions regarding the questions asked in the questionnaire (see the following section) were undertaken to ensure that they were not too invasive, were appropriately worded and all provided useful information that would be required for this specific research project. A copy of the questionnaires used for the TD and DS groups is presented in the appendix (items 5 and 6). Participants and their families were made aware that they could change their minds about participation at any stage and received full information sheets from the researcher regarding the aims of the project before providing written informed consent (for example consent form see appendix item 3).

4.1.1 Procedure

Prior to the recruitment of the children and their families, the materials described below were piloted with an adult to ensure the protocols for each test were clear, and that the measures that were designed were appropriate and well structured. Following this, piloting with a TD bilingual and TD monolingual child was undertaken to ensure that the tasks were of an appropriate level of difficulty with no evidence of ceiling or floor effects. Piloting with TD children also ensured that the requirements of the tasks were clear and easy to follow and also to highlight any other issues with the assessment procedure that needed adapting. Finally, the assessments were also piloted with a bilingual adult (aged 18) with DS to ensure that the tasks were suitable for the target population and to further identify and eliminate any possible ceiling effects within this population. During this phase, it was noted that the *Prawf Geirfa* (a measure of Welsh receptive vocabulary) was of difficulty to the children and subsequently only the first third of the test (the test increased in difficulty as the assessment

continued) was administered during testing for the remainder of participants (see section 4.1.2 for an overview of this assessment).

Upon completion of piloting, suitable children and their families were identified with the assistance of appropriate organisations working in the appropriate regions. Families who expressed an interest in participating in the project were then contacted to ascertain whether they fitted the inclusion criteria for the research, as described above. Arrangements were then made to meet with the parent or guardian and the child at a suitable location. The majority of families chose to meet at their homes as this was an environment that the children were accustomed to. If the parents asked for the testing to take place in their school, the headteacher and staff working directly with the child were contacted to obtain their approval to visit the schools at a convenient time. All children were tested in a quiet environment in a one-to-one setting with the researcher, with short breaks or separate sessions when required.

The bilingual children all had at least two sessions as noted. These were on separate days for the majority of participants. Due to time constraints and geographical restrictions, this was not possible for all participants. The English sessions lasted between 1 hour and 15 minutes to 1 hour 45 minutes, depending on the response latency of the children during testing and how quickly they became comfortable to begin the assessments. The Welsh session lasted between 1 hour and 1 hour and 15 minutes, again dependent on the speed of response times and the children becoming relaxed in the Welsh environment before beginning with the tasks. The testing procedure was in a fixed order for all children with the tasks deemed most important presented first. If any of the children refused or seemed uncooperative with a particular task, this was then bypassed to the next task and continued in the fixed order. The task that had been skipped was then re-introduced at the end of the assessments (although this only occurred for two participants). A summary of the testing protocol is presented in Table 14.

4.1.2 Materials

All families involved in the research received information sheets (appendix item 1), a letter explaining the research and what would be required (appendix item 4) and consent forms (appendix item 3). These were required to be read, understood and completed prior to data collection. Following this, information was obtained about the participants in the study by asking parents or guardians to complete a background questionnaire (appendix items 5 and 6). Assessments were then administered to the children to gather quantitative data regarding the children's cognitive and linguistic development using the standardised and specially designed test materials described below. The measures selected were designed to be suitable for children with DS, specifically in relation to potentially limited memory capacities that are often apparent in children with DS (see section 3.2.1). This included providing visual stimuli when possible and appropriate according to each test manual's instructions. Tasks with visual stimuli were selected wherever possible with the aim of also keeping attention on task as those with DS are often visually orientated (Jarrold et al., 2000). In order to overcome any

speech intelligibility issues and any potential expressive language impairments, responses were non-verbal when possible with the exception of the tasks that were explicitly designed to measure expressive language abilities.

Groups	Assessments administered
Monolingual and bilingual	
Cognitive assessment	KBIT-2: Non-verbal matrices subtest
Working memory assessment	Forward digit span verbal recall
English language assessment	CELF-P-2: Sentence structure, Word structure, Expressive vocabulary, Concepts and following directions, Recalling sentences, Basic concepts and Word classes subtests.
Phonological awareness assessment	Specially designed phonological awareness measure – Rhyme Detection, Rhyme Generation, Syllable Detection, Syllable Deletion, Phoneme Detection, Phoneme Segmenting.
Parent/guardian questionnaire	Background questionnaire relating to the language history and enabled categorization into appropriate groups
Bilingual only	
Working memory assessment	Forward digit span verbal recall in Welsh
Phonological awareness assessment	Specially designed phonological awareness measures in Welsh – Rhyme Detection, Rhyme Generation, Syllable Detection, Syllable Deletion, Phoneme Detection, Phoneme Segmenting
Welsh language assessment	Prawf Geirfa – Receptive vocabulary assessment, first third administered.

Table 14. Order of test administration for all participants.

Background Questionnaire

Questionnaires were provided to parents or guardians in their language of choice (Welsh/English or both) and were completed either before or during one-to-one testing sessions with the children. Two versions of the questionnaire were designed, one for the parents of children with DS and one for the parents of the TD children. The questions were identical for both versions besides the wording of one question, which was either ‘Does your child have any known disability?’ or ‘Does your child have any disability other than Down

syndrome?'. All parents also received the opportunity to ask questions following completion of the questionnaires and provide additional relevant information about their children if they felt that this would be useful or necessary.

The questionnaires aimed at eliciting demographic information relating to the children including date of birth, gender, hearing status, age of first word, attendance at nursery etc. The second section of the questionnaire related to the child's language background, including questions surrounding home language use, current language exposure, lifetime exposure, response language use as well as a parent report of their child's receptive and expressive language abilities. The information provided from the parents enabled appropriate categorisation into target groups of bilinguals and monolinguals following the information relating to language background. Following this was a question concerning any advice received regarding bilingualism. The final section included two questions relating to SES, which were the highest level of parental/guardian education and the highest parental/guardian occupation. Each SES score was calculated by combining scores for both questions. Parents or guardians were also given the option to leave their contact details if they wished to receive further communication about the research and its outcomes.

Kaufman's Brief Intelligence Test- Second Edition (KBIT-II)

The Kaufman's Brief Intelligence Test (KBIT-II; Kaufman & Kaufman, 2004) is a standardised short assessment of an individual's general intelligence and cognitive development. For the purpose of this research, the non-verbal (henceforth, NV) subtest only was administered to the children as it was deemed more appropriate for the target sample of young children and individuals with DS. Children with DS are often reported to be visually orientated and often have further impairments in expressive language. As a result, the non-verbal subtest was selected to obtain a measure of cognitive development that was as accurate as possible for the populations under study. In addition, potential difficulties with speech intelligibility were foreseen and consequently, NV responses were deemed most appropriate, which this assessment is capable of measuring. The verbal element may not have captured the true abilities of children with DS and may have led to underestimations of the actual cognitive abilities. The KBIT-II was selected as a suitable measure for use with the children in this research as the stimuli used for this task comprises of coloured images selected as responses, which does not rely on an individual's ability to read or name objects. The brevity and bright pictures in the measure is designed to be appealing to children and less intimidating given the testing situation.

The NV subtest in the KBIT-II is a matrices task that is designed to assess an individual's capability to solve new problems by relating prior relationships to a novel example. Within the nonverbal subtest, there are a total of 46 items and all of which have a 5-item multiple-choice answer. Administration time for the whole KBIT-II is 15 minutes, with the NV subtest estimated to require 7-8 minutes to complete dependent on the cognitive ability of each child as the items increase in difficulty as the assessment continues. The subtest is discontinued

after four consecutive incorrect responses. Furthermore, the KBIT-II is reported to have a high level of internal consistency with a coefficient of .93 across all ages and a coefficient of .88 for the nonverbal subtest specifically, which is within accepted standards (Kaufman & Kaufman 2014; Chapter 5). Norms have been designed for this assessment, however, for the purpose of this research raw scores only are reported and utilised as some children did not meet the minimum score required for standardization. This assessment was administered in English, however, as it is a NV measure, it was assumed that this should not cause difficulties in responding to the task for the bilinguals, particularly as all children were reported to have measurable language abilities in both English and Welsh.

Prawf Geirfa

The Prawf Geirfa Cymraeg: Fersiwn 7-11 (The Welsh Vocabulary Test: Version 7-11; Gathercole & Thomas, 2007) is the only standardised Welsh language assessment that is normed for children who are Welsh-English bilinguals. It is an assessment of Welsh receptive language ability and is administered solely through the medium of Welsh. The Prawf Geirfa is available as an electronic and manual version. The manual version was used in this research due to potential technical difficulties with administering the electronic versions as some computer systems have updated which led to the electronic version not being compatible with all required computers. The manual version also allowed additional time for responses where required, in considering the target populations. The assessment involves a manual picture selection task whereby a word is read aloud to the child and they are asked to select which of the four illustrations they believe represents the spoken word to the best of their ability. As this is a NV assessment, it was anticipated that this would be an appropriate measure that would work to visual processing strengths as described previously. There are two initial trials that serve as practice items, followed by 111 items which increase in word length and difficulty as the task progresses.

This measure is currently only designed for use and normed on children age 7-11. Consequently, only the first third of the task (with 37 items) was administered to the children following piloting with children outside of the age range. The raw scores were used in subsequent analyses as opposed to the standardised scores as the children did not fall within the age group to provide normed scores. Therefore, the Prawf-Geirfa was used as stimuli for capturing Welsh language receptive ability as opposed to a standardised test. As this task has been standardised, it was appropriate to use this assessment as the selection of words and pictures have been deemed suitable for bilingual populations in Wales. Prior considerations have been given in terms of item selection and appropriateness for children across various

geographical locations in Wales.² This assessment was delivered in the Welsh session when the children were considered to be in a 'monolingual Welsh mode'. No feedback was provided during the assessment items besides general encouragement to continue.

Digit Span

A forward digit span test was administered to all children in order to ascertain working memory capabilities as prior research has deemed this as playing a fundamental role in language outcomes (Baddeley, 2003). This assessment of short-term memory included single digits from 1-9, which were presented in a randomised order. The researcher read a number sequence to the participant, around the rate of one per second. The number sequence was also presented visually to the children simultaneously. The child was then asked to repeat the sequence in the same order with the wording of "can you say..." followed by the digit sequence. Two trials of each sequence length were presented (i.e. 2 x 2 digits, 2 x 3 digits, 2 x 4 digits, etc.) until the participant was no longer able to recall either sequence within a trial. Prior to the assessment, the children were asked to count to ten to ensure that they recognised and could say all the digits and served as a warm-up to the task.

This task was administered as working memory impairments appear to be apparent in those with DS (as discussed throughout section 3.1.1), and are also known to play a role in PA and vocabulary acquisition. Memory span tasks are also used in order to measure the number of items that a participant is capable of attending and organising as a working unit. This ability will be relied on in some of the following assessments and consequently, a measure of WM will enable an analysis of the association of WM with various aspects of language development that are being tested, including PA. Children were assessed in both languages, although it was anticipated that WM scores would be identical in both.

Clinical Evaluation of Language Fundamentals- Preschool (CELF-P) – Second UK Edition

The CELF-P Second Edition (Semel et al., 1998) is a clinical diagnostic tool used for language assessment which is specifically designed and standardised for pre-school aged children between 3-7 years old. This assessment is designed for individual administration and in a clinical setting, can be used to identify language disorders and areas of weaknesses that interventions can subsequently be based upon. The CELF-P is similar to the CELF-4, however, the administration age of the CELF-4 is for children aged five and over. Therefore, the CELF-P was better suited to identifying early language development as it includes simpler tasks that are more appropriate for younger children. This assessment comprises of 9 sub-tests: Concepts and Following Directions, Word Structure, Expressive Vocabulary, Recalling

² Welsh vocabulary occasionally differs according to the speaker's geographical location. For example, a speaker from South Wales may use the word 'porffor' for the colour 'purple', whereas a speaker from North Wales might use 'piws'.

Sentences, Sentence Structure, Basic Concepts, Recalling Sentences in Context, Word Classes and Phonological Awareness.

Within this measure, there is a four-process model and each of the subtests forms a different level depending on the intentions of the administrator. These are: Identifying the problem and determining eligibility, Describing the nature of the disorder, Evaluating early classroom and literacy fundamentals and Evaluating language and communication in context. For the purpose of this research, the first two levels will be assessed as this provides a Core Language Score, Receptive Language Index, Expressive Language Index, Language Content Index and Language Structure Index which will provide useful information in comparing the groups under study. Analyses of errors made within each subtest are also possible in order to specify difficulties within individuals.

This assessment comprises of receptive and expressive tasks, alongside colourful visual stimuli which again should assist with engaging the children and target visual processing strengths in those with DS. Administration of each sub-test can be adapted to the needs of the child. Two example teaching items to familiarise the child with the questions and the appropriate responses precede the test items in each sub-test. There is no set time limit for this assessment, meaning that adaptations can be made for children requiring more or less time. Standardization of this assessment included more than 1,300 children, with reliability and validity being evaluated for 120 children. Reliability for stability coefficients for test-retest measures when considering all ages is reported at .78 to .94. Reliability coefficients across all ages for internal consistency is also within acceptable standards of between .79 and .97. Internal consistency is also high for children from clinical groups (ASD, hearing disorders and language disorders) with the coefficient alphas between .87 and .97. Consequently, this assessment is regarded as providing appropriate internal consistency and test-retest reliability for measuring the language abilities of children with and without language impairments. As many of the children selected for the research are older than the test criteria, they will not be within standardised scaling scores. Consequently, raw scores, z-scores and age equivalent scores for each subtest will be used in the subsequent analyses which involve the children with and without DS. The subtests used for this thesis are:

Sentence Structure

The first subtest is designed to measure children's ability to interpret sentences presented orally which increase in length and complexity. Four pictures are presented and the child is requested to select which image corresponds to the sentence spoken to indicate that they understand the sentence.

Word Structure

The Word Structure subtest is designed to assess a children's ability to complete a sentence following the appropriate grammatical construction. The correct sentence-completion responses increase in complexity as the subtest continues and the correct response is

indicated by the picture presented for each item. This subtest assessed a child's ability to follow and produce the appropriate morphological rules in English.

Expressive Vocabulary

This subtest is used to measure a child's ability to correctly name the items presented in the pictures presented. The pictures vary in presenting people, objects and actions and the child is asked to say what they can see in the images. The items become more complex and less frequent as the subtest continues.

Concepts and Following Directions

This subtest assesses a child's ability to follow oral commands by correctly interpreting, recalling and undertaking a required action. The child is asked to point to the correct item with the correct characteristics (e.g. size or colour) in the correct sequence.

Basic Concepts

The Basic Concepts subtest is used to measure children's abilities in understanding a range of concepts including information relating to position or location, size and quantity. The child is asked to select which item in an image corresponds to the word spoken and the concepts increase in complexity as the subtest progresses.

Recalling Sentences

This subtest is used to measure a child's ability to remember and repeat sentences that increase in length and complexity throughout the subtest. The child is asked to repeat the sentence in the same order and without altering any of the words. The sentences start as fairly simple to begin with but as the subtest continues additional elements and more complex sentence structures are used.

Word Classes

The final subset that was used was the Word Classes subtest. This subtest is used to evaluate if the child is able to identify and communicate patterns or relationships between items. The subtest begins with three items where two items have a relationship with each other (e.g. their use, where they're found) and later increase to four items with two items being related to each other. The child is first asked to point to which items are related to each other and then express why or how they are related.

Phonological Awareness Tasks

Specially designed PA tasks were produced specifically for the purpose of this research in both Welsh and English. The PA assessments were designed to measure the three levels of PA; syllabic, intrasyllabic and phoneme levels, as these have been noted as the three domains within this metalinguistic skill within these languages. Within each of these levels, there was

one identification and one manipulation task which aimed to identify explicit and implicit abilities within each domain of PA. Each of the six tasks is described in more detail below. All tasks were introduced by two practice items and items were presented verbally alongside visual stimuli. Responses were NV wherever possible in order to overcome intelligibility and production difficulties within the target population (i.e. manual picture selection). The positioning of the correct item was altered across trials, with each position having an equal overall number of correct items.

Assessments were designed to be game-like with coloured illustrations that were age-appropriate and as engaging as possible. The PA measures were adapted according to the recommendations reported by Kennedy and Flynn (2003), in that the length of syllables and phonemes should be restricted to 3 or 4 items due to short term memory impairments which are usually apparent in DS. An upper limit of phonemes or syllables was adhered to wherever possible within these measures as research suggests that children with DS do not often have digit spans of more than 4 items (Cossu et al., 1993). In addition, tasks were designed with the aim that words selected would be likely to be known by young children. In designing the PA tasks, the items included were selected for their high imageability, high frequency and the number of syllables and letters. As this measure was completed in Welsh and English for the bilinguals, matching of both sets of stimuli was required. Subsequently, imageability ratings were obtained for both sets of stimuli using norms provided in studies by Clark and Paivio (2004) and Cortese and Fugett (2004) where items were rated on a 7-point Likert scale by large numbers of participants. Statistical analyses show that there were no significant differences between the imageability scores for the Welsh and English stimuli ($p = .103$), with both also having high mean imageability ratings (Welsh = 6.29; English = 6.44). Some items were used for both the English and Welsh sets of stimuli, however, researchers have identified that imageability scales are capable of being used cross-linguistically as high correlations are reported across languages (Rofes et al., 2017). This is of course dependant on cultural suitability and the availability of semantically equivalent words in the second language, however, as Welsh and English have a large intercultural association (i.e. being used simultaneously in Wales), it should be appropriate to use these measures cross-linguistically. Therefore, the imageability ratings were suitable for use with the Welsh and English stimuli (see appendix item 9 for further statistical analyses relating to imageability, frequency, syllable and letter length.)

Consideration was also given to the frequency of words selected for each stimulus set as more frequent words may be easier to process with some researchers reporting that high-frequency words result in higher scores on PA measures (Hogan et al., 2011). In order to obtain word frequencies in Welsh, the Cronfa Electroneg o Gymraeg (CEG) was used (Ellis et al., 2001) which is a lexical database that compiled a million items in the Welsh language. Translated equivalent words in English were then used as a comparison for the Welsh stimuli, in which there were no significant differences between the two sets of stimuli ($p = .143$) and both had high mean word frequencies (Welsh = 114.5; English = 108.8). In addition to this,

the length of the words in the Welsh and English PA measures also needed controlling for. Research suggests that the length of words has important implications on research outcomes with non-words (Stokes et al., 2006) however, this may also extend to real words, particularly due to memory constraints in DS. Therefore, words were also matched in terms of letter and syllable length (for full statistical output, see appendix item 9).

As noted, PA tasks were restricted to three or four phonemes or syllables where possible due to digit span constraints. It was not possible to control for both letters and phonemes due to differences in orthography between Welsh and English and this is also reported by Spencer and Hanley (2003). The stimuli were matched for syllable number with no significant differences between stimuli language ($p=.143$) and length in letters between the two stimuli sets ($p=0.410$). Due to the fact that the Welsh language is more orthographically transparent with one to one matching of letters and phonemes often being apparent, the English stimuli set had fewer phonemes on average. An example of this is that the English language has many silent letters, which resulted in the Welsh stimuli having more phonemes on average. It would not have been possible to control for both phoneme and letter length and previous research has suggested that matching on letter length when assessing PA in Welsh and English populations is most appropriate (Spencer, 2000; Spencer & Hanley 2003). If the stimuli were matched on phonemes only, this would result in longer English words which may have resulted in a disadvantage towards the monolingual groups, and stimuli too difficult for the target groups. The Welsh words contained more phonemes and so if this resulted in increased difficulty, this factor would result in a disadvantage towards the bilinguals in the Welsh testing only. This should make it more difficult to show any significant bilingual advantage. If it was possible to match the word length on letters and phonemes, any effect would more than likely be greater.

The tasks were also designed in line with Kennedy & Flynn's (2003) recommendations of assessing PA in those with DS, which was to ensure that the tasks selected were not overly complicated in that they relied too heavily on cognitive abilities which would subsequently mask true PA abilities. They acknowledge that NV responses (i.e. pointing) should be used and the use of visual stimuli would lower the processing load by lessening memory constraints where appropriate. Similar recent recommendations have also been made by Lemons et al. (2015) who suggest that adapted measures with populations of children with DS would lead to more accurate outcomes. These recommendations included more explicit instructions, decreased WM load by means of repetitions, limiting verbal language requirements and targeted visual support.

In consideration of the factors identified above, the tasks used for this research are described below. The tasks were ordered in terms of perceived difficulty so that the progression of the tasks was more challenging as the testing continued, beginning with larger units (rhymes and syllables) and finishing with the smallest units (phonemes). Within each section, the difficulty varied within each task in order to keep motivation levels high i.e. difficult tasks all together may lower motivation and engagement. The final version of the

stimuli contained 73 items in total, with 2 practice items preceding 10 test items in each of the 6 sections, with the exception of the syllable manipulation task which had one additional practice item when syllable location reversed, in a similar design to Kennedy and Flynn (2003). Examples of each task and the full list of stimuli are provided in the appendix (items 7 and 8). The tasks used to measure PA are described individually below.

Rhyme Detection: The rhyme detection task was designed to measure the ability to identify sets of rhyming word pairs. Testing involved the presentation of a target item, followed by a choice of three items below, one of which rhymed with the target word. Each item was pointed to and named by the administrator, before asking the participant which item ended in the same sound as the target item. For example, the children were asked which item ended with the same sound as 'bat', with the options of 'man,' 'hat' and 'tie'. This task was designed similarly to that used by Kennedy and Flynn (2003) and Cupples and Iacono (2000), however, instead of a choice of two items, this was increased to three for a lower chance rate of selecting the correct item. Positive reinforcement and teaching of the correct responses were provided for the first two trial items only and ten test items followed the practice phase.

Rhyme Generation: Following the rhyme detection task was rhyme generation. Participants were presented with a pair of matching items and told that they should try and think of a word that ended in the same sound as the target items to create matching pairs. This was similar to that used by Boudreau (2002) who also assessed PA in DS. Participants were also told that they could make up a word that rhymed if they could not think of a real word. Each item was shown in picture form and named by the administrator. During the two practice items, if a participant could not think of a rhyming word, the administrator modelled examples words that rhymed with the target word to ensure that the participant understood the aim of the task. If a participant failed to generate a rhyme for four consecutive trials after the practice items, the task was discontinued.

Syllable Detection: This task was designed to measure the participant's ability to segment syllables within words. Similar measures have been used by Swank and Catts (1994) and Boudreau (2002) with participants being required to clap or tap out the syllables of a target word. Words used ranged between two and four syllables and were presented visually and verbally to the participant. The administrator modelled the responses required using the participant's or administrator's name to familiarise them with the task before beginning the further practice and test items. Participants were first asked to clap out each syllable, however, if the participants struggled with this they were encouraged to tap on the table. This was selected as it is a NV measure of syllable judgement.

Syllable Elision: Participants were again presented with picture stimuli and the examiner named the target item. For this task, participants were asked to say the word aloud with

either the first syllable or last syllable missing (first 5 items missing the first and final 5 items missing the last syllable). For example, the word 'doctor' (/ˈdɒktə/) would become 'doc' (/ˈdɒk/) in the final syllable deletion task. This task was based on the measure used by Verucci et al. (2006), which again targeted participants with DS. Similarly to the syllable detection task described above, this was first practised with either the participant's or the administrator's name as to introduce the task. There were two practice items following this and one additional practice item when the location of the deleted syllable was reversed. If the participant failed to respond or responded incorrectly during the practice items, the correct response was modelled by the administrator.

Phoneme Detection: The phoneme detection task was designed similarly to the rhyme detection measure with one initial target item and a picture selection response with three possible items below. The target item was named firstly and the following items were then pointed to and named by the administrator. This task was adapted from the measure in Cupples and Iacono's (2000) study where participants were asked to identify which item began with the same initial sound as the target item. For example, the children were asked which item began with the same sound as 'snake' (/sneɪk/), with the options of 'tree' (/tri:/), 'crab' (/kræb/) and 'sun' (/sʌn/). The number of possible items was increased from two to three again in order to reduce the chance rate. As with all other tasks, two initial practice items with feedback preceded the ten test items.

Phoneme Segmenting: For the final task, participants were presented with target items singularly with pictorial representations. The target item was spoken aloud by the administrator and participants were asked to produce all the individual sounds they could hear in a given target word. Cupples and Iacono (2000) also used this task to measure phonological awareness at the phoneme level within participants with DS. Prompts were also given in this task such as 'what's the first sound in dog?' for participants who did not initially respond. The correct response was modelled by the administrator for the first two trial items and positive reinforcement was also given for correct responses in this phase. Following this, participants were asked to repeat this for the next ten test items.

4.1.3 Participants

After piloting, the full sample included a total of 77 children (M = 31, F = 46). All these children met the initial inclusion criteria described above and participated in the research. The total age range of the children across all groups was between 2;11-16;9. The children with DS were aged between 5;5-16;9 and the TD children were aged between 2;11-7;10. All parents in the DS groups (n = 31) confirmed a clinical diagnosis of DS (Mosaic DS, n = 1) and all children in the TD groups (n = 46) confirmed that their child had no diagnosed or suspected disability impacting on language development or any suspected or diagnosed language impairment. The vast majority of children in the DS groups attended a mainstream school (n = 25), with

the further 4 being educated in a SEN school. Two parents reported that their child attended two schools where they were educated in a SEN school for 3 days a week and a local mainstream school for the remaining 2 days. Of those with DS attending mainstream schools, 5 were included in a resource unit for those with SEN, with the remaining 20 children with DS being included in a full mainstream setting with the majority having one-to-one support. All TD children attended mainstream schools and nurseries.

Four children with DS were also reported to either have a clinical diagnosis ($n = 3$) or a suspected diagnosis of ASD ($n = 1$). These children are excluded in the main analysis, however detailed information regarding these children is reported separately in Chapter 7. All parents reported that all children had no greater than mild hearing loss, and no further language impairments in the DS groups were reported besides general language delay and one reported as having dysarthria. Three children were also reported to have ADHD (DS, $n = 2$; TD, $n = 1$) and some others reported general health concerns frequently reported in DS such as hypotonia and hyperthyroidism, however, as these do not impact on language these children were included in the overall sample. An additional 5 children (DS, $n = 2$; TD, $n = 3$) were removed from subsequent analyses due to being trilingual, bilingual in languages other than Welsh and English, Welsh monolingual, under three or not completing enough of the assessments. The final remaining overall sample consists of 68 children. An overview of the participants included in the overall sample is presented in Table 15.³ Further individual participant characteristics are provided in the appendix, item 9.

³ Missing information for 5 TDB children due to non-returned questionnaires for SES and L2 lifetime exposure.

		DSM n = 15	DSB n = 10	TDM n = 18	TDB n = 25
CA (Months)	Mean	114.9	114.2	53.1	51.0
	SD	35.6	37.7	16.2	16.3
NMVA (Raw)	Mean	10.9	9.6	11.6	11.8
	SD	5.2	6.5	5.6	4.7
WM (Digit span)	Mean	2.9	2.8	3.2	3.3
	SD	0.9	0.8	0.8	0.6
SES*	Mean	10.47	10.9	11.3	10.9
	SD	2.4	1.7	2.9	2.0
L2 Exposure* (Lifetime %)	Mean	2.14	30	8.0	29.0
	SD	3.56	11.55	8.9	15.6

Table 15. Group means and standard deviations. *Note:* CA = Chronological Age, NVMA = Non-verbal Mental Age, SES = Parental Socioeconomic Status, WM = Working Memory, L2 Exposure = Lifetime exposure to second language. *Missing data (n=5)

To ensure matching criteria were met, a one-way multivariate analysis of variance (MANOVA) was used to identify any significant differences between the four groups on the descriptive variables noted above. An overall significant main effect of group was found $F(15,174) = 11.55$, $p < 0.0005$, Wilk's $\lambda = 0.13$, partial $\eta^2 = 0.5$. As expected, significant univariate between-group effects were found for CA, $F(3,60) = 27.57$, $p < 0.0005$, partial $\eta^2 = 0.58$; and L2 Exposure, $F(3,60) = 22.59$, $p < 0.0005$, partial $\eta^2 = 0.53$. Follow up multiple comparisons using Tukey's post-hoc analysis showed that the significant group differences are between the two DS groups and both TD groups ($p < .0005$). Consequent to matching on NVMA, the children with DS were chronologically older than the children in the TD groups as anticipated. No significant differences are found on chronological age between the two TD groups or between the two groups of children with DS.

Significant differences are also found between the two groups of bilinguals and the two groups of monolinguals on L2 exposure ($p < .0005$). This was again expected and confirmed the group status of the children with the bilingual groups receiving significantly more exposure to an L2 (Welsh or English) than the monolinguals. Groups were also matched on SES with no significant main effect of group ($p = 0.85$). The NV mental age of the children ranged from $<4;0 - 8;3$ with raw scores between 1-26 (mean = 11.26, SD = 5.72). No significant group differences were found on this variable ($p = 0.67$), as expected. Working memory scores ranged from 2-6 (mean = 3.15, SD = 0.85) again with no significant differences between groups ($p = 0.28$). Further information about the children's language backgrounds, degree of learning

difficulty and the reported expressive and receptive language abilities according to their parents is presented in Table 16 below.⁴

		DSM n = 15	DSB n = 10	TDM n = 18	TDB n = 25
Home Language	Welsh	0%	0%	0%	16.7%
	English	100%	20%	94.5%	22.2%
	Both	0%	80%	5.5%	61.1%
School Language	Welsh	0%	70%	0%	100%
	English	100%	10%	94.5%	0%
	Both	0%	20%	5.5%	0%
Nursery Language	Welsh	6.6%	80%	5.5%	72.2%
	English	93.4%	10%	72.3%	22.2%
	Both	0%	10%	22.2%	5.6%
Learning Difficulty	None	0%	0%	100%	100%
	Moderate	86.6%	80%	0%	0%
	Severe	13.3%	20%	0%	0%
Parent report	Expressive	3.1	2.55	4.56	4.19
	Receptive	3.7	3.5	4.64	4.44

Table 16. Group means for language background and degree of learning difficulty. *Note:* Group percentages for the degree of learning difficulty and language background for home, school and nursery language. Parental reports for receptive and expressive language average scores from a 1-5 Likert scale where 1 = Poor, 5 = Excellent. *Missing data (n=5)

For the two bilingual groups, the children were required to be exposed to both Welsh and English before 36 months (3-years old) meaning that all children were classified as simultaneous bilinguals according to Paradis et al.'s (2011) classification. All were required to have been consistently exposed to both languages from this age onwards, however, the majority were exposed to Welsh and English from birth or soon after. Of those classified as bilinguals, 78.9% reported having Welsh exposure at home, compared to 3.0% of the monolingual children. Bilingual status was determined by measures of current and lifetime

⁴ Missing information for 5 TDB children due to non-returned questionnaires.

exposure to Welsh and English which were obtained via parent report (with a cut off exposure criteria being above 15% for inclusion in bilingual groups). As English is the majority community language, all children received substantial input to English, with the bilinguals also being exposed to Welsh at home, school or within both contexts. Language dominance in the bilinguals was determined by the language that the participant had received the most lifetime and current input to as estimated by parent report. Of the bilingual children that returned the questionnaires, 42.4% were regarded as English dominant, 39.4% Welsh dominant and the remaining 18.2% being considered balanced bilinguals.

Typically developing children were recruited through new and existing networks with Welsh and English-medium schools and nurseries in South and North Wales who were willing to assist with the project. Recruitment of children with DS was conducted through collaboration with the Down Syndrome Association and Learning Disability Wales who published information on their websites, social media and newsletters as a means of directly communicating information regarding the research to their databases of families who have a child with DS. Five children with DS were also recruited through a mainstream school in South Wales and a family who had existing networks with the researcher and agreed to be involved with the project.

Finally, in terms of the number of sessions that the children attended for the purpose of this study, these ranged between 1-4 and were adapted according to the age, attention and engagement of each child. All bilinguals were assessed in both English and Welsh and consequently had at least two sessions. These were undertaken on separate days wherever possible as described above. Overall, the DS groups had an average number of sittings of 1.93, and the TD groups had an average of 2.44 sittings (due to the fact that the TD children were younger). Subsequent chapters contain sub-samples taken from the overall participant sample described in this chapter. Explanations as to which participants are included within each chapter are presented within each of the chapters.

Chapter 5: Expressive and Receptive Language Abilities of Welsh-English Bilingual Children with Down Syndrome⁵

This chapter will present the findings in relation to the first and second research questions, which were “What are the language abilities of children with Down syndrome who are bilingual?” and “How does the language development compare for children with Down syndrome who are bilingual versus monolingual?” Results relating to core, expressive and receptive language abilities for bilinguals with Down syndrome will be presented and discussed.

5.1 Introduction

Researchers have highlighted that language impairments pose a substantial challenge for children and adults with DS as discussed in Chapter 3. These difficulties extend through all aspects of language including vocabulary, and grammar. The language impairments that are often observed in those with DS appear to be specifically impaired, meaning that these difficulties are not completely accounted for by general intellectual impairments. As a result, there appears to be something specific surrounding language that children and adults with DS find particularly challenging, especially when it comes to expressive language and expressive morphosyntax. Within bilingual communities, such as Wales, a logical progression is to question how the addition of a second language impacts on children with these particular profiles of language impairments. As discussed earlier, bilingualism may be essential within some families and communities. For other families, parents may make an explicit choice regarding language exposure, particularly within the linguistic context of Wales where all child is said to have the right to become bilingual. At the same time, bilinguals are a growing population with over half the world being bilingual or multilingual within increasingly multicultural and multilingual societies. Consequently, the need to identify the role of bilingualism for individuals with a DD becomes even more necessary and relevant.

The purpose of this research was to investigate the linguistic development of bilingual children with DS. This is firstly presented by comparing children who were individually matched for their raw non-verbal mental age (NVMA) scores to the target group of Welsh-English bilingual children with DS whilst also controlling for working memory (WM) and socioeconomic status (SES). Results are then presented for a larger sample who are matched at group level on NVMA, WM and SES. The three other comparison groups are children with DS who are English monolinguals (DSM), TD Welsh-English bilinguals (TDB) and TD English monolinguals (TDM). The aim was to create a language profile of bilingual children with DS (DSB) by providing a comprehensive overview of language development in Welsh and English

⁵ A modified version of this Chapter is under review in the *Journal Communication Disorders* entitled ‘Language Profiles of Welsh-English Bilingual Children with Down Syndrome.’

and contrasting this profile with the other populations under study. More specifically, the focus of this chapter is drawn to core, receptive and expressive language abilities in English. In addition, Welsh receptive language abilities on the Prawf Geirfa will be presented for the two bilingual groups. Factors associated with language abilities for each group will then be explored. Finally, analyses of the advice that parents receive concerning bilingualism for the children with DS and those who were TD are presented for bilinguals and monolinguals.

5.1.1 Language Development in Down Syndrome

Down syndrome (DS) is the most common chromosomal genetic disability which impacts health, general intellectual development and very frequently, language development. Researchers interested in language development in children and adults with DS have identified a particular profile of language, in which some elements of language appear to be specifically impaired i.e. more impaired than expected given general cognitive development and abilities in other aspects of language such as vocabulary (see Abbeduto et al., 2007; Ypsilanti & Grouios, 2008 for a review). In addition, receptive abilities are frequently reported to be greater than expressive language abilities, with language comprehension often developing at a quicker pace than expressive abilities (Chapman, 1997). Intelligibility is also often reduced in the speech of children and adults with DS. Furthermore, many individuals with DS are reported to continue to use inappropriate phonological processes for a longer period of time compared to TD children (Dodd, 1972; Kumin, 2006).

Some elements of language which have been noted to be specifically impaired such as grammatical development. For example, Chapman et al. (1998) investigated the language development of children with DS between 5;6 and 20;6 in comparison to developmentally matched TD children on their grammatical morphology and MLU. They reported that the DS groups produced more simplistic utterances, had a lower MLU and used a smaller range of words in comparison to the TD children at all four age groups studied. Syntactic development did, however, continue across the age groups with individuals in the older groups performing better, suggesting that there is no 'critical period' for syntactic development in those with DS and that there was no evidence of a syntactic ceiling. Grammatical impairments and more specifically expressive morphosyntax have also been noted to be specifically impaired in those with DS (Chapman et al., 1998).

Further impairments have been documented within the syntactic development for those with DS such as difficulties in the interpretation of personal and reflexive pronouns (Perovic, 2002; Perovic, 2006; Sanoudaki & Varlokosta, 2014). For example, Perovic (2006) compared four individuals with DS (aged 17-21) to two TD control groups (matched for either receptive abilities or chronological age). It was reported that the individuals with DS had marked difficulties in interpreting reflexive pronouns. This was not the case for the control conditions, in which the individuals with DS had much higher performance. It was proposed that this is evidence that syntactic development in DS is not only delayed but develops differently to TD children. Typically developing children usually display difficulties in the

interpretation of personal pronouns, not reflexives (commonly referred to as the delay of principle B effect).

In terms of morphosyntax, Laws and Bishop (2003) reported on impairments with grammar production whereby individuals with DS made more omissions of regular tense-related morphemes than a control group of children with SLI. As a result of studies such as these, it has been proposed that this consequently supports the proposal of modularity of language (i.e. not a single general linguistic processing system) as not all aspects of language are impaired to the same extent (Perovic, 2006). Given these findings, it is evident that individuals with DS have substantial difficulties with language which continue to persist in adolescence and adulthood. Identifying how this language phenotype corresponds in bilinguals with DS will provide a novel insight into language development in DS.

5.1.2 Bilingual Development and Developmental Disabilities

The need to understand the impact of bilingualism within increasingly multicultural societies becomes more and more meaningful. As a result of the increased role of bilingualism in societies, the decision to raise children bilingually, or in many cases the necessity of bilingualism (Bialystok et al., 2012) is even more pertinent. Within the last decade, there has been a surge of literature documenting bilingual language development in children with a DD. A number of studies now exist that document language acquisition in bilingual children with an Autism Spectrum Disorder (ASD; Drysdale et al., 2015; Valicenti-McDermott et al., 2013) and Developmental Language Disorder (DLD; Paradis, 2010; Paradis et al., 2003). This literature provides a favourable view of bilingualism within these disorders as researchers have found no negative impact of bilingualism on a range of aspects and across various language combinations.

For example, a longitudinal study conducted by Valicenti-McDermott et al. (2013) investigated very early language development for monolingual and bilingual children with ASD. Parental reports and observations were used to assess babbling, vocabulary sizes, word combinations and vocalizations. The authors found that these early language abilities of the children were comparable between the two groups, with no slower development or reduced communicative efforts for any of the measures. The authors concluded that exposure to an additional language does not lead to any additional delays or difficulties with language development during the early years. In contrast, the bilingual group produced significantly more 'cooing' than the monolinguals, suggesting that if anything, the addition of a second language promoted this aspect of development.

When it comes to DLD, researchers have made similar conclusions. Paradis (2007) assessed English-French bilinguals with DLD on the use of several different grammatical morphemes in comparison to matched English and French monolinguals with DLD. As incorrect morpheme use is a clinical marker for DLD this area was targeted to investigate if the bilingual children had any further difficulties in this area. Results showed that the bilingual children were as accurate at using the correct grammatical morphemes as the monolinguals

with DLD in both languages. This research again supports the suggestion that the addition of a second language did not lead to any additional difficulties for these children. Several other studies have supported this view across various DDs (Kay-Raining Bird et al., 2016) which all appear to support bilingualism, with no apparent evidence to suggest that exposure to additional languages results in hindrance to development or language outcomes.

Finally, a recent review of multilingualism in neurodevelopmental disabilities also found that there were no adverse outcomes on a number of language measures and other aspects of functioning across the studies that were reviewed (Uljarević et al., 2016). A number of linguistic features were documented in this review including vocabulary, lexical retrieval, grammar, phonological awareness and cognitive functioning. Positive effects were, however, found for communication and social functioning in one study (Hambly & Fombonne, 2012) whereby the bilinguals with ASD had enhanced interpersonal skills as measured on a standardised assessment (The Vineland Adaptive Behaviour Scales - Second Edition; Sparrow, Cicchetti & Balla, 2005). The authors do also note that there is a low number of methodologically high-quality studies within this area. It is also worth highlighting that no study has documented any negative outcomes for bilinguals with DDs when compared to matched monolinguals with the same DD.

Of concern is the fact that research investigating the recommendations given to families surrounding bilingualism does not coincide with these research findings. Clinicians are often the primary source of support for many families who have a child with a DD but it appears that these families have previously been advised to limit language to a single language in order to promote language development and avoid confusion with learning two languages. In terms of parents of children with ASD, it has been reported that clinicians believe that bilingualism would be too much of a challenge for a child with ASD and that parents should instead adopt a monolingual environment (Kay-Raining Bird et al. 2012; Kremer-Sadlik, 2005). As a direct result, this also leads parents to report that they feel that two languages would be too confusing for their child, and may delay language development in both the first and second language (Kay-Raining Bird et al., 2012; Yu, 2013).

An international review was conducted recently which investigated policies and practices related to bilingualism in children with a DD (Pesco et al., 2016). In this review, practitioners from Canada, the US, the UK and the Netherlands provided information regarding their views, experiences and practice relating to working with bilingual clients. The main finding of this report was that the majority of the sites had policies that explicitly referenced the inclusion of students with a DD in a bilingual setting, however, in practice, there was some variability in the opportunities that actually existed. For example, not all locations had SEN schools which would accommodate bilingual children with a DD in this recent review. Practitioner's views towards bilingualism seem to be more positive than previously reported, however, it appears that practice may not always follow suit.

5.1.3 Bilingualism in Down Syndrome

Currently, research assessing bilingual language development in children with DS specifically is very limited, with only one empirical group study having directly assessed the expressive and receptive language abilities within this population (Kay-Raining Bird et al., 2005). Research in this field also exists in the form of early case studies (Vallar & Papagno, 1993; Woll & Grove, 1996). These report that there is no evidence to support the suggestion that learning a second language would result in any further difficulties. Kay-Raining Bird and colleagues recruited 8 English dominant bilingual children with DS (English-French bilinguals $n = 7$, English-Cree bilingual, $n = 1$) and assessed their language abilities by obtaining language samples as well as employing standardised language assessments. The group was matched on developmental age to monolinguals with DS ($n=14$), bilingual TD children ($n=11$) and monolingual TD children ($n=18$). At the time of the research, the children with DS ranged between 2;7 and 11;5 years old (mean = 6;7) which corresponded with developmental ages between 1;8 and 3;11 (mean = 2;9).

Results from this study showed that although the typical language profiles were found for the children with DS compared to the TD children, whereby significant differences were reported for MLU and other expressive language measures. In comparing the two groups of children with DS, no significant differences were found on any of the measures, suggesting that the bilingual children were developing comparable abilities in English to the monolinguals with DS. It is noted that there was a large range of individual variation, both in terms of language abilities and the degree of bilingualism. The authors also report that caution should be taken in interpreting the results due to the relatively small sample size. Additionally, the bilingual DS group had a higher chronological and developmental age overall, though these were not statistically different across groups.

In the case study designs which were employed in earlier research, these also documented bilingual and trilingual success for individuals with DS (Vallar & Papagno, 1993; Woll & Grove, 1996). One of these studies also considered multimodality as a form of bilingualism in a study of English-BSL twins (Woll & Grove, 1996) who were reported to have measurable abilities in both BSL and English although both had a preference to use English. Furthermore, longitudinal parent-report measures have been used to indirectly assess vocabulary sizes in French/English bilingual children with DS (Trudeau et al., 2011). This research also reported a large range of variability in L2 abilities, however, again supported the view that the addition of a second language did not impair the development of the first language (English).

One final study was conducted by Cleave and colleagues (2014) which specifically investigated the morphosyntactic abilities in bilinguals and monolinguals with DS in comparison to developmentally matched TD children in a receptive syntactic bootstrapping task. The findings showed marked difficulties for the DS groups compared to the TD groups as expected given that children with DS have been reported previously to have difficulties with syntactic development. In comparing the two DS groups there were no significant

differences found on performance, suggesting that the bilinguals did not show any further impairments for this domain.

Although the number of studies conducted within this area of research is increasing, there is still only very limited information available which documents the language abilities of bilingual children with DS, particularly in comparison to the available research on bilingualism in other DDs as discussed above. In addition, a number of the studies mentioned above have overlapping samples of English-French bilinguals with DS and consequently, the findings of these studies are constrained to a specific population in Canada. Nevertheless, converging evidence across a range of DDs now suggest that children with language learning difficulties do not experience any additional delays or impairments as a result of bilingualism. Crucially, no study has documented any negative outcomes on development in comparing a monolingual control group with the same DD.

In contrast to this emerging evidence, research assessing the professional recommendations given to parents of children with a DD does not seem to concur with these findings. Researchers have reported that parents of a child with a DD have previously been advised to limit language input to a single language (usually that of the community) with the assumption that this would lead to better language outcomes. This has been reported for parents who only have very limited proficiency in the community language (Kay-Raining Bird, Lamond, & Holden, 2012; Kremer-Sadlik, 2005). Additionally, it has been noted that these recommendations may also impact on these family's comfort in communicating in a non-native language and the challenges that emerge as a result (Hampton et al., 2017).

Hudry et al. (2017) also reported that restricting language input to a language in which the parents are less proficient in may inadvertently reduce the quality and quantity of language that the children subsequently receive. More specifically, it was reported that parents produced shorter and less fluent interactions with their children who had ASD and English as an additional language. The authors also expressed their concerns regarding the social development of these children, which is known to be particularly compromised in ASD, to begin with. In children with ASD, the importance of early caregiver-child interactions (Siller & Sigman., 2002) and the role of linguistic input (Bang & Nadig., 2015) have been documented as important factors to language success. The impact of the advice received by parents consequently has many implications and potential repercussions if practitioners do not provide evidence-informed recommendations for parents of a child with a DD.

5.1.4 Predictors of Language Outcomes in Bilinguals with Down Syndrome

Quantity, quality and timing of exposure are three of the important influences on bilingual language development that have been identified in TD children (Hammer et al., 2012; Thordardottir, 2017). Questions are often raised as to how much exposure is enough or adequate to ensure development in both languages. Numerous studies have now investigated the role of input in TD children and these studies generally report that vocabulary sizes are closely associated with the quantity of input in a bilinguals two languages

(Gathercole & Thomas., 2009). Furthermore, researchers generally report that the current amount of exposure or the recency of exposure is a stronger predictor of language ability than exposure over the lifetime (Cohen, 2016; Thordardottir, 2017).

The role of input in bilingual children with DDs has been much less extensively explored. Hambly and Fombonne (2014) investigated the role of expressive language abilities and the quantity of input in bilingual children with ASD. Although a large proportion of the children were non-verbal at the time of their study (10 out of 33), results suggested that current exposure in the children's dominant language accounted for above 60% of expressive vocabulary scores. In contrast, lifetime exposure did not predict expressive vocabulary sizes. This provides some evidence that current exposure plays a significant role in both TD bilinguals and bilinguals with ASD. Preliminary evidence has also been reported in a study of four bilingual children with DS (Feltmate & Bird., 2008b). In these French-English bilinguals, no consistent effect of language status was found, however, the variability observed in English compared to French language ability was accounted for on measures of current language input. This provides a further suggestion that the role of current exposure is influential in the language abilities of bilinguals with DS as well, although caution is warranted due to the very small sample size.

In researching bilingualism and the role of current and lifetime exposure, it is essential to assess both languages of a bilingual speaker, although many neglect to do so by only assessing the dominant language or only rely on parent report measures as opposed to objectively assessing language abilities. Additionally, many further predictor variables should be considered in relation to language outcomes in bilingual and monolingual children, for example, the role of non-verbal mental age (NVMA), chronological age (CA) and working memory (WM). The role of these variables is potentially more complex and multifaceted when it comes to children with a DD, however, to date, there is only limited information about the role of these factors in bilinguals with ASD and almost no available information in bilinguals with DS. As those with a DD and DS have a specific profile of strengths and weaknesses in their development, understanding how these factors play a role in bilingual language acquisition will result in a more comprehensive understanding of the underlying mechanisms associated with bilingual exposure in those with DS.

5.1.5 Current Study

Limited information is currently available concerning bilingual language development in DS specifically. This research aims to increase understanding of the capacity of children with DS to acquire two languages. Furthermore, the majority of information concerning this population has predominantly explored English-French bilinguals dominant in the majority community language (English). Currently, only one empirical group study exists which has assessed general language abilities in bilinguals with DS. The aim of the present research is to determine the language abilities of Welsh-English bilingual children with DS in comparison to developmentally matched monolinguals with DS as well as similarly matched TD bilinguals

and monolinguals. Although Welsh is considered a minority language, provisions exist for all children to become bilingual through the Welsh-medium educational system and immersive nurseries. Furthermore, every child in Wales is said to have the right to learn the two official languages. These are issues of growing clinical relevance and importance due to the increase in multilingual societies and the drive towards inclusivity for those with a DD. More specifically, the following research questions are addressed:

- I. What are the language abilities of bilingual children with Down syndrome compared to monolinguals with Down syndrome?
- II. How does bilingual acquisition in children with Down syndrome compare to typical bilingual language acquisition?
- III. Do parents of children with Down syndrome and typically developing children in Wales receive any advice regarding bilingual language development?
- IV. What is the impact of age, socioeconomic status, working memory, non-verbal cognitive abilities and amount of language exposure in typically developing children and children with Down syndrome?

5.2 Method

Detailed information relating to research design, procedure, assessments and further participant information is provided in Chapter 4. In summary, one-to-one sessions were conducted with the participants on a range of cognitive and linguistic assessments in order to obtain a comprehensive overview of the children's language abilities. Parents also completed background questionnaires comprising of three sections; information about their child, language background and information about themselves. Assessments were completed in a pre-defined order and bilinguals were assessed in both of their languages; Welsh and English. Measures of NVMA, working memory, core, receptive and expressive language were obtained using the KBIT-II (Kaufman & Kaufman, 2004), CELF-P Second Edition (Semel, Wiig, & Secord, 1998), *Prawf Geirfa Cymraeg: Fersiwn 7-11* (The Welsh Vocabulary Test: Version 7-11; Gathercole & Thomas, 2007) and a Forward Digit Span task. Full ethical approval was obtained from the University's departmental ethical review board prior to data collection. Informed consent was provided by both parents/caregivers and schools.

5.2.1 Participants

The participants and results sections are presented in two parts. Firstly, children from the target group of bilinguals with DS were identified and then individually matched by ± 2 NVMA raw score points to one child in each of the other three groups; Down syndrome monolingual (DSM), typically developing bilingual (TDB), and typically developing monolingual (TDM). The second section includes all children who met the inclusion criteria and who completed the assessments. In this sample, participants are not matched individually, but at matched on

NVMA at the group level. Presenting both individually matched participants and the results of the matched groups provide both a methodologically stringent sample and a larger sample with greater statistical power. Further individual participant characteristics are provided in the appendix, item 9.

Individually Matched Sample

An initial cohort of 77 children was recruited for inclusion for this research specifically and participated in the language assessment sessions. Of these, there were 14 Welsh-English bilingual children with DS. Three were excluded due to having a dual diagnosis of ASD and DS and one additional participant was excluded for being outside of the target age range of 5-14. The remaining 10 children in the target DSB group were subsequently individually matched by ± 2 NVMA raw score points to one child in each of the other groups, giving a total sub-sample of 40 children. This led to a four group 2 by 2 matched design. Analyses confirmed that the groups were also matched overall for NVMA ($p = .972$). Children with DS were required to be between the ages of 5-14 (mean = 9;6) at the time of data collection in order to capture children with enough receptive and productive vocabulary who were still acquiring language. As the TD children were matched on NVMA, the TD participants were chronologically younger than the participants in the two DS groups who were aged between 3-7 years of age (mean = 4;2). As the participants in each group were individually matched on their developmental level, both of the TD groups were significantly younger ($p < .05$). The two groups of children with DS were statistically matched on chronological age ($p = .890$) as were the two groups of TD children ($p = .907$). Group matching for all four groups was also achieved for parental socioeconomic status (SES; $p = .790$). Working memory scores were also comparable across groups with no significant differences found on this measure either ($p = .884$).

Analyses of the children's cognitive profiles as measured using the non-verbal matrices subtest of the KBIT-II (Kaufman & Kaufman, 2004) showed that none of the TD children displayed any evidence of a learning disability. The two groups of children with DS were also comparable for the degree of cognitive impairments with 20% of each group displaying severe learning difficulties and the remaining 80% with moderate difficulties. Gender distribution was also comparable between the four groups with no significant difference found ($p = .662$). In order to ensure that the Welsh-English bilinguals received more input to Welsh than the monolingual groups, analyses were conducted using information obtained from the parental questionnaires. These confirmed the validity of the bilingual status as both bilingual groups had significantly more current and lifetime exposure to Welsh ($p < .05$) than the two monolingual groups. A summary of the group characteristics is presented in Table 17.⁶

⁶ Mean scores are reported with standard deviations in parenthesis. Non-verbal IQ represent raw scores on the non-verbal matrices subtest of the KBIT-II (Kaufman & Kaufman, 2014). Socioeconomic status was obtained via

	Down Syndrome		Typically Developing	
	Bilingual (n=10)	Monolingual (n=10)	Bilingual (n=10)	Monolingual (n=10)
Age in months ^a	114.2 (37.7)	112 (31.9)	51.6 (17.4)	50.7 (16.5)
Non-verbal IQ	9.6 (6.5)	10.7 (6.3)	10 (6.2)	9.5 (6.0)
SES	10.9 (1.7)	9.9 (2.6)	10.8 (2.0)	10.6 (3.1)
Current Welsh exposure in % ^b	49.1 (24.9)	5.01 (9.4)	57 (21.6)	12.8 (14.9)
Lifetime Welsh exposure in % ^b	50 (24.0)	2.79 (4.4)	45.8 (24.1)	8.3 (9.9)

Table 17. Group characteristics for individually matched participants.

Group Matched Sample

The original sample was the same 77 children who participated in the research as described above and in Chapter 4. As opposed to individually matching the participants across groups, all the children who met the inclusion criteria were included in the group matched sample consisting of 65 children. From the original sample, 12 children were removed for the following reasons, a diagnosis or suspected diagnosis of ASD ($n = 4$), trilingual ($n = 1$), English-Greek bilingual ($n = 1$), Welsh monolingual ($n = 1$) and not completing enough assessments ($n = 5$). The sample of 65 participants were again distributed across the same 4 groups, DSB ($n = 10$), DSM ($n = 15$), TDB ($n = 22$) and TDM ($n = 18$). The age range for the participants with DS were between 5;5-16;9 (mean = 9;6, SD = 35.67) and the TD children ranged between 2;11 and 7;10 (mean = 4;3, SD = 15.75). As for the individually matched sample, both of the TD groups were significantly younger ($p < .05$) than the DS groups as expected due to matching at group level for NVMA.

The DS groups were statistically matched on chronological age ($p = .965$) as were the two groups of TD children ($p = .377$). Statistical analyses confirmed that the groups did not differ on NVMA ($p = .755$), SES ($p = .714$), gender ($p = .775$) or WM ($p = .336$). Analyses again confirmed the validity of bilingual status for the bilingual groups with both having significantly more current exposure to Welsh ($p < .001$) and lifetime exposure to Welsh ($p < .001$) than the monolingual groups. Finally, analyses of the cognitive profiles of the participants obtained from the KBIT-II again confirmed no evidence of a learning disability in any of the TD participants. For the DSB group, 20% displayed severe learning difficulties with the remaining 80% showing moderate difficulties. This was similar to the DSM group where 13.33% had

parent report in terms of parental education and occupation (scale from 2-14). ^a Indicates a between-group effect of diagnosis with $p < .05$. ^b Indicates a between-group effect of language status with $p < .05$.

severe learning difficulties and 86.77% with moderate learning difficulties. The sample used for the group matched analyses are provided in table 18.⁷

	Down Syndrome		Typically Developing	
	Bilingual (n=10)	Monolingual (n=15)	Bilingual (n=22)	Monolingual (n=18)
Age in months ^a	114.2 (37.7)	114.9 (35.6)	49.2 (16.1)	53.1 (16.2)
Age of first word in months ^a	24.9 (2.27)	21.7 (11.2)	12.0 (3.2)	10.8 (2.6)
Non-verbal IQ	9.6 (6.5)	10.9 (6.3)	11.2 (6.2)	11.6 (5.6)
SES	10.9 (1.7)	10.5 (2.4)	11.1 (1.9)	11.3 (2.9)
Current Welsh exposure in % ^b	49.1 (24.9)	3.5 (7.9)	48.6 (21.1)	8.2 (10.4)
Lifetime Welsh exposure in % ^b	50 (24.0)	2.14 (3.6)	42.1 (24.12)	8.0 (8.9)

Table 18. Group characteristics for the group matched participants

5.2.2 Recommendations Given to Parents Regarding Bilingualism

Answers provided from the parental questionnaire indicated whether or not parents/carers of the participants had received any professional recommendations in favour of or against bilingualism. Of the children in the DSB group, the majority reported that they did not receive any information or advice surrounding bilingualism (60%), two received positive support for bilingualism (20%), one received advice to drop the additional language and concentrate on English only (10%). The final respondent did not respond.

Of the 15 participants in the DSM group, the majority responded that they did not receive any information about bilingualism (46.7%). Four reported that they did not see it as applicable to their circumstances (26.7%), two were advised to stop speaking a second language or not to introduce a second language (13.3%), one was told that speaking a second language is generally beneficial for all children (6.7%) and one heard generally positive advice through another parent but did not directly receive any information (6.7%). Overall, the majority of children with DS did not receive any information concerning bilingualism (52%). Three received advice to restrict language input to English only (12%) and three others received positive support for two languages (12%). Four families did not think that

⁷ Mean scores are reported with standard deviations in parenthesis. Missing data for 5 children from the TDB group for SES, parent reported current exposure to Welsh and parent reported lifetime exposure to Welsh due to non-return of the parental questionnaire. Age of first word missing data for 20 participants due to non-return of questionnaire and parents not being able to recall this information. ^aIndicates a between-group effect of diagnosis with $p < .05$. ^b Indicates a between-group effect of language status with $p < .05$.

bilingualism applied to their situation (16%), one did not receive direct information besides through other parents (4%) and one did not respond (4%).

For the families of the TD bilingual children, the majority again did not receive any direct information about bilingualism (61%). Two families did receive positive information about bilingualism (9%), and the remaining families did not respond to this question (30%). No families reported receiving any negative information about bilingualism. For parents of the TD monolingual children, the majority did not receive any information specifically about their child learning a second language (53%). Three reported that they knew that bilingualism had positive effects (17.6%), two reported that it did not apply to them (11.7%), two reported that they had received information but did not elaborate on what the information was (11.7%), and one parent did not answer this question (5.9%). Overall, the majority of parents of TD children did not receive information about bilingualism (57.5%), five received positive information (12.5%), two stated that they received advice but did not explain what the advice was (5%), two felt the question was not relevant for them (5%) and the remaining families did not respond (20%). No families of children who were TD were reported to receive any negative information about bilingualism. This information is presented in Figure 6.

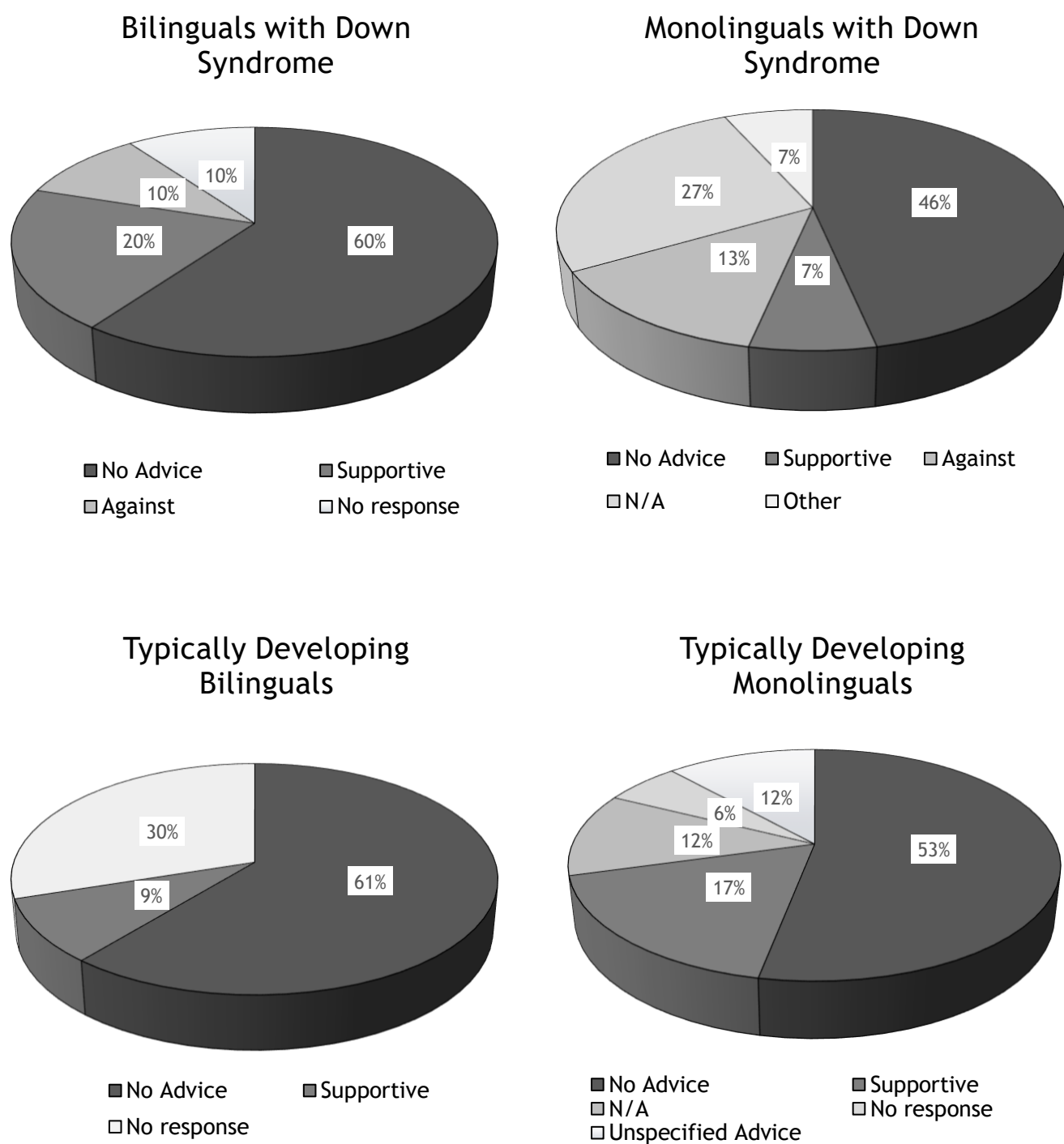


Figure 6. Distribution of parents who received supportive advice for bilingualism, advice against bilingualism, no advice or other response across the four groups.

5.2.3 Procedure

All of the children completed the English language assessments and the bilingual children were also assessed on their Welsh language development in order to get a full picture of their abilities. Assessments were all completed in the same predefined order as shown in Table 14 in Chapter 4 where more detail is provided on the procedure employed. Sessions were split into separate sessions according to the needs of each child, however, the Welsh and English sessions were separate and on different days when possible. For the focus of this chapter, expressive and receptive language abilities, the results for the language assessment in English were obtained from seven subtests of the CELF-Preschool Second Edition (Semel, Wiig, & Secord, 1998). For Welsh receptive language, a subset of the Prawf Geirfa Cymraeg: Fersiwn 7-11 (The Welsh Vocabulary Test: Version 7-11; Gathercole & Thomas, 2007) was utilized.

5.2.4 Data analysis

Standardised scores from the assessments were not appropriate as the participants were outside of the age range. Raw scores were instead used for the Welsh receptive assessment. For the English language assessments, raw scores were converted to Z scores.⁸ These enabled comparisons between groups and direct comparison between the different components of this measure; receptive, expressive and core language. A series of two-way analyses of variance (ANOVAs) were conducted to examine the effect of language status (monolinguals versus bilinguals) and diagnosis (DS versus TD). Any significant effects were followed up using Bonferroni corrections to decrease the familywise error rate given multiple comparisons or simple main effects analyses where appropriate.

5.3 Results

5.3.1 Core, Receptive and Expressive Language – Individually Matched Sample

Descriptive statistics for converted z-scores on the CELF-P are presented in Table 19 for individually matched participants and Table 20 for group matched participants⁹.

⁸ Analyses were also undertaken with scores converted to percentages for completeness and these returned identical results.

⁹ CELF-P subtests are as follows: SS = Sentence Structure; WS = Word Structure; EV = Expressive Vocabulary; C&FD = Concepts and Following Directions; RS = Recalling Sentences; BC = Basic Concepts.

CEL F – P Component	Component Sub-tests	Down Syndrome		Typically Developing	
		Bilingual	Monolingual	Bilingual	Monolingual
Core Language	SS	-0.178	-0.314	0.382	0.110
	WS	-0.524	-0.282	0.379	0.110
	EV	-0.383	-0.012	0.115	0.281
	Overall	-0.412	-0.113	0.253	0.272
Receptive Language	SS	-0.178	-0.314	0.382	0.110
	C&FD	-0.414	-0.050	0.441	0.023
	BC	-0.347	0.091	0.164	0.091
	Overall	-0.360	-0.127	0.442	0.045
Expressive Language	WS	-0.524	-0.282	0.379	0.110
	EV	-0.383	-0.012	0.115	0.281
	RS	-0.544	-0.466	0.709	0.301
	Overall	-0.412	-0.113	0.253	0.272

Table 19. Mean standardised z-scores on CELF-P subtests across groups (individually matched sample).

CEL F – P Component	Component Sub-tests	Down Syndrome		Typically Developing	
		Bilingual	Monolingual	Bilingual	Monolingual
Core Language	SS	-0.303	-0.341	-0.021	0.312
	WS	-0.669	-0.317	0.048	0.403
	EV	-0.503	0.081	-0.182	0.394
	Overall	-0.542	-0.130	-0.0166	0.523
Receptive Language	SS	-0.303	-0.341	-0.021	0.312
	C&FD	-0.561	-0.262	0.078	0.224
	BC	-0.440	-0.102	-0.017	0.185
	Overall	-0.470	-0.222	0.080	0.426
Expressive Language	WS	-0.669	-0.317	0.048	0.403
	EV	-0.503	0.081	-0.182	0.394
	RS	-0.769	-0.516	0.226	0.499
	Overall	-0.718	-0.259	0.115	0.556

Table 20. Mean standardised z-scores on CELF-P subtests across groups (group matched sample).

Univariate between-group ANOVAs for core, receptive and expressive language revealed an effect of diagnosis for expressive language $F(1, 36) = 6.666, p = .014, \eta_p^2 = .710$ but no effect of diagnosis for core or receptive language. After including CA as a covariate, an effect of diagnosis on expressive language remained. No effect was found for language status or any interaction for any language component. Given the effect of diagnosis on the expressive language component, follow up analyses were conducted which showed that the DS groups scored significantly lower than the TD groups on the word structure $F(1, 36) = 7.280, p = .011, \eta_p^2 = .168$ and recalling sentences sub-tests $F(1, 36) = 13.132, p = .001, \eta_p^2 = .267$. After controlling for CA as a covariate, a significant effect of diagnosis was found for all sub-tests. No effect of language status was found for any sub-test nor any significant interaction. Analyses also confirmed no significant differences between males and females, $t(38) = .025, p = .980$ and subsequently are not reported on further. Figure 7 presents a summary of core, receptive and expressive scores across the four groups.

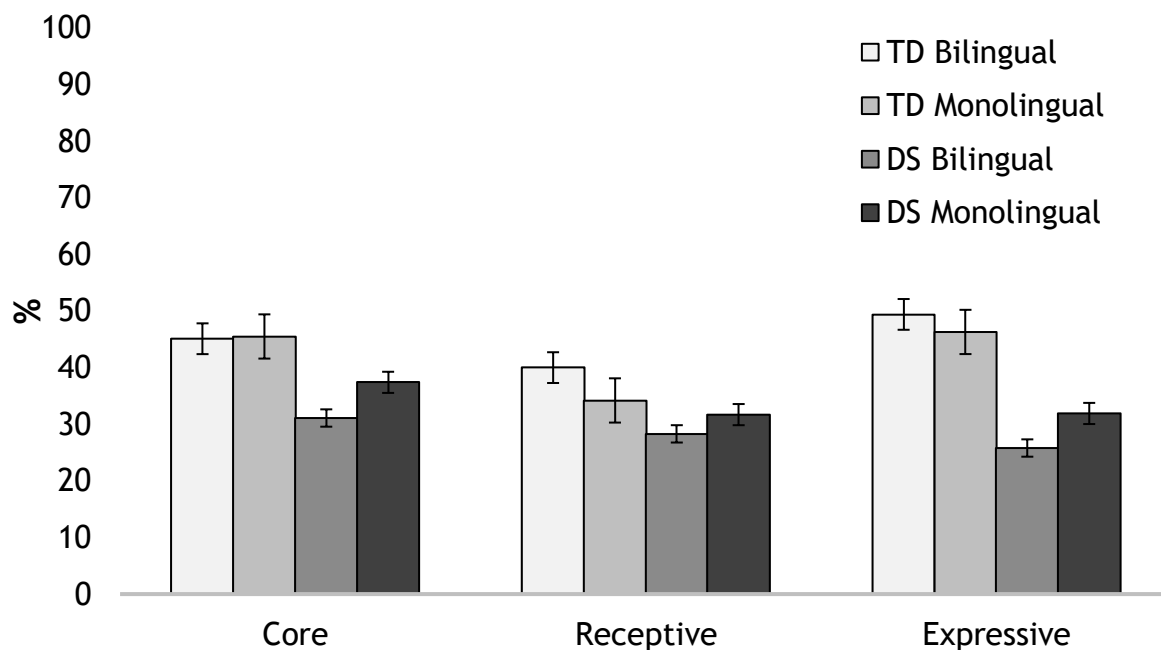


Figure 7. Mean scores on core, receptive and expressive language in English as measured by the CELF-P distributed by diagnosis and language status for individually matched sample.

In order to analyse Welsh language abilities in the bilinguals, further analyses were conducted for the TDB and DSB groups on the Welsh receptive language assessment. An independent-samples t-tests involving raw receptive language scores was conducted which found no significant differences between TDB ($M = 21.6, SD = 8.91$) and DSB ($M = 22.8, SD = 6.76$) groups $t(18) = .340, p = .738$ on this measure. Figure 8 shows group performance for the TDB and DSB group on this measure.

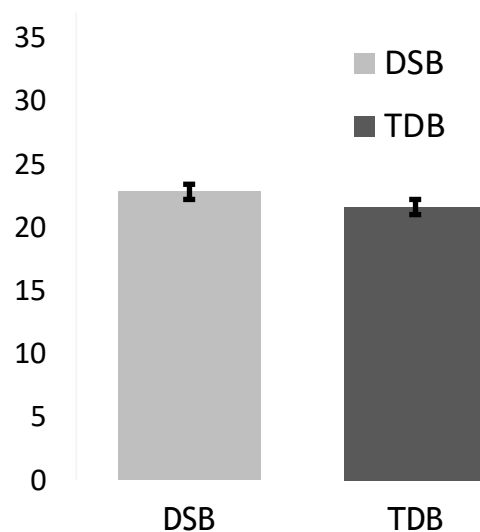


Figure 8. Mean raw scores on the Welsh receptive vocabulary assessment for bilingual groups.

5.3.2 Core, Receptive and Expressive Language – Group Matched Sample

Between-group ANOVAs for core, receptive and expressive language revealed an effect of diagnosis for expressive language $F(1, 59) = 11.322, p = .001, \eta_p^2 = .161$, core language $F(1, 59) = 5.796, p = .019, \eta_p^2 = .089$ and receptive language $F(1, 59) = 6.340, p = .015, \eta_p^2 = .097$. No effect was found for language status or any interaction between language status and diagnosis. Follow up analyses showed that the DS groups scored significantly lower than the TD groups on the sentence structure $F(1, 59) = 5.014, p = .029, \eta_p^2 = .078$, word structure $F(1, 59) = 7.746, p = .001, \eta_p^2 = .168$, concepts and following directions $F(1, 59) = 4.705, p = .010, \eta_p^2 = .078$ and recalling sentences sub-tests $F(1, 59) = 14.797, p < .001, \eta_p^2 = .257$.

After controlling for CA as a covariate, a significant effect of diagnosis was found for all sub-tests. The effect of diagnosis on core, expressive and receptive language also remained after controlling for CA. After including CA as a covariate, an effect of language status was also found for expressive language $F(1, 58) = 4.126, p = .047, \eta_p^2 = .066$. Analyses again confirmed no significant differences between males and females $t(63) = -1.034, p = .305$ and subsequently are not reported on further. Given the significant effect of language status on expressive language after controlling for CA, further analyses were conducted which showed that the bilinguals scored significantly lower for the expressive vocabulary subtest only with $F(1, 59) = 5.091, p = .028, \eta_p^2 = .079$. Analyses were run separately for DS and TD groups but no effect of language status reached significance for expressive language after controlling for CA as a covariate with $p = .210$ and $p = .199$ respectively. Figure 9 presents a summary of core, receptive and expressive scores across the four groups.

Analyses were again conducted for the TDB and DSB groups on the Welsh receptive language assessment. An independent-samples t-tests found no significant differences between TDB ($M = 22.77, SD = 8.33$) and DSB ($M = 22.8, SD = 6.746$) groups with $t(30) = .009$,

$p = .993$. Figure 10 presents the two groups performance on this measure. Additionally, a correlation analysis showed that there was a highly significant correlation between performance on the Welsh receptive assessment with receptive ($r = .704$, $n = 20$, $p = 0.001$), expressive ($r = .685$, $n = 20$, $p = 0.001$), and core language ($r = .752$, $n = 20$, $p < 0.001$) in English. The correlation between performance for the Welsh and English assessments is presented in Figure 11.

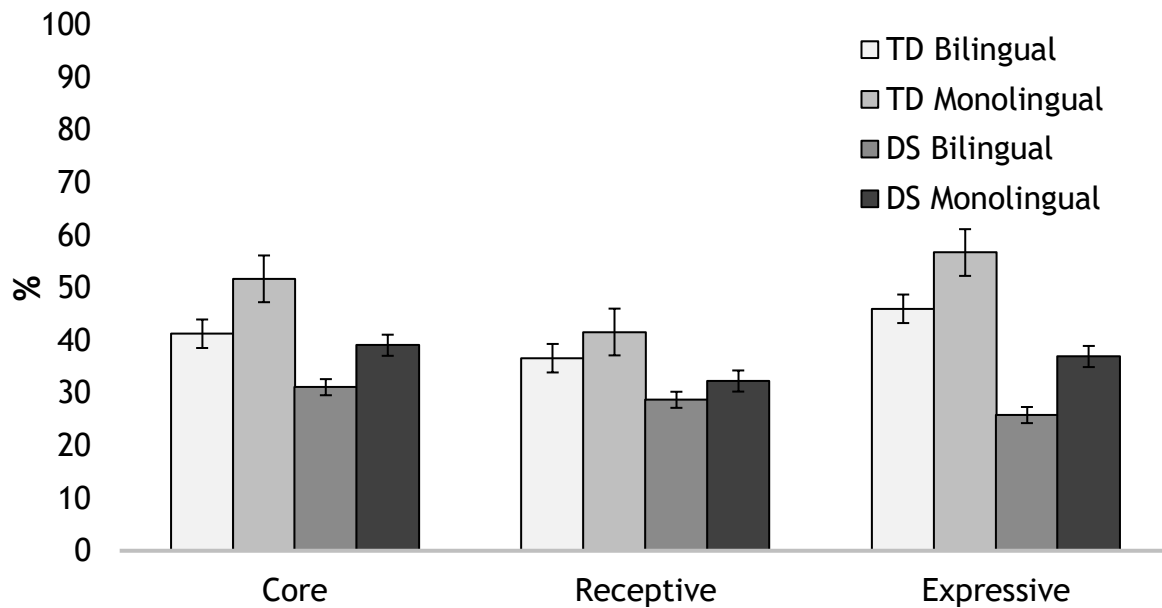


Figure 9. Mean scores on core, receptive and expressive language in English as measured by the CELF-P distributed by diagnosis and language status for group matched sample.

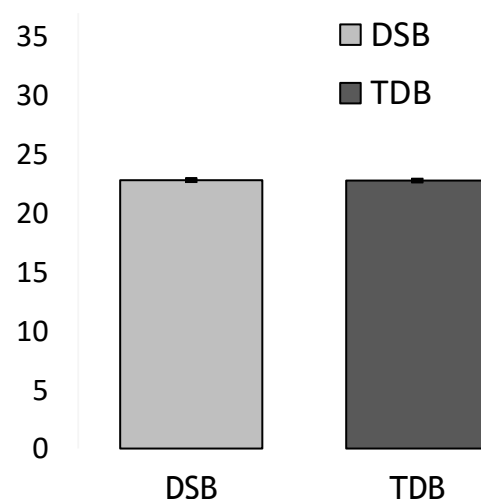


Figure 10. Mean raw scores on the Welsh receptive vocabulary assessment for bilingual groups

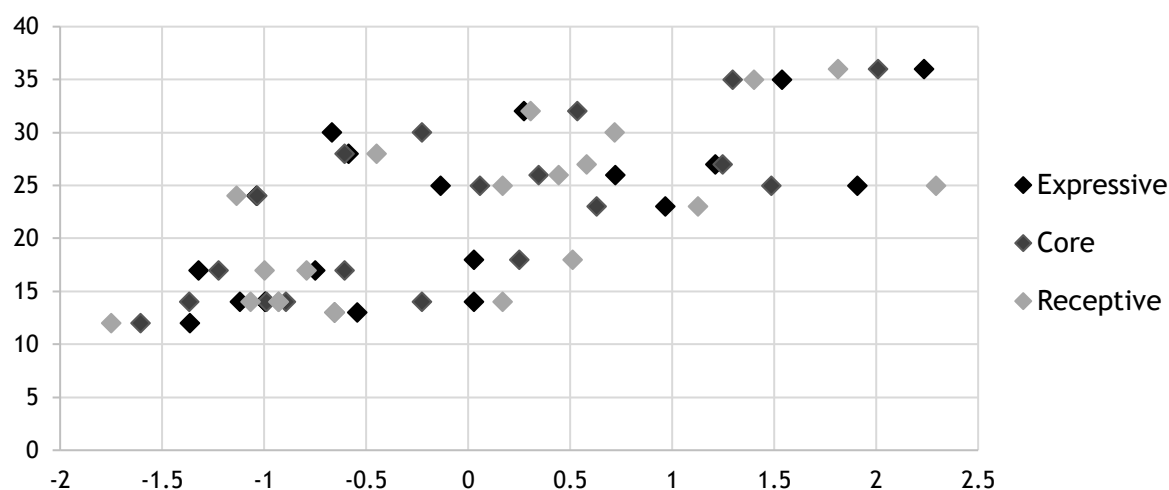


Figure 11. Correlation between Welsh vocabulary scores and English measures for the bilingual groups. *Note:* English scores reports as standardised z-scores (x-axis) and Welsh scores reported as % (y-axis).

5.3.3 Factors Relating to Language Outcomes

In order to address the final research question, linear mixed-effects regression models were employed to examine the predictive role of language input, SES, CA, WM and NVMA on receptive language abilities. To avoid multicollinearity, current and lifetime exposure to English could not both be entered into the regression model as they were both highly correlated with each other ($r = .92$). As current exposure is reported to play more of a role in predicting language outcomes (Hambly & Fombonne, 2014), this variable was used in subsequent regression models.

The model which accounted for the most amount of variance in considering all groups included CA, NVMA, English PA, WM and SES which explained a significant amount of variation on receptive language abilities ($F(5,34) = 33.78, p < 0.001, R^2 = 0.832$). Within this model WM, CA, NVMA and English PA were all significant predictors of English receptive language abilities ($p < .05$). Current exposure to English and SES did not significantly increase the explained variance. The variables that were significant predictors of English receptive abilities were then entered into separate models for both the DS and TD groups. All variables remained significant predictors for both populations ($F(4,15) = 35.150, p < 0.001, R^2 = 0.904$), and ($F(4,15) = 15.494, p < 0.001, R^2 = 0.805$) respectively. For DS groups, WM was the strongest significant predictor ($p = .001$), but for TD groups NVMA was the strongest significant predictor ($p = .015$) of receptive English language outcomes.

For Welsh receptive language abilities, a significant amount of variance was also explained by WM, CA, Welsh PA and current exposure to Welsh ($F(4,15) = 42.369, p < 0.001, R^2 = 0.919$). All of these variables were significant predictors of Welsh receptive abilities ($p < .05$). As with the English measures, SES did not increase explained variance. Current exposure to Welsh was a significant predictor of Welsh receptive vocabulary. Follow up Pearson's

correlations found a significant correlation for Welsh receptive ability and Welsh lifetime exposure for the DSB participants ($r = .773, p = .016$) only but not for the TDB group ($r = .18, p = .619$).

5.4 Discussion

The purpose of this research was to examine the language profiles of Welsh-English bilingual children with Down syndrome in comparison to English monolinguals with DS as well as bilingual and monolingual TD children. Given the difficulties in language acquisition documented in children with DS, this research aimed to examine how language profiles compare for bilingual children with DS with these developmentally matched control groups.

5.4.1 Language Abilities of Bilinguals with Down Syndrome

In this study, the language abilities of the bilinguals with DS were comparable to that of the children with DS who were only exposed to English after carefully considering developmental age, chronological age and socioeconomic status. For measures of expressive, receptive and core language ability in English, the bilingual groups performed comparably to that of the monolingual groups. This supports the suggestion that bilingualism does not lead to any additional delays or difficulties with language for children with DS. These results also support and strengthen previous findings with French-English bilinguals with DS (Kay-Raining Bird et al., 2005) in that they also report no detrimental impact of bilingualism.

Overall, no detrimental impact of bilingualism was found for any of the language measures assessed which included core, receptive, and expressive language. No evidence was found to support the view that bilingualism was detrimental to the language development of those with DS. For the group matched sample, only one measure resulted in a significant difference between bilinguals and monolinguals after considering chronological age, which was the expressive vocabulary subtest of the CELF-P. This finding is not surprising as many of the bilingual children were Welsh dominant and many of the TD bilinguals had only received limited exposure to English. This finding appears to reflect the role of language input on expressive vocabulary sizes within bilingual children, and this is frequently reported in the literature of TD children whereby bilinguals initially have smaller vocabulary sizes in each of their languages when considered separately (Bialystok et al., 2010). This finding was not replicated in considering the DS and TD groups separately, nor was this found in the individually matched sample.

As expected, a significant effect of diagnosis was found in that the TD children outperformed the DS groups, which was also reported by Kay-Raining Bird and colleagues. Previous research has documented that language is specifically impaired in those with DS and this finding reflects this. This was most evident for expressive language which is in line with literature that documents a specific weakness in expressive abilities for these individuals (Chapman et al., 1998; Roberts et al., 2007; Martin et al., 2009). More specifically, the results

show the greatest effect of diagnosis on assessments relating to word structure and recalling sentences. This finding coincides with previous research reporting marked difficulties with speech production and expressive morphosyntax (Andreou & Katsarou., 2013).

The findings of this study reinforce the suggestion of a specific language profile for those with DS. This research has also extended this profile to bilinguals with DS, as the same patterns of strengths and weaknesses emerged for both bilingual and monolingual groups with DS as the same aspects of language were challenging for both groups. This profile was found to be characteristic of the DS groups only as both of the TD groups displayed similar levels of receptive and expressive language abilities. Further comparisons between the DS and TD groups found that parental reports of early language development also coincides with marked language delays as the two groups of children with DS were reported to produce their first word significantly later than the two TD children. For the DS groups, the mean age of first word was almost double that of the TD children, at around 23-24 months. This again is in line with the literature that reports early language delays in children with DS (Oliver & Buckley, 1994). No significant difference was found between the age of first word between the bilingual and monolingual children.

Previous research suggests that parents of children with a DD such as DS have been advised to restrict input to a single language, which in most cases is the majority community language (Marinova-Todd et al., 2016a). These recommendations are reported to have stemmed from the view that bilingualism may result in additional delays or impairments in language development. If this view were to be true, it would be expected that impairments would be most apparent in aspects of language which are already particularly compromised in children with DS. As a result, the finding that no further deficits within the domains of expressive language and grammar is a meaningful outcome as these aspects of language are frequently reported to be impaired to a greater extent than other aspects of language.

Bilingual groups were also assessed on their Welsh receptive vocabulary. No differences were found for receptive vocabulary sizes as measured using a subset of the Prawf Geirfa (Gathercole & Thomas, 2007) between the bilinguals with DS and the developmentally matched TD bilingual children. Consequently, the bilinguals with DS also had measurable language abilities in Welsh, which were comparable to the receptive vocabulary sizes of the developmentally matched TD Welsh-English bilinguals. All bilingual children were considered simultaneous bilinguals due to the fact that all were exposed to two languages before the age of 3, which is frequently used as a cut-off point between sequential and simultaneous bilinguals by researchers (Paradis et al., 2011), although the majority were exposed to both Welsh and English from birth. As only a small number were exposed to a second language after the age of 2, it was not appropriate to conduct any analyses to identify any differences between children who received exposure to two languages from birth and those exposed at an older age.

In summary, no differences were found between the bilinguals and monolinguals with DS, and the bilinguals with DS were developing receptive Welsh language abilities that were

commensurate with TD bilingual children who were at the same stages of development. In addition to strengthening previous findings within this field, this research has extended these results to a different population as many of the children in the current study were dominant in the minority language (Welsh) or balanced bilinguals, whereas the majority of children in Kay-Raining Bird et al.'s study were dominant in the majority language (English). In addition, older children were included in the current study than in the previous group study of bilinguals with DS. The children in the current study were chronologically older with higher developmental ages with more advanced language abilities than the children with DS in Kay-Raining Bird et al.'s study. Therefore, this research has increased understanding of bilingual language development in individuals with DS in children who had progressed further in their language development and explored novel aspects of language development.

5.4.2 Professional Recommendations

As highlighted in the literature review in Chapter 2 and at the beginning of this chapter, previous research suggests that parents of children with a DD such as DS have been advised to restrict input to a single language, which in most cases is the majority community language (Marinova-Todd et al., 2016a). These recommendations may be due to the belief that two languages would post too many additional challenges on a child with DS, given that children with DS generally experience substantial difficulties with language, to begin with. One aim of this chapter was to identify what advice parents and caregivers of children with DS and TD children receive regarding exposure to a second language.

Findings of this research show that only a small number of families of a child with DS were recommended to limit language input to a single language (12%) with the suggestion that exposure to two languages would be too challenging, in the current study. At the same time, an equally small number of families (12%) received encouragement in their decision to expose their child to both English and Welsh or any supportive information about the utility of their child being included within bilingual families and schools. This is in contrast to the parents of the TD children, in which no family reported that they had ever received any negative information about bilingual language development. Surprisingly, only a small number of parents of the TD children had ever received any positive information regarding outcomes of bilingualism (12.5%) also.

Perhaps most concerning was the fact that the vast majority of parents of children with DS did not receive any information about how the addition of a second language may impact on their language development (52%). Although some responded that bilingualism did not apply to their situation (16%), even for the children attending English-medium schools, it is still required that a small amount of Welsh is taught due to the protected status of the Welsh language (Welsh Language Commissioner, 2011). Consequently, it appears that there is a substantial lack of information surrounding this issue which may have led to families feeling unsupported and having to speculate about whether they have made appropriate decisions regarding their choice to expose their child to a second language or not, for those

families that felt they were in a position to make such choices. This consequently has further implications on education policy in Wales which is discussed later.

Although no piece of research has explored the advice that parents of children with DS receive specifically, previous research investigating the advice that parents of children with DDs in general and parents of children with ASD have reported that a large proportion of families received negative advice relating to bilingual outcomes. For example, Kay-Raining Bird and colleagues (2012) conducted a survey study in which they reported that only 8% received positive support, 30% received advice against bilingualism and a further 14% received conflicting advice. Parents in the present study report to have received more positive advice and less negative recommendations than previous studies, however, more parents were left with a complete lack of information. This may be an indication that practitioners are becoming more aware of the current research in the field and view bilingualism more favourably than has previously been reported, as suggested by Marinova-Todd et al. (2016). Alternatively, it may simply be due to differences in expectations in regards to language abilities between children with DS and ASD. Guidelines have recently been introduced by the Royal College of Speech and Language Therapists (RCSLT) in 2019 which now states that families should “never be advised to abandon home language in favour of the majority language (typically English, Welsh or Gaelic) under any circumstances” (Bilingualism-Guidance Key Points: RCSLT, 2019, p.23) and that it is recognized that bilingualism is in itself an advantage.

On the whole, it appears that views held by professionals towards bilingualism for those with DS are not as negative as previous reports from studies undertaken with other DDs, with only a small number of families receiving explicit advice to restrict language input to English only. In contrast, a small number were supported in their decision to raise their child bilingually. On the other hand, a large proportion of parents did not receive any information or advice on how to approach bilingualism within the Welsh language context. Therefore, there still needs to be an increase in understanding of the research that currently exists in relation to bilingualism in DS to ensure that evidence-informed recommendations can be provided to families in the future. Following this, tailored bilingual speech and language therapy sessions, assessments, and interventions should be employed in order to appropriately support Welsh-English bilingual children with DS.

5.4.3 Factors Associated with Language Outcomes

In order to gain an insight into the relatively high degree of within-group variability which is often observed in those with DS, several factors were considered in relation to receptive language abilities. These were: current language input, chronological age, developmental age, parental socioeconomic status, phonological awareness and working memory. For the DS and TD groups, significant predictors of language comprehension were working memory, developmental age, chronological age and phonological awareness which explained 90% of the variation in the DS groups and 81% in the TD groups. The strongest predictor of receptive

language for the DS groups was WM, however, for the TD groups, non-verbal IQ had the largest impact. This may have some important clinical implications if the role of WM contributes the most to language comprehension, as targeting both language and working memory in therapy sessions may prove to be more successful with this population.

As reported elsewhere, NVMA seems to play less of a role in determining language outcomes in bilinguals with DS (Kay-Raining Bird et al., 2005) and monolinguals with DS (Abbeduto et al., 2003) in comparison to TD children. In addition, current exposure to English did not significantly improve this model. In contrast, the amount of lifetime input in Welsh was significantly related to Welsh receptive vocabulary scores for the bilingual DS group only. This suggests that receptive language abilities in children with DS are somewhat related to the amount of input, particularly in minority language contexts. Previous research with TD children (Haman et al., 2017) and children with ASD (Gonzalez-Barrero & Nadig, 2018) have reported that in bilinguals, the amount of current input is predictive of language abilities. In researching those with DS specifically, Kay-Raining Bird and colleagues also did not find that duration of exposure to their L2 was predictive of language abilities in bilinguals with DS.

Similar findings to the current research are reported in the only study which examined the frequency of L2 exposure in bilinguals with DS in a parent report study (Trudeau et al., 2011). In their research, the amount of input to the majority language (English) was not related to language abilities, however, input to an L2 (simply defined as a language other than English as opposed to the second or weaker language necessarily) was related to vocabulary development. The lack of continuity between these studies may be due to the specific profile of receptive and expressive abilities found for individuals with DS specifically. This also provides some evidence that the amount of input does play a role in determining language abilities for those with DS when there is one minority language and one majority language. It is also possible to consider the other factors which were included in the analyses which play a role in determining language abilities for this population. These factors may be stronger than the impact of current exposure, the age of first exposure or duration which may outweigh these effects. The variability within and between groups may also explain why a relationship was found for Welsh input but not English as for the bilingual groups, large variations in exposure to Welsh and English were reported. Nevertheless, research employing longitudinal designs and more thorough assessments of language input and language use are needed to explore the relationship between input and language outcomes in this population further. Socioeconomic status as measured by parental education and occupation also did not significantly predict language abilities in either Welsh or English for TD and DS groups.

5.4.4 Limitations and Implications

The first limitation to note is the relatively small sample size which is often characteristic of research assessing language development in children with DDs and in particular bilingual children with DDs. Although the sample is relatively small, the overall sample included more children with DS and more bilingual children with DS than all current studies investigating

bilingualism in those with DS to date. In addition, the small sample size is reflective of the small overall target population (i.e. Welsh-English bilinguals with DS specifically). Alternatively, the number of bilinguals with DS may also be a reflection of the advice that they received in relation to bilingualism or the lack of information available. This may have led parents to avoid exposing their children to an additional language, for instance selecting English-medium schools as opposed to Welsh or bilingual schools. As the individually matched sample is relatively small, analyses were undertaken with a larger group matched sample in order to overcome possible lack of effect sizes which may be apparent with a smaller sample. The fact that the findings reported for both samples led to the same results strengthens the conclusions of the research.

Secondly, the focus of this research was on simultaneous bilinguals with DS, however, the sample was not completely homogeneous in terms of the degree of bilingualism. The majority of bilingual children were dominant in Welsh as they received input in Welsh at home and school, however, a few were English dominant or balanced bilinguals with English as a home language and input to Welsh at school only or mixed language use at home and school. Consequently, there may be differences between these groups of bilinguals that were not identified in the current research. Likewise, differences may have emerged if it were possible to sub-divide the bilingual speakers according to the age of exposure to each language. Again, as the vast majority received input to both languages from birth, it was not possible to separate groups further according to the age of exposure to Welsh and English and were consequently all examined as bilinguals which encompassed a range of children with varying current and lifetime exposure to Welsh and English. As reported in section 4.2.1, analyses were conducted to ensure the validity of the bilingual status of the groups compared to the monolinguals, however, future research should employ larger samples wherever possible in order to differentiate between those receiving exposure to two languages from birth and those receiving exposure within the first 3 years in order to ascertain if there are any differences in language outcomes.

One important implication arising from this work concerns the inclusion of children with DS within Welsh-medium educational provisions. The linguistic situation in Wales in terms of schooling provides the opportunity for all children to receive rich input in Welsh and English and subsequently become bilingual if parents choose for their children to attend a Welsh-medium or bilingual school. The findings of this research support the proposal that children with DS should also be able to access and attend Welsh-medium or bilingual schools and can flourish in these settings. Recent research examining the experiences of bilingual children with ASD in Wales reported that children who were included in Welsh-medium schools felt more positively towards bilingualism in general and felt more socially active in their classrooms (Howard, Katsos & Gibson, 2019). This suggests that including children with ASD in Welsh-medium mainstream schools offers a positive experience and a positive impact on social development. The inclusion of children with DS within Welsh-medium educational settings may subsequently have a positive impact on the social development of these children

and also be a positive experience for the children themselves, although further research is needed to examine if this is also found for children with DS.

In light of these findings, policies and support within Welsh-medium schools need to be available which reflect the research evidence that exists in the field of bilingualism and DS. Furthermore, the findings of this research and previous research investigating bilingualism in DS should be taken into consideration when decisions are made regarding home, school and therapy language use. Clinicians should employ evidence-informed practice when it comes to providing SLT and when giving advice to parents and schools pertaining to bilingualism. Recent research suggests that professionals working with children who have a DD may be more positive towards bilingualism than in previous studies (Marinova-Todd et al., 2016b). Nevertheless, policies and resources in light of these findings are required to ensure that appropriate practice takes place which ultimately needs to be in line with each child's needs which will then allow them to be fully included and supported in bilingual communities and educational provisions.

5.4.5 Conclusion

To conclude, the current study is the first piece of research that reports on the language profiles of bilingual children with DS in the UK in comparison to developmentally matched monolingual children with DS as well as TD control groups. In summary, this research reports that bilinguals with DS perform comparably to monolinguals with DS on all aspects of language under study and similar developmental profiles for both populations after considering chronological age, socioeconomic status and working memory. Comparable profiles of language in terms of linguistic strengths and weaknesses were also found for bilingual and monolingual children with DS. Caution is warranted due to the relatively small sample size within this population, although the sample contained early simultaneous bilinguals with English both as an L1 or L2. Furthermore, consideration should be given to the fact that bilingual speakers are a heterogeneous population and a bilingual's consistency of exposure to each language may be dynamic and dependant on a number of factors. This may particularly be the case in the context of children with DS whereby the functions of each language may differ according to the context and environment of testing (Oller, Pearson & Cobo-Lewis, 2007).

Further research is required to increase understanding of this field further and should employ longitudinal methods in order to ascertain the developmental trajectories of bilingual children with DS. Exploration of further elements which may impact on language acquisition for this population is required, for example, the role of early parent-child interactions and gesture usage. Finally, this research provides evidence that children with DS should be included in bilingual services and educational systems and future professional recommendations should be made in line with research evidence. In contrast, if one language is discontinued as a result of receiving advice against bilingualism, this may inadvertently hinder development if the consistency of language input is altered and the quality and

quantity of input are reduced. This research supports and extends previous findings that document no adverse outcomes for bilingual children with DS as well as developmental disabilities as a whole.

Chapter 6: Phonological Awareness in Bilinguals with Down Syndrome

This chapter will address the third research question: “How does phonological awareness develop in bilinguals and monolingual with Down syndrome?” The findings which relate to the phonological awareness abilities of Welsh-English bilingual children with Down syndrome in comparison to developmentally matched control groups will be presented, including relative strengths and weaknesses for each population. Towards the end of the chapter, the theoretical, clinical and educational implications of the research will be highlighted and discussed, as well as future research directions in this field.

6.1 Introduction

Phonological awareness (PA) is a well-established concept that refers to an individual’s ability to separate the form of language from its meaning. In developing PA, individuals learn to identify and manipulate units of speech of various sizes. In this respect, it is often considered a multidimensional metalinguistic ability in which a listener or speaker is capable of attending to and altering the phonological sound structure of a language by objectively viewing language. In alphabetic languages (e.g. English and Welsh), there are three levels of PA; syllable level, intrasyllabic (onset/rhyme) level and a phoneme level. In tonal languages (e.g. Mandarin and Chinese), tonal awareness is also considered an additional element with children learning these languages being able to discriminate between tones. Many factors play a role in the development of PA, one of these being the language(s) being learnt, particularly concerning the transparency of the orthography and the consistency of the grapheme to phoneme mappings. In addition, the age of formal reading instruction, the complexity of the language and the nature of each level of PA and how they are assessed will impact the emergence and detection of PA.

The importance of PA resides in the fact that it has been reliably demonstrated to play a crucial role in early reading and spelling development in children who are learning an alphabetic language who show typical and atypical developmental profiles (Carnine et al., 2004; Melby-Lervåg, Lyster & Hulme, 2012). Resultingly, this association has attracted much research interest in recent years. Research has shown that PA is the strongest predictor of later reading abilities, with PA accounting for more variability in reading scores than vocabulary and cognitive abilities (Melby-Lervåg, Lyster & Hulme, 2012). Likewise, observations of children who have marked difficulties in developing PA have been identified as being negatively impacted in terms of their later reading attainment. In turn, these difficulties may also extend to other aspects of language such as expressive and receptive vocabulary sizes, syntactic abilities and morphological abilities, as these aspects are found to correlate with levels of PA (Dickinson et al., 2003; Smith & Tager-Flusberg, 1982; Chaney, 1992).

Typical measures and tasks used to assess PA generally include identifying, segmenting, blending and manipulating levels of speech sounds. In TD children, these skills are believed to begin to emerge at around 2-3 years old, and the majority of children will have mastered these abilities by the time they reach age 7 (Lieberman et al., 1974; Lonigan, Burgess, & Anthony, 2000). The following sections will summarise the development of PA in three groups of children; TD children, bilingual TD children and children with DS.

6.1.1 Phonological Awareness in Typical Development

In TD populations, researchers have proposed two alternative accounts of PA. The first, suggests that PA is one single unitary construct that encompasses all three elements of PA. This proposition is supported by research which suggests that children's abilities on a range of measures across the three levels of PA can usually be statistically explained as one overarching ability in designs that employ factor analyses (Branum-Martin et al., 2012). It is also suggested that the fact that PA abilities appear to transfer across languages (Bialystok, Majumder & Martin, 2003) and the fact that this cross-language transfer has also been reported to be bidirectional (Dickinson et al., 2004), supports the proposal of PA being a single ability. Although evidence also supports the suggestion that the language being learnt influences the rate of development (Durgunoğlu & Öney, 1999), as these cross-language transfer effects appear to be fairly universal, this also suggests that PA is a single general ability.

The contrasting view proposes that PA is a multidimensional construct that is separated according to the three levels of PA mentioned above (i.e. syllable, intrasyllabic and phoneme). This proposal stems from the idea that there may be different underlying cognitive processes that are responsible for the development of each aspect of PA. Within both of these interpretations, the developmental progression of PA is believed to begin with the larger units (i.e. syllables and rhymes) before the smaller units (i.e. phonemes; Treiman & Zukowski, 1991; Carroll et al., 2003; Anthony et al., 2011), at least in TD children. Metsala and Walley (1998) argue that this progression of development is due to the increase in vocabulary size that occurs with language development (also referred to as the Lexical Restructuring Hypothesis). More specifically, as children acquire more words, they are required to employ more fine-grained representations to successfully distinguish between similar words in their expanding vocabularies.

Further research has supported these claims, for example, Newman et al. (2006) reported that children who displayed greater speech segmentation abilities as infants had larger expressive vocabulary sizes at the age of 2. Furthermore, the same children had better performance on a range of language assessments at 4 years of age. Interestingly, this was not the case for general cognitive development, suggesting a unique relationship between vocabulary size and PA specifically. More recently, Singh et al. (2012) also reported that early speech segmentation skills were strongly correlated with expressive vocabulary sizes at two

years of age in a longitudinal study. Although this supports the claim that increased vocabulary size is related to PA, it is not clear whether the increase in vocabulary is responsible for the earlier PA abilities or vice versa. Alternatively, it is plausible to suggest that infants' ability in speech segmentation facilitated the development of expressive vocabulary. A further pattern of development that has been identified in TD children refers to the abilities in relation to implicit and explicit PA. Although perhaps intuitive, children are able of identifying phonological discriminations before they are able to alter or manipulate them. Furthermore, Anthony et al. (2003) reported that children were able to blend phonological information before they are able to segment them, a finding which was also reported earlier by Yopp (1988). These patterns will be interesting to consider in relation to other populations.

In languages that have a more transparent orthography (i.e. those with a close phoneme to grapheme correspondence), the development of PA is reported to be accelerated (Cossu et al., 1988; Goswami et al., 1997; Öney & Durgunoğlu, 1997). This has been referred to as the Orthographic Depth Hypothesis (Frost, Katz & Bentin, 1987; Katz & Frost, 1992) and it is suggested that if children are acquiring languages which are more orthographically transparent than English (which is known for having a very opaque orthography), they will have more advanced awareness of phonemes, and be more accurate readers than age-matched English learners, however, this pattern is believed to disappear when English learners 'catch-up' with increased formal reading instruction. Additionally, further linguistic features of the language being learnt may also influence the development of PA. For example, researchers have identified that the increased complexity of the language being learnt may result in slower progression of PA. Children learning languages which have more simple syllable structures (for example fewer clusters) or more obvious syllable boundaries (e.g. Turkish or Italian) are found to acquire syllable awareness quicker (Demont & Gombert, 1996; Durgunoğlu & Öney, 1999). This provides further evidence that the language being learnt in itself influences the rate at which PA develops.

Finally, as aforementioned, the relationship between PA and educational outcomes (i.e. reading and spelling development) has been frequently documented in the literature. Phonological awareness has been robustly reported as the key foundation for later reading and spelling development (Carnine et al., 2004; Melby-Lervåg, Lyster & Hulme, 2012). It may be useful to consider the directionality of this process (i.e. whether PA influences reading development or vice versa) or whether this process is bidirectional. Although implicit PA may develop prior to formal reading instruction, Gombert (2002) suggests that reading acts as a catalyst for the development of explicit PA knowledge. Further evidence of the directionality or bidirectionality of this relationship has emerged from intervention studies. Bradley and Bryant (1983) reported that interventions that targeted the development of PA were successful in enhancing not only PA but also the speed of reading development, suggesting that PA is responsible for stimulating reading abilities.

6.1.2 Phonological Awareness in Typically Developing Bilinguals

As highlighted above, one key factor that influences the development and the progression of the acquisition of PA is the language being learnt. Therefore, if a child is acquiring two (or more) languages, it is appropriate to consider the possibility that these languages may interact or influence each other in some way (for recent reviews, see Branum-Martin et al., 2012; Kuo et al., 2016). For other aspects of metalinguistic awareness (e.g. executive functioning and executive control), many researchers report superior performance compared to monolingual control groups (Adesope et al., 2010; Bialystok, 2015), although literature in the field is mixed (Paap & Greenberg, 2013) and claims made are often met with criticisms. The explanation behind this proposed benefit is that a bilingual speaker is required to constantly monitor, inhibit and switch attention between two (or more) languages. In turn, this is believed to enhance the cognitive mechanisms that are responsible for undertaking these skills.

For PA specifically, it has been proposed that benefits (i.e. accelerated development) are also observed (Campbell & Sais, 1995; Bialystok et al., 2003; Chen et al., 2004; Canbay, 2011). Positive effects have been found for bilinguals and this has been reliably shown in terms of word awareness (Cummins, 1978; Eviatar & Ibrahim, 2000). Although mixed effects have also been reported for other aspects of PA, the majority of studies that have reported these advantages have included observation of bilingual speakers in which the L2 is simpler, more regular or more salient in terms of phonological structures than the L1. For example, a positive effect of bilingualism was reported by Campbell and Sais (1995) in young English-Italian bilingual children, whereby the bilinguals outperformed the English monolinguals on tasks assessing syllable deletion and phonemic identification. It was proposed that as the children were learning Italian, which has a more regular syllable structure than English, this subsequently facilitated the development of PA among the bilinguals at the syllable level. This suggestion is also supported by research which investigated the development of tone, onset and rhyme awareness in Cantonese-Mandarin bilinguals in comparison to Mandarin monolinguals (Chen et al., 2004). This study reported that the bilingual group was more accurate in detecting tonal differences. As Cantonese is a rich tonal language, it was suggested that this feature accelerated the development of tonal awareness for this group of bilinguals.

Alternatively, if the L2 is substantially different from the first language or the L1 has phonological structures that are simpler than the L2, the bilingual advantage should not be evident according to this theory (Cross-Language Transfer Theory). In this instance, PA would not be enhanced in either language. Null effects have been reported which supports this proposal. For example, Bialystok and colleagues undertook a range of studies that encompassed a number of PA tasks in Mandarin-English bilingual children in comparison to matched English monolinguals (Bialystok, Majumder & Martin, 2003; Bialystok, Luk & Kwan, 2005). In these studies, there was no evidence to suggest that the bilinguals had enhanced performance compared to the monolinguals on any of the measures employed. Negative transfer or the lack of transfer in these studies have been rationalised by the fact that English

is a more complex language than the L1 (Mandarin), and that the languages did not share enough similar properties, such as English being an alphabetic language, whereas Mandarin is not.

Initial theories proposed that this bilingual benefit was due to transfer effects between a speaker's L1 and L2. In order to investigate this, Dickinson et al. (2004) undertook a large longitudinal study with Spanish/English bilingual children who were aged between 2-5 years old. The children completed PA assessments in both languages at various time points. The authors found evidence of cross-language transfer in which PA development was facilitated by earlier abilities in the other language. It was concluded that PA is highly influential in developing PA in the other language and vice versa. The positive effects of bilingualism and the null effects reported above, appear to support the proposal of cross-language transfer effects within PA. This theory has been extended further by Kuo and Anderson (2010) in what they term as the Structural Sensitivity Theory and relates to the research that suggests that bilinguals have heightened cognitive flexibility in terms of PA specifically.

This theory proposes that bilingual speakers have a "greater readiness to reorganize linguistic input and impute linguistic structure" (Kuo & Anderson, 2012; p.457). It is suggested that developing PA is necessary to avoid interference between a child's two languages, whereby they become more attuned to attending to the structure of language by identifying structural similarities and differences between words. This is suggested to consequently form more abstract representations of the languages being acquired. Furthermore, the more experience and variety that is observed within languages is believed to lead to quicker development of these abilities in abstracting the word form independently from meaning, which accelerates the acquisition of PA. The way in which this theory differs to that of the Cross-Language Transfer Hypothesis is that the Structural Sensitivity Theory postulates that experience in both languages assists in facilitating the development of PA as opposed to a 'carry-over' effect from one language to the other (and vice versa) as suggested by Cross-Language Transfer Hypothesis. In sum, this theory proposes that the bilingual experience in itself leads to quicker processing of phonological stimuli.

A different but related theory is the proposal of the Lexical Restructuring Hypothesis (Walley, 1993; Metsala & Walley, 1998) and that of Psycholinguistic Grain Size (Ziegler & Goswami, 2005). The process of acquiring new vocabulary requires the listener to discriminate new items from similar-sounding competing items. As more and more items enter an individual's vocabulary (or their lexicon), the lexical representations need to become more detailed and more fine-grained to be successful. This lexical restructuring is believed to occur each time a new word is acquired and the representations of these words need to include fine phonetic details of these words. This is believed to assist with the development of PA in bilinguals, as speakers of two languages may encounter more phonological competitors with more potential competing words. This would also help to explain the developmental progression of PA, with larger units being acquired first and the smaller units

(i.e. phonemes) being acquired later when more fine-grained representations are made (Kuo et al., 2016).

Finally, one further proposal that has been presented to explain differences between the development of PA in bilingual and monolinguals concerns the orthography of the languages. This factor was mentioned above as the languages being learnt appears to influence the rate of reading and PA development (Goswami et al., 1997; Öney & Durgunoğlu, 1997). This was explored within the Welsh language context by Spencer and Hanley (2000; 2003) whereby they investigated reading and phoneme awareness in Welsh-English bilingual children learning to read Welsh in comparison to English monolinguals learning to read in English. As Welsh is an orthographically transparent language whereas English is not, it was hypothesised that the children learning Welsh would show superior reading and phoneme detection abilities. Across the two experiments, the Welsh-English bilingual children learning to read in the transparent language (Welsh) outperformed the monolinguals learning to read English for both real words and nonwords. In addition, the children learning Welsh performed better than the children learning English on the phoneme awareness task. This is reported as evidence to support the Orthographic Depth Hypothesis, however, it could also serve as evidence for the bilingual advantage hypothesis. The authors to consider this possibility, however, as they did not find an increase in the difference between the bilinguals and monolinguals at the second time point (when the bilinguals were considered to be 'more bilingual'). They conclude that learning Welsh was responsible for these gains as opposed to bilingualism. Given that previous research suggests that bilingualism does play a role in enhancing PA in languages with differences in their orthographies, it would be justified to explore this factor further.

In summary, although caution is raised due to null effects being reported, researchers have proposed that a bilingual advantage does exist for PA and theories have been proposed which aim to explain why this benefit is found in some circumstances but not in others. If these advantages do exist for bilingual speakers in these specific contexts, this will provide a further understanding of the relationship between PA, literacy, and cognitive development in bilinguals. These will be especially useful in considering those with specific challenges in developing PA.

6.1.3 Phonological Awareness in Children with Down Syndrome

Individuals with DS are reported to have a specific language phenotype whereby some linguistic aspects are specifically impaired, whereas other areas are not impaired to the same extent. Within this phenotype, PA has been reported to be specifically impaired with children with DS displaying weaknesses in both identifying and manipulating various units of speech sounds (Cossu, Rossini & Marshall, 1993; Fletcher & Buckley, 2002; Næss et al., 2015; Næss, 2016) in relation to their cognitive abilities. Initial investigations of PA in individuals with DS suggested that these children were unable to develop PA abilities altogether and that PA was

not associated with reading development in this population. These claims received numerous criticisms due to the complexity of the tasks utilised and due to the interpretations of the results. Recent research now reports that with appropriate instruction and assessments, children with DS do develop PA, and these skills are associated with reading and spelling development (Fletcher & Buckley, 2002). It is acknowledged that PA is specifically impaired in individuals with DS, but it is not the case that these individuals lack these skills completely. Furthermore, a recent systematic review conducted by Lemons and Fuchs (2010), concluded that children with DS are usually outperformed by TD children, although commensurate abilities are reported in research that employs matching based on NVMA. Others have similarly concluded that marked difficulties in acquiring PA abilities lead to specific difficulties in learning to read for those with DS (Hulme et al., 2012).

Individuals with DS may adopt a sight-word reading approach as opposed to a decoding strategy that most TD children employ. In whole word reading strategies, children learn to associate and recognise words based on their appearance as opposed to identifying relationships between letters and phonemes. As children with DS often have visual processing strengths in comparison to their verbal skills (Yang, Conners & Merrill, 2014), researchers have sought to identify if sight-word reading interventions are more successful in developing PA in children with DS than decoding strategies. Although sight-word interventions have been found to be successful in developing reading skills in children with cognitive disabilities (Browder et al., 2006), interventions employing decoding strategies have also been successful in developing PA and letter knowledge for young children with DS specifically (van Bysterveldt, Gillon & Moran, 2006). Therefore, it appears that children with DS are capable of developing PA if provided with phonics-based instruction, which is usually the case in educational provisions for children learning alphabetic languages, such as English and Welsh.

Researchers have also sought to identify if children with DS develop PA in a similar progression to that of TD children in terms of the skills associated with each of the three main components of PA; rhymes, syllables and phonemes. Fletcher and Buckley (2002) reported that the developmental progression for the children in their study appeared to conform to the trajectory of development reported for TD children. In their study, different abilities were found for different tasks, which implies that the tasks were measuring different skills within PA. Within these tasks, the children were reported to have higher skills in terms of blending and segmenting items. The authors suggested that children with DS mastered PA in a similar manner to that of TD children, albeit at a slower pace. In contrast to this view, others have proposed that individuals with DS do not display the same developmental profiles to TD children. More specifically, researchers have reported a pronounced difficulty with tasks designed to assess the rhyme component of PA (Cardoso-Martins et al., 2002; Kennedy & Flynn, 2003; Hulme et al., 2012). This finding is in line with the view that PA is not a single ability but rather a multidimensional ability (Newman et al., 2006).

Furthermore, Snowling, Hulme and Mercer (2002) reported on three studies in which the PA profiles of 29 children with DS were compared to a TD control group, matched for their

reading abilities. Although both groups performed similarly to each other for word and non-word reading, the children with DS performed more poorly on the PA measures. After considering verbal abilities in their analyses, the DS group still exhibited deficits in rhyme detection for all three studies. The authors reported that children with DS appear to have a specific difficulty with detecting rhymes and many of the children with DS in the study were only performing at chance. In a recent review and empirical study, Næss (2016) also reported a specific weakness for PA in children with DS as well as additional challenges with rhyme awareness specifically. This was true for the meta-analysis that was conducted (with large effect sizes) and for the longitudinal study whereby PA was assessed at two-time points.

This finding led to the proposal that children with DS may exhibit a specific difficulty in attending to word endings. Snowling et al. (2002) reported difficulties with rhyme awareness in children with DS but also with phoneme awareness when the phoneme being targeted was in the word-final position. This was not the case for targeted phonemes that were in word-initial position. As grammar and tense also often rely on attending to word endings, it has been suggested that this proposal may also assist in at least partly explaining some of the observed impairments within this domain (Laws & Bishop, 2003; Perovic, 2006). Understanding the development of PA in children with DS has several important implications and considerations should be given to teaching, assessment and interventions for those with DS.

6.1.4 Phonological Awareness in Bilingual Children with Down Syndrome

Given that bilingualism research reports that PA is enhanced in bilinguals in comparison to TD children, and the marked deficits that are observed in children with DS, it is logical to question how PA will develop in bilinguals with DS. In a single case study which investigated reading and PA in a Russian-English bilingual child with DS (Burgoyne et al., 2016), the authors noted that although she displayed reading skills and speech at a rate that was expected in comparison to the developmentally matched TD children, the participant did not show any enhancements in measures of PA. Unusually, performance on PA in Russian was relatively higher than that of the participant's English PA abilities. It is possible to propose that the lack of a bilingual advantage for the individual in this study may be due to the fact that Russian and English do not share the same alphabet system. Alternatively, in line with the Cross-Language Transfer Hypothesis reported on earlier, as English was the L2 and is more complex and less orthographically transparent, an emerging benefit in PA in this specific situation would not be expected. It should also be noted that the participant was specially selected to participate in this study and she had previously received a reading intervention program that may explain her competent reading abilities and her uneven profile of PA in Russian and English.

Finally, in an early case study of a 23-year old trilingual adult with DS, although the aim was to document her language abilities in Italian, English and French, it emerged that she displayed unusually high PA abilities for an adult with DS (Vallar & Papagno, 1993). It was

proposed that multilingualism was the reason behind these surprising findings. Given that Italian and French are fairly transparent in their orthographies, the high degree of lexical similarities, and the simplicity and consistency of phonological structures between these languages, theories behind the bilingual benefit would explain why these well-developed PA abilities were observed in this context. Although it is intriguing to hypothesise the nature of these reported benefits in bilinguals with DS, it is imperative to recognize that this is a single case study with no control group. Nevertheless, this area warrants further investigation.

If such an advantage is observed for TD bilingual children, specifically in Welsh-English bilinguals as reported by Spencer and Hanley (2003), a compelling argument can be made to propose that Welsh-English bilingual children with DS may also experience more successful PA acquisition if they receive exposure to a bilingual environment. Alternatively, bilingual children with DS may develop PA in a distinctive or divergent way to TD bilinguals. Any emerging benefits, interactions or interference effect will have crucial significance to theoretical understanding and practical implications on how to approach teaching, assessment and interventions for this ability in this population specifically.

6.1.5 Current Study

To date, no single study has investigated PA in an empirical group study of bilingual children with DS. This study aims to undertake a comprehensive assessment of the PA abilities in Welsh-English bilingual children with DS. This research will identify PA abilities and developmental profiles in both English and Welsh in comparison to developmentally matched monolinguals with DS as well as similarly matched TD bilinguals and monolinguals. Given that PA abilities have been reliably shown to predict later reading and spelling development in both TD children and children with DS, it is imperative to understand how this skill emerges in these bilingual children. As children with DS are reported to display significant challenges with PA to begin with, identifying any differences in the development for bilinguals with DS will have substantial educational implications. This is particularly important within increasingly multilingual communities, specifically in Wales with initiatives to increase the number of Welsh speakers and the drive towards inclusivity for children with DS in mainstream schools.

The focus of this chapter is to answer the following research questions:

- I. How does phonological awareness develop in bilinguals and monolingual with Down syndrome?
- II. How does bilingualism impact phonological awareness in children with Down syndrome compared to typically developing bilingual children?
- III. Does the developmental trajectory of phonological awareness differ for bilingual children with Down syndrome?

6.2 Method

Information relating to the detailed methodology is provided in Chapter 4. For the current chapter, methods relating to the PA tasks only are relevant. In summary, novel tasks were designed to assess PA which included detection and production tasks, which aimed at identifying both implicit and explicit awareness of the three levels of PA under study; rhyme, syllables and phonemes. The tasks were designed specifically for the target populations recruited for this research whilst also considering recommendations from previous studies conducted with similar populations. For example, forced-choice tasks were used for implicit tasks as these have been used successfully with children with DS previously (Carroll et al., 2003; Gombert, 2002). Assessments were conducted in Welsh and English for the bilingual groups and in English only for the monolingual groups. Statistical analyses were undertaken to ensure that the Welsh and English stimuli were comparable (see section 4.1.2. for detailed information) in terms of the number of syllable ($p = .143$), letters ($p = .410$) as well as imageability ($p = .103$) and word frequency ($p = .143$).

6.2.1 Participants

As before, two participant groups (individually matched sample and the group matched sample) are used in this analysis, as described above in participant section 4.2.1, thus presenting a smaller sample with a more stringent matching process and a larger sample which provides greater statistical power. For clarity, a summary of the group characteristics is again presented below in Table 21 and 22.¹⁰ Further individual participant characteristics are provided in the appendix, item 9.

6.2.2 Procedure

Assessments were completed in a predefined order which was designed from larger, simpler units (rhyme) to smaller units (phonemes) as the tasks progressed. This procedure was designed with the aim that the initial tasks would be relatively easy for the participants and they would increase in difficulty as the assessments continued. Implicit tasks assessing awareness of each component were administered before the productive tasks for each component of PA. Each task had 2 practice items and 10 test items. As there were two tasks for each component as described above, the ceiling score was 60 for each of the English and Welsh PA assessments.

¹⁰Non-verbal IQ represent raw scores on the non-verbal matrices subtest of the KBIT-II (Kaufman & Kaufman, 2014). Socioeconomic status was obtained via parent report in terms of parental education and occupation (scale from 2-14). ^a Indicates a between-group effect of syndrome with $p < .05$. ^b Indicates a between-group effect of language status with $p < .05$.

	Down Syndrome		Typically Developing	
	Bilingual (n=10)	Monolingual (n=10)	Bilingual (n=10)	Monolingual (n=10)
Age in months ^a	114.2 (37.7)	112 (31.9)	51.6 (17.4)	50.7 (16.5)
Non-verbal IQ	9.6 (6.5)	10.7 (6.3)	10 (6.2)	9.5 (6.0)
SES	10.9 (1.7)	9.9 (2.6)	10.8 (2.0)	10.6 (3.1)
Current Welsh exposure in % ^b	49.1 (24.9)	5.01 (9.4)	57 (21.6)	12.8 (14.9)
Lifetime Welsh exposure in % ^b	50 (24.0)	2.79 (4.4)	45.8 (24.1)	8.3 (9.9)

Table 21. Group characteristics for individually matched participants. Mean scores are reported with SD in parenthesis.

	Down Syndrome		Typically Developing	
	Bilingual (n=10)	Monolingual (n=15)	Bilingual (n=25)	Monolingual (n=18)
Age in months ^a	114.2 (37.7)	114.9 (35.6)	51.04 (16.3)	55.4 (16.7)
Age of first word in months ^a	24.9 (2.27)	21.7 (11.2)	11.5 (3.4)	10.4 (2.5)
Non-verbal IQ	9.6 (6.5)	10.9 (6.3)	11.4 (5.2)	12.4 (6.55)
SES	10.9 (1.7)	10.5 (2.4)	11.0 (1.9)	11.2 (2.9)
Current Welsh exposure in % ^b	42.1 (25.1)	3.5 (7.9)	51.5 (24.8)	8.5 (10.3)
Lifetime Welsh exposure in % ^b	50 (24.0)	2.14 (3.6)	47.5 (27.87)	7.7 (9.1)

Table 22. Group characteristics for the group matched participants. Mean scores are reported with SD in parenthesis.¹¹

6.2.3 Data Analysis

Raw scores from the PA assessments were used in all analyses. This enabled comparisons between groups, between each component (rhyme, syllable and phoneme tasks) and also between languages (Welsh and English). A series of two-way analyses of variance (ANOVAs) and multivariate analyses of variance (MANOVAs) were conducted to examine the effect of language status (monolinguals versus bilinguals) and diagnosis (DS versus TD). Any significant

¹¹Data is missing for 5 children from the TDB group for SES, parent reported current exposure to Welsh and parent reported lifetime exposure to Welsh due to non-return of the parental questionnaire. Age of first word missing data for 20 participants due to non-return of questionnaire and parents not being able to recall this information.

effects were followed up using Bonferroni corrections to decrease the familywise error rate given multiple comparisons or simple main effects analyses where appropriate.

6.3 Results

6.3.1 Phonological Awareness - Individually Matched Sample

A summary of the descriptive statistics for the PA results is presented in Table 23 for each condition (syllable, rhyme and phoneme) as well as the individual sub-tasks within each condition.¹² Analyses also confirmed no differences based on gender $t(38) = 0.363$, $p = .719$ and subsequently gender was not included in any further analyses.

Phonological Awareness Component	Component Sub-task	Down Syndrome		Typically Developing	
		Bilingual	Monolingual	Bilingual	Monolingual
Syllable	SA	6.6 (2.07)	6.0 (2.91)	6.9 (1.73)	4.4 (2.72)
	SM	0.2 (0.63)	1.0 (2.16)	3.1 (3.98)	2.0 (3.23)
	Total	6.8 (2.39)	7.0 (4.14)	10 (5.31)	6.4 (5.66)
Rhyme	RA	4.6 (2.50)	5.7 (2.26)	4.9 (2.02)	4.9 (2.02)
	RP	0.4 (0.97)	0.6 (1.90)	0.6 (1.90)	1.4 (2.99)
	Total	5 (3.20)	6.3 (4.14)	5.5 (5.31)	6.3 (5.66)
Phoneme	PA	5.9 (3.51)	5.6 (2.76)	6.7 (3.47)	4.4 (3.41)
	PM	4.1 (3.60)	3.4 (3.86)	3.4 (4.01)	2.7 (4.14)
	Total	10.0 (5.79)	9.0 (5.87)	10.1 (6.90)	7.1 (7.31)
Overall Total		21.8 (7.52)	22.3 (12.98)	25.6 (13.7)	19.8 (16.67)

Table 23. Mean raw scores for English phonological awareness measures in English (individually matched sample) with SD in parenthesis

Between-group analyses were conducted for each component (syllable, rhyme and phoneme) for language status and diagnosis. No effect of language status, diagnosis or any interaction was found. As chronological age varied across groups, this was entered as a covariate in this model. After entering the covariate in the model, a significant effect of diagnosis emerged for syllable $F(1, 35) = 6.573$, $p = .015$, $\eta_p^2 = .158$, rhyme $F(1, 35) = 6.671$, $p = .014$, $\eta_p^2 = .160$, phoneme $F(1, 35) = 13.757$, $p = .001$, $\eta_p^2 = .282$ and overall PA $F(1, 35) = 12.756$, $p = .001$,

¹² Component sub-tasks are as follows: SA = Syllable Awareness; SM = Syllable Manipulation; RA = Rhyme Awareness; RP = Rhyme Production; PA = Phoneme Awareness; PM = Phoneme Manipulation.

$\eta_p^2 = .282$. Language status did not result in any significant effect, nor did any interaction effect emerge.

Analyses were conducted to further explore the significant effects of diagnosis on each sub-task. An effect of diagnosis was found for the syllable manipulation sub-task $F(1, 36) = 4.846, p = .034, \eta_p^2 = .119$ where the TD groups outperformed the DS groups. Chronological age was included as a covariate as before which displayed a significant effect of diagnosis in favour of the TD groups for all sub-tasks except the syllable awareness sub-task. Further analysis found a significant effect of language status for the syllable awareness sub-task $F(1, 36) = 4.164, p = .049, \eta_p^2 = .104$ whereby the bilinguals outperformed the monolinguals. Pairwise comparisons revealed that this was significant for the TD bilingual group only, ($p = .026$) not the DS bilingual group ($p = .580$). This effect remained after controlling for CA ($p = .047$). No interactions were significant. A summary of the results across syllable, rhyme and phoneme for all groups is presented in Figure 12.

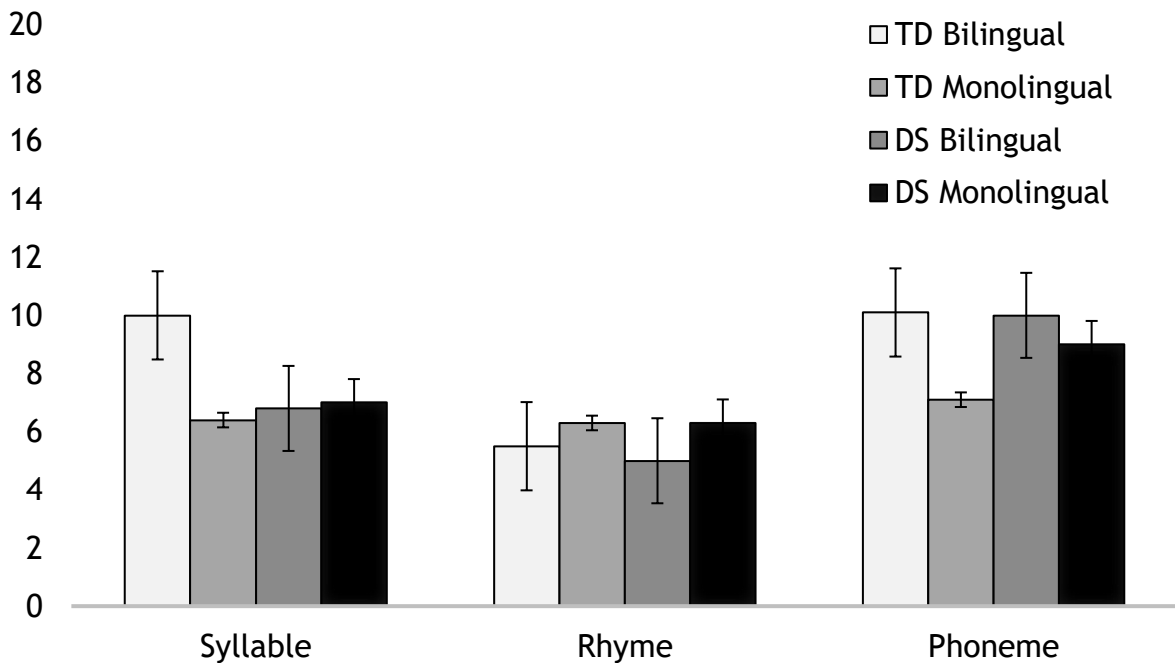


Figure 12. Mean raw scores on phonological awareness assessments for rhyme, syllable and phoneme levels distributed by diagnosis and language status. *Note:* Maximum raw scores for each condition is 20.

Multivariate analyses were conducted to compare performance between the conditions (rhyme, syllable and phoneme) for all groups which displayed an effect of condition $F(2, 35) = 8.515, p = .001, \eta_p^2 = .327$. Separate analyses for the DS and TD groups showed an effect of condition for the DS groups only $F(2, 17) = 6.874, p = .006, \eta_p^2 = .447$ but not the TD groups $F(2, 17) = 3.203, p = .066, \eta_p^2 = .0274$. Post-hoc comparisons were performed following the significant result for the DS groups which found that phoneme scores were significantly higher than rhyme scores ($p = .005$). No differences were found between performance on syllable

and rhyme conditions ($p = .297$) or for syllable and rhyme conditions ($p = .109$). No significant interaction was found for PA condition and language status for any group.

Further analyses were conducted for the bilingual groups on the Welsh PA measures (Table 24). A between-group independent samples t-test found no difference for overall PA in Welsh $t(18) = -.066, p = .948$ for the DSB group ($M = 26.6, SD = 8.00$) and the TDB group ($M = 27.0, SD = 17.46$). No differences emerged for the three components with syllable (TDB: $M = 7.4, SD = 4.77$, DSB: $M = 7.5, SD = 2.88$) and $t(15) = .057, p = .955$, rhyme (TDB: $M = 6.9, SD = 5.99$, DSB: $M = 5.6, SD = 2.76$) with $t(13) = -.623, p = .544$ and phoneme (TDB: $M = 10.4, SD = 7.57$, DSB: $M = 13.5, SD = 4.72$) with $t(15) = 1.098, p = .289$. A mixed-design ANOVA was also performed with diagnosis as the between-group variable and scores on Welsh and English PA as the within-participants variables. No significant differences were found between performance on the English assessments compared to Welsh for syllable $F(1, 18) = .175, p = 0.681$, rhyme $F(1, 18) = .843, p = 0.371$ or phoneme $F(1, 18) = 2.208, p = 0.155$. No interactions between language and diagnosis were significant. A summary of the bilingual group's performance for the English, Welsh and average PA is presented in Figure 13.

Phonological Awareness Component	Component Sub-task	Down syndrome	Typically Developing	<i>P</i>	Effect Size (η_p^2)
Rhyme	RA	5.4	5.1	.802	.004
	RP	0.2	1.8	.156	.108
	Total	5.6	6.9	.541	.021
Syllable	SA	6.9	7.0	.903	.001
	SM	0.6	3.0	.076	.164
	Total	7.5	10.0	.187	.095
Phoneme	PA	7.1	6.5	.667	.011
	PM	6.4	3.9	.122	.128
	Total	13.5	10.4	.287	.063
Overall		26.6	27.3	.948	.000

Table 24. Mean raw scores for phonological awareness measures and significant effects for Welsh assessments (individually matched sample).

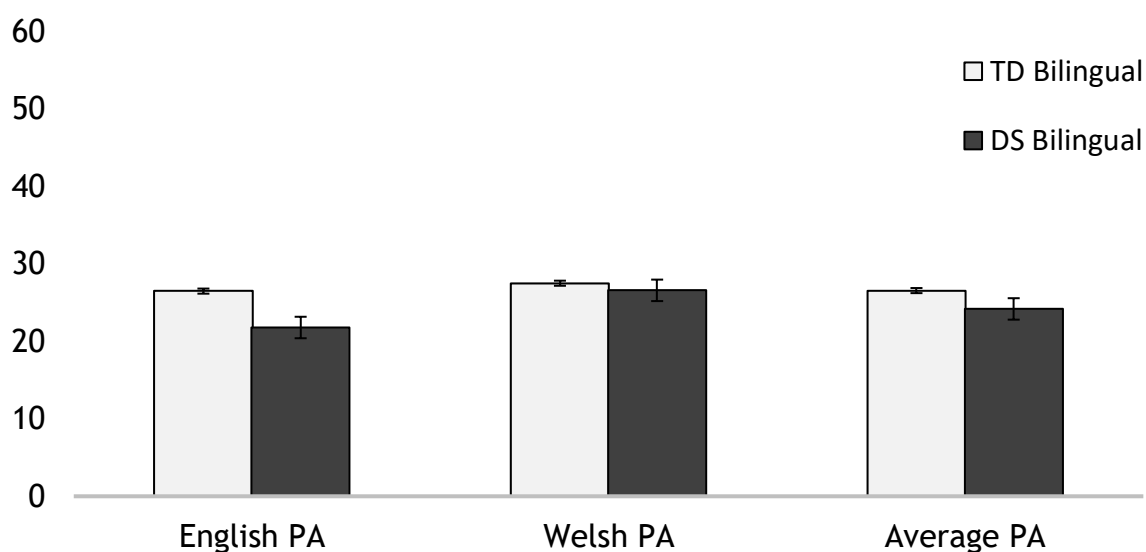


Figure 13. Mean raw scores on English, Welsh and average scores on phonological awareness for the bilingual groups.

6.3.2 Phonological Awareness - Group Matched Sample

A total of 68 children were included in the group matched sample (Table 15). Group performance on the PA tasks (including each condition and sub-tasks within each condition) is presented in Table 25. Analyses again confirmed that there were no differences in gender, $t(64) = 0.9, p = .928$ and are not reported on further.

Phonological Awareness Component	Component Sub-task	Down Syndrome		Typically Developing	
		Bilingual	Monolingual	Bilingual	Monolingual
Syllable	SA	6.6 (2.06)	6.13 (2.64)	7.46 (2.26)	5.24 (2.91)
	SM	0.2 (0.63)	1.93 (3.26)	2.25 (3.44)	3.0 (3.98)
	Total	6.8 (2.40)	8.06 (4.98)	9.71 (4.35)	8.24 (6.39)
Rhyme	RA	4.6 (2.50)	5.6 (2.50)	4.92 (2.45)	6.24 (2.59)
	RP	0.4 (0.97)	1.6 (2.97)	1.25 (3.07)	2.24 (3.38)
	Total	5 (3.20)	7.2 (4.69)	6.17 (5.07)	8.47 (5.46)
Phoneme	PA	5.9 (3.51)	6.07 (2.89)	7.0 (3.22)	6.35 (3.62)
	PM	4.1 (3.60)	3.8 (3.78)	3.62 (3.70)	2.59 (3.76)
Overall Total		21.8 (7.52)	25.1 (14.38)	26.5 (13.86)	25.65 (16.84)

Table 25. Mean raw scores for English phonological awareness measures in English (group matched sample) with SD in parenthesis.

A series of ANOVAs were conducted for each of the three components of PA (syllable, rhyme and phoneme). Including the covariate showed significant effects for diagnosis on syllable $F(1, 61) = 19.990, p < .001, \eta_p^2 = .247$, rhyme $F(1, 61) = 25.443, p < .001, \eta_p^2 = .294$ and phoneme $F(1, 61) = 22.890, p < .001, \eta_p^2 = .273$ scores. Language status was not significant for any component, although the rhyme condition approached significance $F(1, 61) = 3.995, p = .05, \eta_p^2 = .061$. No interactions were found. Follow up analyses identified that the DS groups performed significantly lower than the TD groups on all subtests ($p < 0.005$) besides the syllable awareness subtest ($p = .052$). For language status, a significant effect was found for the syllable awareness subtest only $F(1, 61) = 5.101, p = .028, \eta_p^2 = .077$, in favour of the bilingual groups. Pairwise comparisons showed that this significant effect of language status on syllable awareness was evident for the TD group ($p = .003$) but not for the DS group ($p = .620$). No interactions were significant. Results for the three components of PA are summarised in Figure 14.

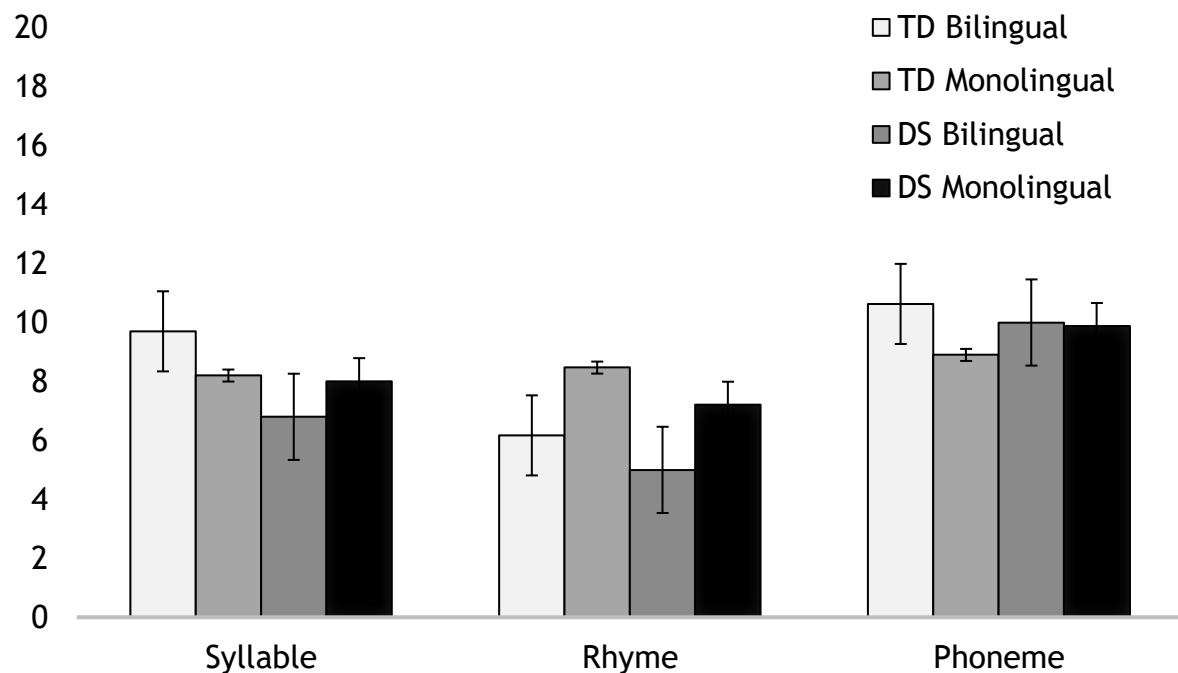


Figure 14. Mean raw scores on phonological awareness assessments for rhyme, syllable and phoneme levels distributed by diagnosis and language status. *Note:* Maximum raw scores for each condition is 20.

Multivariate analyses were conducted to identify differences in performance for the three conditions (rhyme, syllable and phoneme). Performance for all participants showed a significant effect of condition $F(2, 64) = 12.460, p < .001, \eta_p^2 = .280$. Follow up analyses were conducted with the DS and TD groups as a result of the effect of condition. An effect of

condition was found for both the DS groups $F(2, 23) = 6.365, p = .006, \eta_p^2 = .356$ and the TD groups $F(2, 39) = 6.171, p = .005, \eta_p^2 = .240$. Post-hoc comparisons for the DS groups found that performance on the phoneme tasks was significantly better than performance on the rhyme tasks ($p = .005$). No differences were found between performance on syllable and rhyme conditions ($p = .238$) or for phoneme and rhyme conditions ($p = .096$). For the TD groups, their performance was significantly higher on the phoneme ($p = .004$) and syllable conditions ($p = .033$) in comparison to the rhyme condition. No differences were found in performance between syllable and phoneme conditions for the TD groups ($p = .721$). No interaction between PA condition and language status was found for the DS groups, however, the interaction was significant for TD groups $F(2, 38) = 4.515, p = .017, \eta_p^2 = .192$.

Performance on the Welsh PA measures (Table 26) was not significantly different for PA overall between the TDB ($M = 27.95, SD = 16.98$) and DSB groups ($M = 26.60, SD = 8.00$) with $t(30) = -.307, p = .761$. One sub-task showed a significant effect between the TDB and DSB group, the syllable manipulation task where the DSB group performed lower than the TDB group $t(30) = -2.142, p = .040$. A mixed-design ANOVA with diagnosis as the between-group variable and language as the within-participants variable (Welsh vs English) found no significant effect of language $F(1, 30) = 3.763, p = .062, \eta_p^2 = .111$. The interaction between language and diagnosis was not significant $F(1, 30) = 1.818, p = .188, \eta_p^2 = .057$. A summary of the bilingual group's performance for the English, Welsh and average PA is presented in Figure 15.

Phonological Awareness Component	Component Sub-task	Down syndrome	Typically Developing	<i>P</i>	Effect Size (η_p^2)
Rhyme	RA	5.40	5.55	.889	.001
	RP	0.2	2.05	.104	.085
	Total	5.6	7.59	.314	.034
Syllable	SA	6.9	7.32	.548	.012
	SM	0.6	3.23	.040	.133
	Total	7.5	10.55	.083	.097
Phoneme	PA	7.1	6.14	.446	.019
	PM	6.4	3.68	.059	.114
	Total	13.5	9.92	.161	.064
Overall		26.6	27.95	.813	.002

Table 26. Mean raw scores for phonological awareness measures and significance level for Welsh assessments (group matched sample).

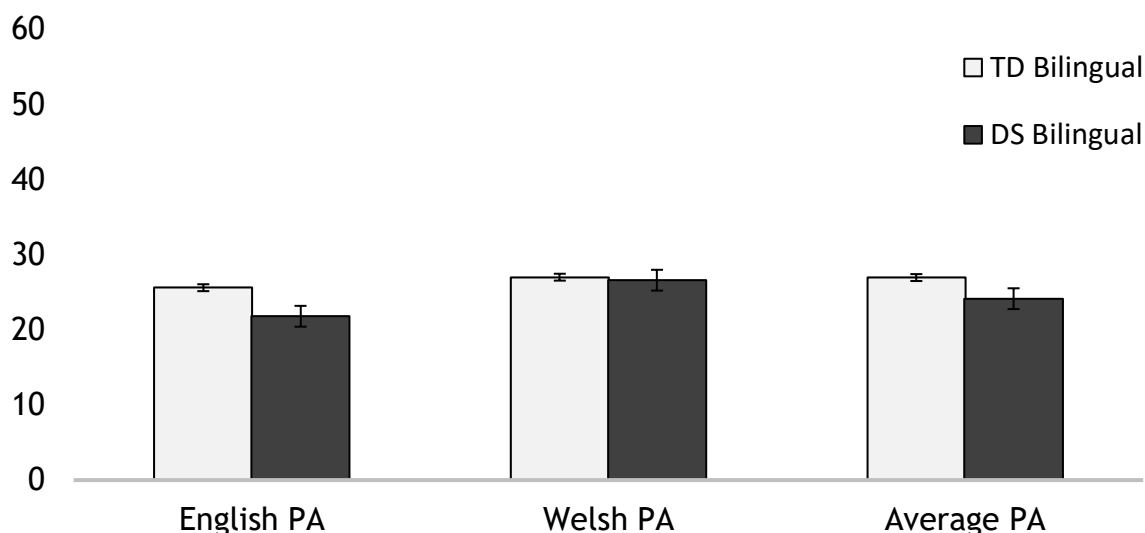


Figure 15. Mean raw scores on English, Welsh and average scores on phonological awareness for the bilingual groups.

6.4 Discussion

The purpose of this research was to examine the PA abilities in Welsh-English bilingual children with DS in comparison to monolinguals with DS and TD control groups. Given that bilingualism has been reported to accelerate the development of PA and the marked difficulties that children with DS have in regards to PA specifically, this research sought to identify how bilingualism impacted the development of PA in children aged between 5-16 with DS. The crucial role of PA on later reading and spelling development means that identifying the emergence of PA within this population is fundamental to educational provisions within these populations. Alongside the increasing bilingual and multilingual speakers and communities, especially in Wales, this research has substantial implications on policy, clinical practice and educational provision.

6.4.1 Phonological Awareness in Bilinguals with Down Syndrome

Findings of this study reveal that overall, the bilingual groups performed similarly to the monolingual groups on overall PA. This was true for both the individually matched sample and the group matched sample. Results show that bilingual children with DS develop PA in a similar way to monolinguals with DS and show the same profiles of development with a marked deficit in rhyme awareness and production. Similarly, the TD bilingual children displayed equivalent skills to the monolingual TD children for the majority of the PA assessments. Evidence for a bilingual advantage was only found for the TD children for the syllable awareness task. An effect of diagnosis was found in that the DS group performed lower than the TD groups, as would be expected given the previous research that documents

a specific deficit in this domain for children with DS. This was most pronounced for the syllable manipulation sub-task, although after considering CA in the analysis, the DS groups showed impairments compared to the TD groups for all tasks besides the syllable identification sub-task. Both bilingual groups showed comparable abilities in Welsh and English PA. Findings from the individually matched sample and the group matched sample show very similar results which demonstrates that the findings are valid and reliable.

Given that children with DS show deficits in the development of PA, it is plausible to suggest that any potential negative impact of bilingualism would be most evident in areas of development such as PA. Findings show that PA is no more compromised in bilinguals with DS in comparison to monolinguals with DS, highlighting the capacity for children with DS to acquire two languages and show measurable abilities of PA in both languages. In contrast, no additional adversities were observed on the development of PA in bilinguals with DS. Findings are in line with a specific deficit in PA for children with DS (Cossu, Rossini & Marshall, 1993; Fletcher & Buckley, 2002; Næss et al., 2015; Næss, 2016). Given the crucial role of PA on later reading and spelling development in both TD and DS populations (Fletcher & Buckley, 2002), the finding that PA is comparable in bilinguals and monolinguals with DS has implications on educational provisions which are discussed later. Furthermore, bilingualism may have impacted the reading abilities of the children under study, however, this was not directly assessed in this research. Reading abilities were outside the scope of the current study, however, this warrants further investigation in future research.

All groups displayed the greatest performance on the syllable and phoneme tasks and lower performance on the rhyme assessments. This was particularly true for the bilingual groups who had a larger discrepancy between phoneme and rhyme tasks than the monolingual groups. For DS groups, this supports previous research that documents a specific weakness in rhyme abilities (Cardoso-Martins et al., 2002; Kennedy & Flynn, 2003; Hulme et al., 2012). This also provides support for the proposal that children with DS have a specific deficit in attending to elements of language which are in word-final position (Næss, 2016). In previous research, it has been reported that participants with DS had marked difficulties with rhyme and final phoneme trials but not initial phoneme or syllable tasks (Snowling et al., 2002). This proposition could also explain the findings reported here with the identification of rhymes and attending to final syllables being particularly compromised for both DS groups. Furthermore, this suggestion may also assist in partly explaining some of the deficits which have also been observed in expressive and receptive morphosyntax. Applying appropriate grammatical inflections often relies on attending to word-endings in English and Welsh and consequently this suggestion may explain some of the specific deficits reported here between the TD and DS groups for expressive morphosyntax and PA.

In considering metalinguistic awareness, previous research has proposed that the development of PA in bilinguals is accelerated due to an automatic increase in implicit and explicit attention to word forms and more fine-grained representations of language as a result of receiving input to two languages (Bialystok, Majumder & Martin, 2003; Verhoeven, 2007).

Some evidence to support this was found for syllable awareness, however, this was only true for the TD children. Bilingual TD children outperformed the TD monolingual children on the task that required the children to detect syllables only. No other evidence was found to support the proposal of a bilingual benefit for any of the other tasks, nor on the working memory task as measured by a forward digit span. It is important to note that “an absence of evidence is not evidence of absence” (Altman & Bland, 1995), however, there are some plausible reasons why no effect of language status was found within this context specifically.

Firstly, in line with the Cross-Language Transfer Hypothesis, the order of acquisition of the two languages may have impacted the findings in this study. The Cross-Language Transfer Hypothesis postulates that there will be ‘carry-over’ effects from the L1 to the L2 if the languages share linguistic features and that the linguistic features are more complex in the L1. According to this theory, if the second language that is being acquired has more complex phonological structures, then the proposed bilingual advantage would not emerge in this specific bilingual situation. As Welsh and English have some different linguistic features (particularly in terms of opacity) and English is more phonologically complex, this may result in differences dependent on whether Welsh or English is considered to be the first or second language. If English is acquired first, according to this theory, a bilingual advantage would emerge as carry-over effects from the more complex language (English) would subsequently transfer to the phonologically simpler language (Welsh). This would result in a positive transfer of phonological properties, thus enhancing PA. In contrast, if children acquire Welsh first followed by English, then the opposite would be expected (i.e. null effect of bilingualism). As the majority of the bilingual children in the sample for the current study were L1 Welsh, this may explain the lack of an effect of language status. Therefore, further research that differentiates between the order of the languages acquired is required in order to disentangle these effects.

Furthermore, researchers investigating bilingualism in populations of Welsh-English bilingual children and adults have found mixed results with some studies failing to replicate research which supports the bilingual advantage hypothesis for executive functioning and metalinguistic abilities (Gathercole et al., 2014). The authors of these studies have suggested that the particular linguistic context in Wales may result in enhanced automaticity in linguistic processing between Welsh and English. Within fully bilingual communities in Wales, Welsh and English are often used interchangeably which may consequently require less cognitive control, monitoring and inhibition than within other bilingual settings. It is suggested that in these fully fluent bilinguals, no superior performance on tasks assessing cognitive control was found as their two languages are both “on-line” at all times and speakers are not required to monitor their linguistic surroundings to the same extent within these fully bilingual communities. This proposal may also be relevant for explaining why only limited evidence was found for a bilingual advantage in the current study. If Welsh and English are often used alongside each other, children may not be required to rely on identifying patterns of speech sounds to differentiate between Welsh and English.

The only available evidence that has been conducted in terms of PA in Welsh-English bilingual children specifically was conducted by Spencer and Hanley (2003). In this study, it was reported that the children who are learning Welsh outperformed children who were learning English only on a phoneme awareness task and at reading words and non-words. It was proposed that the differences between groups were due to the fact that the children who were learning Welsh were learning a language with a transparent orthography, whereas those who were learning English were not. The authors also consider the fact that bilingualism may have resulted in this effect, although they propose that the transparent orthography was the most likely factor due to the finding that there was not a larger between-group effect when the children were older and would be considered to be 'more bilingual'. Spencer and Hanley suggest that the children learning to read Welsh had consequently adopted a different strategy for word recognition compared to the children learning English. The bilingual children in this study were all L1 Welsh speakers, which may explain why an advantage in favour of the bilinguals was observed in this study.

An alternative explanation may be that the developmental ages of the children in the current study may not have reached a point to which an advantage on the PA tasks would be expected. Duncan and colleagues (2009) proposed that such advantages on PA would only emerge in children with chronological ages above 4 years old. As many of the children were three years old, it may not be surprising that only limited evidence of a bilingual advantage was found. In addition, many of the children with DS also had developmental ages below 4 years old, which may also explain why no evidence of a bilingual advantage emerged in this population. Given that a bilingual advantage was detected in the TDB group for syllable awareness, this may be evidence that a bilingual advantage may emerge in this population as the chronological ages and developmental ages increase, given that typically, syllable awareness is the first component of PA to be acquired (Caravolas & Bruck, 1993; Fox, & Routh, 1975; Treiman & Zukowski, 1991). Similarly, Bialystok and Barac (2012) proposed that a certain level of proficiency is required in both languages in order for metalinguistic advantages to emerge. Subsequently, these young children may not be "proficient enough" in their languages for an advantage on PA to have developed at this stage.

A further explanation to be explored in future research is that children with DS may have adopted different strategies of reading and decoding to the TD children. Previous research has suggested that children with DS may employ a sight-reading approach initially if they are not provided with phonics-based reading instruction (Cossu & Marshall, 1990; Fowler et al., 1995). This may explain why a syllable awareness advantage was discovered for the TD group but not the DS groups. If the participants under study had adopted whole sight-reading approaches, it would not be surprising that no bilingual advantage emerged for this group, although the majority of research to date now supports the suggestion that PA does play a central role in reading abilities in those with DS (Cupples & Iacono, 2002; Kennedy & Flynn, 2003; van Bysterveldt, Gillon & Moran, 2006), and therefore this proposal seems unlikely. Finally, a lack of statistical power may have lowered the potential to identify meaningful

differences between the groups, particularly due to the variation within groups which is often characteristic of research in individuals with DS. Including two samples, one being a larger sample intended to increase these effect sizes, however, this factor cannot be dismissed.

It is also possible that the development of PA in children with DS is fundamentally different from the way in which PA develops in TD children. Consequently, the underlying mechanisms responsible for PA which are impacted by bilingualism may be different for children with DS, which would explain why no evidence of a bilingual advantage was found. This finding would then substantiate previous findings where no bilingual advantage was found in a case study of a bilingual child with DS (Burgoyne et al., 2016) or a group study of bilingual children with ASD either (Pereda, 2013). Both of these studies sought to identify if there was any evidence of a bilingual advantage for PA in their respective samples, but neither documented any enhanced performance of bilingualism. Although the small sample sizes could again be responsible for these findings, an alternative explanation could be that the advantages found in TD children do not extend to children with DDs. Crucially, this study and the small-scale previous work has not found any disadvantages in this area. Given that those with DS have marked difficulties in the development of PA, the finding that this area is not further impaired is an important finding.

One further interesting finding that was found in the current study was that there was a larger discrepancy between the rhyme and phoneme components for the bilingual groups. This interaction between bilingualism and the PA component was significant for the TD children, suggesting that bilinguals may be developing PA differently to the monolinguals who did not show such large differences in performance. All groups had the highest performance for the phoneme tasks (which may be due to task effects), however, the bilinguals consistently had significantly higher phoneme than rhyme scores which was evident for the TDB group specifically and both bilingual groups when considered together. It is intriguing to note that this finding was also reported by Spencer and Hanley (2003) who also studied Welsh-English bilingual children and found that the English monolingual children were consistently better at the rhyme tasks (although not significantly so in either study). It was proposed that this was due to orthographic differences between Welsh and English as rhyme may be more of a salient feature for children learning English. If children learning Welsh were applying grapheme to phoneme rules in English, this may lead to these children pronouncing words incorrectly and may explain why children learning Welsh had greater difficulties with rhyme assessments but displayed greater performance than the English monolinguals on the phoneme tasks.

In summary, bilingualism is a concept that is complex and multifaceted. The complexity and the number of variables to consider in researching this phenomenon in children with DS becomes even more multidimensional. Overall, both DS groups showed similar profiles of PA with them displaying marked deficits in PA as anticipated. Furthermore, both groups of bilingual children performed comparably to the groups of monolingual children for the most part. An advantage in favour of the bilinguals was found for syllable

awareness in the TD children only. Some possible explanations in relation to the lack of a bilingual advantage in this population may be due to the specific linguistic situation in Wales, the order of languages acquired or the developmental and chronological ages of the children in this study. Crucially, we find no evidence that the bilinguals performed any worse than the monolinguals. As children with DS show specific impairments in this domain, any negative impact of bilingualism may be best observed in areas of weaknesses, however, no further impairments were found. Findings reveal that bilinguals with DS also show similar patterns to monolinguals with DS with similar areas of weakness and relative strengths.

6.4.2 Limitations and Implications

The first limitation to consider is that the sample sizes were relatively small and employed a large age range. This may have led to difficulties in identifying group differences in the current population. Future studies should employ matching principles with children who are closely paired on chronological age as well as developmental age, although the groups recruited in this study were also matched for age across groups. Research that explores individual differences may also be particularly informative and useful for the populations under study, given the large amount of individual variation that is often observed in children with a DD and children with DS specifically. Case studies may consequently be particularly insightful for exploring specific instances where individuals have shown distinct instances of well-developed language or PA abilities.

A second important potential constraint to consider is that participant's experiences with similar tasks to those designed for this study may have varied according to their speech and language therapy and their school's approach to teaching phonics and PA explicitly. This may particularly be the case for the groups of children with DS whereby engaging in game-like activities may often feature as part of their therapy schedules. It may, therefore, be possible that the children with DS may have had greater performance depending on their experiences with such tasks. Future studies may be able to control for this potential factor by using non-words which will not have been directly used in interventions previously, however, it is unlikely that it will be possible to eliminate this factor completely. Nevertheless, it is important to recognize that this may impact on the results of such studies investigating PA in a population such as those with DS.

One further potential limitation is the difficulty of the tasks designed for this research. Many of the TD children who were at the lowest end of the age range and some of the younger children with DS scored at floor for some of the tasks. This seemed particularly true for the rhyme level tasks and the rhyme production task specifically. The tasks were designed to include a range of tasks that varied in difficulty and consequently it was expected that the participants would have some difficulty with the tasks. In addition, these tasks were designed for the specific population at hand and adaptations were made to accommodate the needs of children with DS such as providing visual stimuli alongside spoken stimuli and enabling non-verbal responses where possible. The tasks were also designed in accordance with previous

research that investigated PA in children with DS specifically and consequently these tasks were considered appropriate for this population. To overcome potential floor effects in the future, researchers may select children older than 5 for assessing PA in those with DS. Some studies have also matched participants based on reading ability as opposed to developmental age and consequently, these children are usually slightly older (Cardoso-Martins, Michalick & Pollo, 2002; Snowling, Hulme & Mercer, 2002; Verucci, Menghini & Vicari, 2006). Given the range in language abilities in this population, however, selecting older children with DS may not necessarily eliminate some children having specific challenges with PA.

In terms of implications arising from this work, the primary meaningful outcome of this work is that bilinguals with DS are not at any disadvantage to the development of PA in comparison to monolinguals with DS. Given the fundamental role of PA on later reading and spelling development, this finding means that those with DS should not be discouraged against bilingual exposure or indeed bilingual education. Similar trajectories of development seem to be apparent for bilinguals and monolinguals with DS, however, both groups appeared to have substantial challenges with the tasks which assessed rhyme awareness and rhyme production. Consequently, interventions that explicitly target the development of this component may be successful in not only enhancing PA but also reading and spelling development, which in turn has been shown to increase further language abilities such as later MLU (Laws & Gunn, 2002) as well as vocabulary knowledge and grammar (Sue Buckley et al., 2007). All these elements may contribute to higher academic success which may provide better outcomes in adulthood such as the ability to live independently or semi-independently and obtain employment. As interventions in reading and PA have been successful for children with DS previously (Cologon, Cupples & Wyver, 2011), it would be anticipated that future interventions with bilinguals with DS will be equally beneficial, if not more so given the possibility of cross-language transfer and interaction effects.

6.4.3 Conclusion

In conclusion, this is the first group study of PA in bilingual children with DS. Given that PA poses a specific challenge for individuals with DS and the apparent bilingual advantage observed in aspects of metalinguistic awareness and more specifically PA, this research aimed to identify if bilinguals encounter further challenges to the development of PA by the addition of a second language, or whether any evidence of a bilingual advantage would emerge for this population. Findings reveal a specific language phenotype in terms of PA for the children with DS, which is in line with previous research, such as a specific weakness in rhyme awareness. No evidence of a bilingual advantage was observed in the DSB group, and only limited evidence was observed for the TDB group in comparison to the monolingual control groups. Importantly, no evidence was found to suggest that the bilinguals with DS were in any way disadvantaged in this aspect of development compared to the monolinguals with DS. Furthermore, similar developmental profiles were observed for these two groups. Given the crucial role that PA plays in later outcomes of reading and spelling for both those with DS and

TD children, this finding has important implications on educational provisions within these populations.

Future studies should endeavour to examine the role of PA in this population further by directly assessing reading and non-word reading alongside PA measures. Given the challenges posed in developing rhyme awareness for this population, intervention studies and longitudinal studies may provide pragmatic and constructive information as to how to develop these skills in these populations in the future. Identifying the developmental trajectory of PA may also be productive for individuals who work with those with DS in order to identify if an individual is having greater difficulties with this area than would be expected for monolingual and bilingual populations with DS. Subsequently, if interventions are found to be successful, these can be tailored to individuals who might benefit from them the most and will provide evidence-based practice as to how to stimulate the development of PA in this population further. Finally, longitudinal research that explores the possibility of cross-language transfer may be particularly useful to furthering the understanding of PA in bilinguals with DS, given the research that currently exists in this field for TD bilingual children.

Chapter 7: Findings – Bilingualism in Children with a Dual Diagnosis of Down Syndrome and Autism Spectrum Disorder¹³

7.1 Introduction

This chapter explores the unique language profiles of children who have Down syndrome (DS) and an additional diagnosis of an Autism Spectrum Disorder (ASD) who are in a bilingual setting. Researchers and practitioners have identified that a considerable number of children with a diagnosis of DS also meet the diagnostic criteria for ASD at a later stage in development. As both children with DS and ASD are known to have language impairments, the examination of how children with a dual diagnosis acquire more than one language will provide a further novel insight into the linguistic profiles of children within a bilingual environment. This chapter will identify how language and PA develop in a case by case analysis of four children with a dual diagnosis of ASD and DS. The profiles of the four children are presented here and compared in relation to bilingual and monolingual children with DS as well as TD bilingual children.

7.1.1 Prevalence of DS-ASD Dual-Diagnosis

Practitioners and researchers initially considered a dual diagnosis of ASD and DS as a rarity, with the suggestion that DS had some protective function against behaviours associated with ASD. For example, social interaction appears to be a relative strength in some children with DS and research reports that those with DS generally engage appropriately with others in social situations (Rosner et al., 2004). More recently, however, these claims have been questioned as has the validity of using the same screening measures for ASD with those who additionally have an intellectual disability as ASD may present itself differently within these populations (Kent et al., 1999). More recent large scale studies of individuals with DS have reported high rates of participants also reaching the diagnostic criteria for a diagnosis of ASD, particularly in those who have a more severe learning disability by means of greater impairments in general intellectual development and functioning (DiGuseppi et al., 2010; Warner et al., 2014).

Recent research now suggests that there is a considerably higher occurrence of ASD in children with DS than that of the general population with current estimations suggesting a prevalence rate of between 5-37.7% (DiGuseppi et al., 2010; Moss, Howlin & Oliver, 2012; Warner et al., 2014), compared to around 1.1%-2.24% in the general population (Brugha et

¹³ A modified version of this chapter is under review in the *Journal of Clinical Linguistics and Phonetics* entitled 'Bilingual Language Development in Children with a Dual Diagnosis of Down Syndrome and Autism Spectrum Disorder.'

al., 2012; Zablotsky, et al., 2015). In general, overall understanding and awareness of ASD has increased, and changes have been made in the diagnostic procedures and accuracies of diagnostic assessments (Blumberg, et al., 2013). This has led to an increase in the number of children receiving an appropriate diagnosis of ASD across all ages, as well as an increase in the number of children receiving an earlier diagnosis of ASD (Hertz-Picciotto & Delwiche, 2009). At the same time, more specified and accurate identification of children with DS who also have ASD has increased. A greater understanding of how ASD manifests itself in children with DS and the implications that this has on cognitive and linguistic development has informed recent research and practice within these populations.

Current estimates of children who have this dual-diagnosis (henceforth DS-ASD) suggest that between 5-37.7% of children with DS additionally have a diagnosis of ASD (DiGuseppi et al., 2010; Moss, Howlin & Oliver, 2012; Warner et al., 2014). As aforementioned, these figures are higher than the number of children within the overall population receiving a clinical diagnosis of ASD, suggesting that children with DS are more likely than TD children to have further difficulties associated with ASD. These difficulties as reported in the most recent edition of the diagnostic and statistical manual (DSM-5, American Psychiatric Association, 2013) relate to “persistent difficulties with social communication and social interaction” and “restricted and repetitive patterns, behaviours, activities or interests” which “limit and impair everyday functioning.” Many children with ASD also experience substantial language delays or remain non-verbal/minimally verbal (Tager-Flusberg & Kasari, 2013). Furthermore, there appears to be a particular language phenotype that is associated with children and adults with ASD (Tager-Flusberg & Caronna, 2007). Consequently, these additional difficulties with language in those with DS-ASD may have further implications in relation to bilingual populations.

Generally, a diagnosis of ASD in DS is received at a later age than in children without DS which could be due to the fact that language and general cognitive delays and impairments are known to be a part of DS also. Further language and cognitive delays may consequently not be recognised and assumed to be a part of the developmental trajectory of DS. Researchers have reported, however, that early identification of ASD in children with DS is important in order for these children to receive early access and provisions that are better suited to their needs. Those with DS-ASD are likely to need targeted educational support and tailored services with clinicians who are best suited to meet the needs of children with DS-ASD (Kent et al., 1999). In contrast, other researchers have raised concerns about attempting to diagnose ASD too early in children with DS. Buckley (2005) suggested that there may be a risk of finding false positive symptoms associated with ASD within populations of individuals with DS, due to the lower cognitive and intellectual development found in those with DS which may lead to an overdiagnosis of DS-ASD.

Given that there appears to be this unique phenotype of ASD in DS, this may lead to further difficulties for clinicians to formally diagnose DS-ASD. Consequently, Buckley (2003) suggests that families and clinicians should wait until a child with DS is aged 5 or 6 years old

until suggesting or seeking a formal diagnosis. Finally, Howlin (2000) similarly suggests that caution is warranted when considering and assessing any dual diagnosis of DS-ASD. She reports that it is fundamental to consider the level of cognitive functioning and language skills in a range of settings in order to appropriately decipher and understand any underlying comorbid diagnosis of ASD. As there are often overlapping symptoms associated with both DS and ASD, this may also make it more difficult to identify if the challenges that a child is displaying is due to DS alone, or if there is an underlying ASD. The delayed and often absent speech in infancy, poor hearing and reduced opportunities to develop social skills may also make identifying a possible dual DS-ASD diagnosis more complex.

This specific profile of development generally appears to result in additional language delays as both children with ASD and children with DS exhibit language delays and impairments. Consequently, it appears that having both of these DDs leads to even further language delays, however, there is a profile of development that appears to be specific to those with DS-ASD. Research in relation to this specific language and cognitive profile is discussed further below.

7.1.2 Language and Cognitive Profiles in Dual-Diagnosis

Researchers have noted that it is difficult to identify those with DS-ASD as there is a specific profile associated with this dual diagnosis which may not coincide with conventional assumptions and tendencies that are either associated with ASD or DS separately. This may subsequently be one of the reasons behind the disagreement and uncertainty of the true prevalence of DS-ASD. Researchers have, however, reported that there are further language and communication difficulties associated with a dual diagnosis. For example, Molloy et al. (2009) found that children with DS-ASD had increased difficulty with both receptive and expressive language abilities and that some children with DS-ASD also experienced a language regression, something which is usually associated with ASD but not DS. Castillo et al. (2008) also reported that children with DS-ASD in their study also experienced a language regression but that this was later in development than what would be expected in children with a single ASD diagnosis. Furthermore, a regression was found in children with DS-ASD in terms of overall development as opposed to just a language regression.

Warner et al. (2014) also noted that there appeared to be a specific phenotype associated with DS-ASD specifically in relation to language development. This was a large scale study with 499 children from England and Wales which explored autistic tendencies in children with DS compared to children with a diagnosis of ASD only. This research explored language characteristics within these groups and aimed to identify the number of children with DS who reached the typical cut-off point for a diagnosis of ASD. This was found to be very high with 37.5% displaying characteristics of ASD, and 16.5% reaching the criteria for autism. The majority of these were male at 67% for ASD and 75% of those reaching the diagnostic criteria for autism being male. In this research, it was reported that children with DS-ASD displayed fewer impairments relating to communication than children with ASD alone

but more impairments than those with just DS. These included impairments in the use of gestures, imitative social play, eye gaze and general shared interactions. There was one aspect found to be more greatly impaired than both children with a single diagnosis of ASD or DS, which was a greater tendency to display compulsions and more obsessive behaviour. All other measures assessed in terms of restricted, repetitive and stereotyped behaviour were found to be similar in both the DS-ASD and the ASD group.

In terms of speech development, Warner et al. (2014) also found that those with DS-ASD displayed lower levels of development with significantly more children at the single word level or below compared to those with DS. They were also less likely to communicate in sentences or phrases. In terms of the age of first word, children with DS-ASD reached this stage at a significantly later stage than the children with DS alone, with this stage being delayed on average an additional 6 months. Furthermore, the DS-ASD group displayed significantly more emotional symptoms, conduct problems, hyperactivities, peer problems and significantly less prosocial behaviour than the DS group. Compared to the ASD group, the children with DS-ASD displayed more impairments on pronoun reversal, and on the use of neologisms, however, the DS group also displayed difficulties with pronoun reversal, suggesting that this difficulty was associated with DS. The authors concluded that children with DS-ASD do show a specific phenotypic presentation of language which needs to be understood and considered when formulating and implementing tailored interventions.

Further research has emphasised the importance of the screening tools used to confirm or explore a possible comorbid diagnosis of ASD within children who have low levels of intellectual functioning (Capone et al., 2005). A recent study sought to evaluate the effectiveness of the aberrant behaviour checklist (ABC; Aman et al., 1985) and the DSM-IV in terms of its reliability in identifying children with DS-ASD (Capone et al., 2005). The researchers found a high occurrence of DS-ASD (in line with previous research) with a prevalence rate of 12.9% (although the authors acknowledge the possibility of a referral bias). As reported by Warner et al. (2014), a similar gender distribution was found with males being disproportionately affected with males accounting for 75% of those reaching clinical criteria for ASD. It is reported that scores on the ABC were able to explain a high proportion of the variability of autistic tendencies and, on the whole, it is reported to be one of the easiest and most reliable ways of identifying ASD in children with low levels of intellectual functioning, specifically in those with DS. This study concludes that the ABC can be a very useful tool that is feasible and reliable for use with children who have DS in making accurate diagnoses and identifying ASD traits.

This study does, however, express that caution is required in assessing language in relation to an ASD diagnosis, as many assessments use spoken language skills in their diagnostic criteria (including the ABC), which puts these children at a higher likelihood of ASD diagnosis due to specific difficulties with expressive versus receptive language associated with DS. Standardised assessments may not take these factors into account and should ideally be avoided if they are being used for assessment or diagnostic purposes in these populations. A

high degree of variability in language and cognitive outcomes in those with DS is also noted in this study and it is expressed that general low cognitive development should not be the only factor resulting in a DS-ASD diagnosis. Those with DS-ASD in the study showed impairments with both verbal and non-verbal language abilities, and this appears to be necessary for consideration of this dual diagnosis. The concluding remarks of the article question the need for a dual diagnosis and suggest that perhaps an additional diagnosis may not be useful for all children if they are already receiving targeted support.

Complementing this research is a further study that recruited 20 children with DS to be screened on a range of assessments which are typically used to identify ASD in otherwise TD children (Hepburn et al., 2008). The research aimed to identify autism symptoms in very young children and infants with DS. Comprehensive assessments were administered to these children at 2 years of age, and later at 4 years of age for the majority of the children (n=18). Of these, 3 children were considered to have ASD according to the diagnostic cut-off with a further 2 meeting the criteria for autism. The authors do raise concerns about using a single diagnostic tool as the ADOS-G was found to over-identify autism diagnoses in their sample of children with DS. It is suggested that misdiagnosis of autism may be particularly prevalent in young children with DS who display substantial language and communication deficits. To date, there is no formally, universally accepted tool for screening children with a dual diagnosis of DS-ASD.

In those children who have received a formal diagnosis of DS-ASD, there appears to be very limited information available about the clinical management of these children in terms of language and communication interventions. Researchers have highlighted that interventions used with children who have ASD and low intellectual development have been successful in managing behaviour and enhancing communicative abilities (Ben-Itzhak & Zachor, 2007). Limited information is available concerning children with comorbid disabilities in children with ASD such as DS. As highlighted in chapter 2.3, efforts have been made to identify and tailor interventions and strategies to enhance language development for children with DS, however, there is very little information about the development or assessment of language for children with DS-ASD. As a specific profile appears to exist for cognitive and linguistic development for those with this dual-diagnosis, information is needed which is specifically tailored to this group of children. Current research in this field tends to focus on clarifying prevalence rates of DS-ASD and identifying ways to formally assess and diagnose ASD in children with DS.

The largest study to date within this population specifically (n = 30) showed that, as with children with a single DS diagnosis, there is a large range in symptoms and the severity of impairments, however, there are two main sub-types of DS-ASD that have been identified (Medlen, 1999; Capone, 2005). The first type, referred to as Group 1, display symptoms of ASD during infancy or shortly afterwards and often engage in repetitive motor behaviours and display obsessions with sensory stimulation. In this sub-type, receptive language is impaired and expressive language is either absent or repetitive (similar to Echolalia in those with ASD).

The second subtype that was identified, Group 2, usually show ASD symptoms at a later age and were reported to experience a substantial regression (between 3-7 years old) or a plateau in language and social development. Repetitive behaviours were also observed in this subtype as well as anxiety. Group 2 DS-ASD is believed to be most common in this population (Heyn, 2008). Considerations should be given with regard to these sub-types in contemplating the most appropriate interventions as children with different subtypes of DS-ASD may benefit from different types of clinical and educational practices leading to more successful outcomes.

As the knowledge that children with DS also have a high occurrence of ASD is relatively recent, little empirical research is available concerning language development in this dual diagnosis specifically. Consequently, the available information concerning language development is limited with only two studies specifically researching language development in this dual diagnosis, which includes single case study designs with the aim of increasing understanding of linguistic development in this population. Kroeger and Nelson (2006) conducted a language intervention that followed applied behaviour analysis techniques in order to assist in the development of language abilities in a 9-year-old boy with DS-ASD. The participant was identified as having DS-ASD at the age of two when a regression in spoken language development was observed. He was assessed on a range of measures shortly after participating in the language intervention and again following another 9 months post-intervention.

The language training intervention was administered for one hour over 14 consecutive days and it was designed in relation to previous interventions employing reinforcement techniques. The participant was required to orally name objects and the items were withheld until the correct behaviour was made (i.e. naming the object). Verbal prompts were initially given with a range of activities with the required responses becoming longer and more complex as the intervention progressed. The intervention was reported to be successful as verbal production increased substantially following the intervention and also at follow up. This was particularly true for responsive language. The findings of this study provide promising evidence that interventions in this population can be advantageous, especially as the increase in speech was maintained at follow-up. Although this is a single case study design, this study provides a useful starting point to further language interventions for this population and future research.

A second study that investigated the language and cognitive profiles of adults with DS-ASD was conducted by Dressler et al. (2011). Individuals with DS-ASD ($n = 8$) were compared on a range of measures to adults with a singular diagnosis of either ASD ($n = 8$) or DS ($n = 8$). The DS-ASD group performed lower on all measures in comparison to the DS group which included receptive and expressive language, communication, socialisation and daily living. There were no group differences for written skills on the Vineland adaptive behaviour scales (Sparrow, Balla & Cicchetti, 2003). The DS-ASD also performed more poorly on the majority of measures compared to the ASD group, with the exception of the receptive and domestic

skills subdomains. On the whole, this study reports that adults with DS-ASD show similar profiles to that of adults with DS with relative strengths in receptive language and weaknesses in expressive language and interpersonal relationships, however, the DS-ASD group's profile was generally lower. The authors propose that the profiles of those with DS-ASD were more similar to that of the adults with DS as opposed to those with ASD, however, the comorbidity of ASD had impacted their adaptive skills to a greater degree than the DS. This study also suggests that early identification of ASD in children with DS is required as inappropriate educational strategies may otherwise be implemented which are not as well suited for those with DS-ASD

In considering bilingualism in children with a dual DS-ASD diagnosis, there is no study to date which reports on the development of children from this population who have been exposed to two languages. When it comes to bilingualism in children with a DD, as reported in Chapter 2, and more specifically subsection 2.2, there is a growing body of literature which documents the bilingual language development for children with ASD, and a smaller growing body of literature reporting on bilingual language acquisition within DS. This literature suggests that neither children with DS or ASD show any additional delays or impairments as a result of being exposed to an additional language (Kay-Raining Bird et al., 2005; Kay-Raining Bird, 2009; Hambly et al., 2013; Cleave et al., 2014; Hambly & Fombonne, 2014). This is also in line with the findings in Chapters 4 and 5. To date, there is no published literature on bilingual language development for children with a dual diagnosis of DS-ASD. As DS-ASD appears to have its own behavioural and linguistic phenotype, research relating to DS or ASD bilingual language development alone may not be appropriate when recommendations are made or when interventions are formulated for these children. The current research aimed to provide a preliminary insight into bilingual language profiles in children with DS-ASD.

7.1.3 Current Study

To date, the research concerning children with a dual diagnosis of DS-ASD predominately investigates prevalence rates, diagnostic specifications and case studies documenting language development and language interventions. No information currently exists in relation to language development in bilingual populations who have a dual DS-ASD diagnosis. Given the growing number of bilingual speakers and the linguistic context in Wales which promotes bilingualism and provides opportunities for children to become bilingual, case studies that document bilingual language profiles within this population specifically will provide a useful and novel insight into bilingual language outcomes. This research will extend the understanding of bilingualism in children with a DD further by examining the bilingual experiences of four children with DS-ASD.

More specifically, the following research questions will be addressed:

- I. What are the language abilities of bilingual children with a dual diagnosis of Down syndrome and Autism Spectrum Disorder?
- II. How does bilingual acquisition in children with a dual diagnosis of Down syndrome and Autism Spectrum Disorder compare to bilingual language acquisition in children with Down syndrome and typical bilingual acquisition?
- III. Do bilingual children with a dual diagnosis of Down syndrome and Autism Spectrum Disorder display similar profiles to bilingual and monolingual children with Down syndrome?

7.2 Method

Four children with DS were excluded from prior analyses due to the fact that they had a dual diagnosis of DS and ASD. Consequently, a multiple single case study design was used in order to document four unique cases of Welsh-English bilingual language development in children with a dual DS-ASD diagnosis. Each of these case studies is presented in a case by case approach alongside three control groups. These control groups comprise of bilinguals with DS, monolinguals with DS and TD bilingual children and are described further below.

7.2.1 Participants

Case study participants

Four children with a dual DS-ASD diagnosis were recruited from four locations in Wales. Parents confirmed that three participants had a confirmed clinical diagnosis of DS (trisomy 21) and ASD. The final participant had a diagnosis of mosaic DS and a suspected ASD diagnosis as she had previously scored within the clinical criterion for ASD on clinical assessments.¹⁴ All parents gave informed consent for their children to take part prior to data collection. A summary of the participant's demographic characteristics is presented in Table 27 below.¹⁵ Case descriptions containing further information about each case study participant is provided below.

Case Study 1: Dylan

Dylan was 10 years and 5 months old at the time of data collection and had a developmental age below 4 years. He had a confirmed diagnosis of ASD and DS although it was noted by the parent that the participant's ASD appeared to be the most overarching diagnosis. He attended a mainstream Welsh-medium school and received one-to-one support. Dylan also received Makaton input alongside Welsh and English, particularly for commands. His home language

¹⁴ Follow-up correspondence with the family confirmed that the fourth participant had since received a confirmed diagnosis of autism.

¹⁵ Pseudonyms are used throughout in order to preserve the identity and anonymity of the participants.

was Welsh and he was Welsh dominant with the majority of input currently and over his lifetime being Welsh (both at 80%). Dylan responded in the same language that he was spoken to. Exposure to English and Welsh begun from birth and he had been continuously exposed to both languages. Dylan's mum did not receive any advice regarding bilingual language exposure, however, her native language is Welsh and she subsequently did not question whether she would speak to him in Welsh or not.

	Dylan	Catrin	Owain	Rhiannon
Age	10;5	13;3	16;8	6;8
Gender	Male	Female	Male	Female
NVMA	<4;0	<4;0	<4;0	6;3
AoE	Birth	*	Birth	Birth
Schooling	Mainstream	Mainstream	Mainstream (Resource Unit)	Mainstream
Lol	Welsh	English	Welsh	Welsh
Hearing Impaired	No	No	No	No
First word	12 Months	48 Months	24 Months	15 Months

Table 27. Demographic information relating to the four participants identified as having a dual DS-ASD diagnosis. *Note:* NVMA: Non-verbal mental age, AoE: Age of Exposure, Lol: Language of Instruction. **Indicates missing information*

Case Study 2: Catrin

At data collection, Catrin was 13 years and 3 months old. She also had a developmental age of below 4 and had a confirmed diagnosis of both DS and ASD. Catrin had initially received input in Welsh and English before the age of 5 through her family and she had been attending

a Welsh-medium mainstream school. At the age of 5, she was fostered and her foster parents were advised to discontinue exposure to Welsh as this would be detrimental to her language development, given that she still had very limited language abilities. Her parents were also concerned as Catrin used to mix Welsh and English words when she was younger. Catrin's parents decided for her to attend an English-medium school, and her statement disappplied her from participating in Welsh language lessons, although some limited Welsh phrases were still used at the school. Makaton is used alongside English to communicate with Catrin. Her parents estimated that her lifetime exposure to Welsh was around 20%, however, currently, her language input is in English 99% of the time and she only responds in English.

Case Study 3: Owain

Owain was the oldest participant at the time of data collection at 16 years and 8 months old. Assessments showed that his developmental age was below 4 years old. Owain had a confirmed diagnosis of DS, ASD and also ADHD. He was attending a mainstream Welsh-medium school which had a resource unit in which he attended for the whole of the school day. Owain was Welsh dominant and received Welsh input as the home and school language. He had previously used Makaton but was not reported to use it anymore. Lifetime exposure to Welsh was reported to be 80%, although, he had attended an English-medium nursery prior to beginning Welsh-medium schooling. Owain was exposed to Welsh from birth and had been continuously exposed to both languages. In terms of responding, parental reports estimated that he would choose to respond in Welsh 90% of the time and 10% of responses would be in English.

Case Study 4: Rhiannon

The youngest participant in this case study analysis was Rhiannon, who was aged 6 years and 8 months at data collection. Rhiannon was the only participant with a diagnosis of mosaic DS and also had the highest developmental age at 6 years and 3 months. In terms of developmental age, she scored closest to her CA with only 6 months difference between these. Rhiannon was the only participant with suspected ASD which had not been formally confirmed. It was reported that Rhiannon had previously scored within the lower percentile on certain assessments and it was likely that she would receive a diagnosis of ASD at a later stage.¹⁶ She was not reported to have any hearing impairments and the age of her first word was at 15 months old. Rhiannon was attending a small mainstream Welsh-medium school, in a class with only 3 other pupils. She had received exposure to Welsh from birth, although the primary home language was English. She received input to Welsh and English on a daily basis with Welsh exposure at school and small amounts of Welsh were also used at home. Parental

¹⁶ Follow-up correspondence with the family confirmed that the fourth participant had since received a confirmed diagnosis of autism.

reports estimated that she had a current and lifetime exposure to Welsh of 20%, and would respond according to the language being spoken.

Control Groups

Three control groups were recruited (see Chapter 3 regarding recruitment procedure), the first being Welsh-English bilingual children with a diagnosis of DS only (DSB; $n = 10$), secondly Welsh-English TD children (TDB; $n = 25$) and also English monolingual children with DS (DSM; $n = 15$). This enabled a comparison between each participants language profile with these control groups. The control groups included the same samples as reported on previously throughout this thesis. In summary, the children in the DSB were aged between 5;5 and 13;9 at the time of data collection ($M = 114.2$ months or 9;2, $SD = 37.7$), the DSM group were between 6;0-16;9 ($M = 114.9$ months or 9;6, $SD = 35.6$) and the TDB group were aged between 3;0 and 7;10 ($M = 51$ months or 4;3, $SD = 16.1$). For the DSB group, 6 were female and 4 were male and they had an average current exposure to Welsh of 49.1%. All except one were attending Welsh-medium schools, 7 of which were mainstream schools. Two had a mix of schooling between a SEN school and a mainstream school. The final participant in the DSB control group had recently transferred to an English-medium SEN school from a mainstream Welsh-medium school. All parents confirmed that they had no known or suspected additional diagnosis of ASD. The DSM group comprised of 5 males and 10 females and had very limited exposure to Welsh with a mean current exposure to Welsh of 3.5%. Two were attending SEN schools and the remaining were attending mainstream schools. Parents of these children confirmed no additional diagnosis that impacted language development such as ASD. The final control group of TDB children included 12 males and 13 females and all were attending mainstream schools or nurseries. The children had an average current exposure to Welsh of 51.5%¹⁷ and no parent reported that their child had any known or suspected disability or language impairment.

7.2.2 Procedure

Children were assessed on a range of cognitive and linguistic assessments in a one-to-one setting. The three children who still received Welsh-medium input were assessed in English and Welsh in a separate session. The final participant, Catrin was assessed in English only. Participants in the three control groups completed identical assessments using the same protocols. Assessments were conducted in a pre-defined order as described in Chapter 4. In summary, assessments comprised of; non-verbal cognitive abilities, core, expressive and

¹⁷ Missing data for 5 children due to non-return of the background questionnaire.

receptive language in English, receptive language in Welsh, phonological awareness in English and Welsh, and finally, working memory in English and Welsh.¹⁸

7.2.3 Data Analysis

Descriptive information is initially presented for each participant in the case study as well as the demographic information of the three control groups. Following this, each case study participant is described in detail as well as an overall description of their language scores and language profiles. Subsequent to this, the Revised Standardized Difference Test (RSDT; Crawford & Garthwaite, 2005; Crawford & Garthwaite, 2007), which is a modified t-test developed for use with analysing single-case studies in comparison to a control group was utilized for each case study participant in comparison to each control group in order to ascertain if there are any significant differences in their language and cognitive abilities. This test was recently developed to enable the analysis of neuropsychological case study research (singlebayes.exe program was used for the current analyses) which also provides point and interval estimates of effect sizes, controls for Type 1 errors regardless of small sample sizes and is also robust with skewed data (Crawford & Howell, 1998; Crawford et al., 2003).

7.3 Results

From the overall sample of children with DS recruited for the research ($n = 32$), 4 were identified as having a diagnosis or suspected diagnosis of ASD. This corresponds to a prevalence of 12.5% in the current sample of children with DS. A summary of each participant's performance on the assessments in comparison to each control group is presented in Table 28. On average, parents of children with DS-ASD reported their children's receptive language abilities as 2.5 (maximum score of 5 whereby 1 = poor, 5 = excellent) and expressive language abilities as 2 (as before with a maximum score of 5). Parents of the DSB children reported their average receptive language as 3.5 and expressive language as 2.6. Parents of the monolingual children with DS reported an average receptive language score of 3.67 and an expressive language score of 3.07. The parents of the TDB children estimated their children's receptive language as a score of 4.45 and their expressive language at 4.18.

The four children with DS-ASD's mean scores for the English language assessments were 26.5 for Core Language, 23.5 for Receptive Language, 22 for Expressive Language, 26.25 for Language Content and 18.5 for Language Structure. This is in comparison to the DSB control group who scored 31.1 for Core Language, 28.3 for Receptive Language, 25.8 for Expressive Language, 35.5 for Language Content and 21.8 for Language Structure. The DSM control group scored 39.07 for Core Language, 32.27 for Receptive Language, 36.93 for Expressive Language, 43.93 for Language Content and 26.93 for Language Structure. The TDB

¹⁸ For detailed information relating to the assessments and testing sessions, see the methods chapter (Chapter 4).

group scored 44.23 for Core Language, 39.5 for Receptive Language, 49.2 for Expressive Language, 47.56 for Language Content and 44.28 for Language Structure. For Welsh Receptive Vocabulary the DS-ASD group scored an average of 21.66, the DSB group scored an average of 22.8 and the TDB an average of 22.5. Phonological awareness scores were 23 in English and 21.8 in Welsh for the DS-ASD participants. In comparison, the DSB group's mean score for these was 26.6 in English and 28.6 in Welsh. The TDB group averaged 26.5 in English and 27.5 in Welsh and the DSM group averaged 25.2 in English. In order to evaluate the unique profile of each participant with DS-ASD, each case study is presented individually below.

	KBIT	Working Memory	Parent Report - Receptive	Parent Report - Expressive	Core Language (CELF)	Receptive Language (CELF)	Expressive Language (CELF)	Language Content (CELF)	Language Structure (CELF)	Welsh Receptive Language	English PA	Welsh PA
Dylan	4	3.5	2	2	10	7	12	13	6	19	22	27
Catrin	5	2	4	3	29	32	20	31	21	-	25	-
Owain	3	3	1	1	17	16	11	13	11	18	16	26
Rhiannon	20	3	3	2	50	39	45	48	36	18	29	33
DSB Group	9.6	2.8	3.5	2.55	31.1	28.3	25.8	35.5	21.8	22.8	21.8	26.6
TDB Group	11.8	3.4	4.45	4.18	44.23	39.5	49.2	47.56	44.28	22.5	26.5	27.5
DSM Group	10.9	2.93	3.67	3.07	39.07	32.27	36.93	43.93	26.93	-	25.2	-

Table 28. Summary of individual case study participant's scores on language measures, working memory, non-verbal intelligence and parent reported language abilities.

7.3.1 Case Study Analysis

Dylan

Dylan was classified as having severe learning difficulties as measured on the NV subtest of the KBIT and his IQ equivalent was 40. On the digit span task, he was able to recall 3 items in Welsh and 4 items in English. Dylan's parents reported that he spoke his first word at around 12 months of age. Dylan's expressive language was marginally better than his receptive language in English, and his language content was higher than his language structure scores. For the English language assessments as measured on the CELF, he scored 10/86 for core language, 7/62 for receptive language, 12/101 for expressive language, 13/80 for language content and 6/83 for language structure. Dylan scored 19/37 on the Welsh Receptive Language assessment and scored an average of 24.5/60 for the PA assessments. Performance was highest on the phoneme component and lowest for the rhyme component on the PA measures. In terms of age equivalent scores, Dylan's performance on the CELF corresponded to an age-equivalent score of below 3 for every sub-test. A further breakdown of his results is presented below in Table 29.¹⁹

Measure	Score	% Correct
Sentence Structure	1	4.5
Word Structure	2	8.3
Expressive Vocabulary	7	17.5
Concepts & Following Directions	3	13.6
Recalling Sentences	3	8.1
Basic Concepts	3	16.6
Word Classes	*	*
Welsh Receptive Vocabulary	19	51.4
Syllable	10	50.0
Rhyme	3	15.0
Phoneme	11.5	57.5

Table 29. Summary of Case Study 1 results.

¹⁹ *Represents missing data.

Catrin

Catrin was also classified as having severe learning difficulties as measured on the NV subtest of the KBIT with an IQ equivalent to 40. For the digit span task, she could correctly recall 2 items in the correct order. Catrin was reported to have spoken her first word at 48 months of age. For the English language assessments, her receptive language was considerably higher than her expressive language, and her language content was higher than her language structure. On the CELF, she scored 29/86 for Core Language, 32/62 for Receptive Language, 20/101 for Expressive Language, 31/80 for Language Content and 21/83 for Language Structure. Catrin had an age equivalent score of below 3 for word structure, concepts and following directions and recalling sentences sub-tests. For sentence structure and expressive vocabulary, her score was an age-equivalent of 3;3, for basic concepts, her score equaled to 4;0. For the English PA measures, Catrin performed equally as well on the Rhyme and Phoneme tasks but scored lower on the Syllable assessments. Overall, she scored 25/60. As Catrin has not received any exposure to Welsh for 8 years and her statement disappplied her from Welsh lessons, the Welsh assessments were not administered. A summary of Catrin's scores is presented in Table 30.²⁰

Measure	Score	% Correct
Sentence Structure	11	50.0
Word Structure	8	33.3
Expressive Vocabulary	10	25.0
Concepts & Following Directions	6	27.3
Recalling Sentences	2	5.4
Basic Concepts	15	83.3
Word Classes	2**	5.0**
Welsh Receptive Vocabulary	*	*
Syllable	5	25.0
Rhyme	10	50.0
Phoneme	10	50.0

Table 30. Summary of Case Study 2 results.

²⁰ * Represents missing data.

** Represents incomplete assessment score as the participant became uncooperative towards the end of testing.

Owain

Owain's scores on the NV cognitive assessment indicated that he had been classified as having a severe learning difficulty indicated by an IQ score of 40. On the digit span task, he was able to recall 3 items in Welsh and 3 items in English. Owain was reported to have spoken his first word at around 24 months. Owain's receptive language was higher than expressive language score on the CELF assessment and his language content was slightly higher than his language structure. He scored 17/86 for core language, 16/62 for receptive language, 11/101 for expressive language, 13/80 for language content and 11/83 for language structure. On the Welsh receptive vocabulary test, he scored 18/37. Owain's scores corresponded to an age-equivalent of below 3 for all sub-tests of the CELF. In terms of PA, Owain scored highest on the phoneme tasks and lowest on the rhyme component. Overall he scored 21/60 on average between the Welsh and English PA assessments. A further breakdown of his results is presented below in Table 31.

Measure	Score	% Correct
Sentence Structure	8	36.4
Word Structure	1	4.8
Expressive Vocabulary	8	20.0
Concepts & Following Directions	4	18.2
Recalling Sentences	2	5.4
Basic Concepts	4	22.2
Word Classes	2	5.0
Welsh Receptive Vocabulary	18	48.6
Syllable	7	35.0
Rhyme	3.5	17.5
Phoneme	10.5	52.5

Table 31. Summary of Case Study 3 results.

Rhiannon

Rhiannon's performance on the NV matrices subtest classified her as being within the normal range of cognitive ability with an IQ of 98. She had a digit span of 3 as measured by the forward digit span task in Welsh and English. Rhiannon was reported to have spoken her first word at around 15 months of age. Rhiannon was stronger in her receptive language compared to expressive language and stronger in language content than language structure. On the CELF, Rhiannon scored 50/86 for core language, 39/62 for receptive language, 45/101 for

expressive language, 48/80 for language content and 36/83 for language structure. For the Welsh receptive language test, Rhiannon scored 18/37. Her age-equivalent scores on the CELF were: sentence structure, 3;11; word structure, 4;5; expressive vocabulary, 4;8; concepts and following directions, 4;2; recalling sentences, 3;6; basic concepts 3;7 and word classes, 4;9. Performance on the PA tasks showed that she was considerably stronger on the phoneme tasks and scored equally lower on the syllable and rhyme components with an average overall score of 31/60. Table 32 presents a further breakdown of Rhiannon's performance on these measures.

Measure	Score	% Correct
Sentence Structure	13	59.1
Word Structure	15	62.5
Expressive Vocabulary	22	55.0
Concepts & Following Directions	12	54.5
Recalling Sentences	8	21.6
Basic Concepts	14	77.8
Word Classes	26	65
Welsh Receptive Vocabulary	18	48.6
Syllable	6	30.0
Rhyme	6	30.0
Phoneme	17	85

Table 32. Summary of Case Study 4 results.

Control Group - DSB

The DSB control group had an average raw score on the KBIT of 9.6 (average IQ equivalent of 55.2), with the majority displaying moderate learning difficulties (8/10) and the remaining having severe learning difficulties (2/10). For the digit span assessment, the group could recall 2.8 digits in the correct sequence on average. The average age of first word was 24.9 months. Higher levels of receptive language were found compared to expressive language with an average of 28.3/62 (45.6%) for receptive and 25.8/101 (25.5%) for expressive. Language Content was higher in the DSB control group than Language Structure with an average of 35.5/80 (44.4%) for Language Content and 21.8/83 (26.3%) for Language Structure. Performance on the subtests showed that, on average, they scored below an age-equivalent of 3 for word structure, concepts and following directions and recalling sentences. For sentence structure, the group had an average age-equivalent of 3;3, expressive vocabulary of

3;7, basic concepts of 3;2 and word classes of below 4 years old. The average for the Welsh receptive vocabulary assessment was 22.8/37. For PA, the control group scored an average of 24.1/60 and had the highest performance on the phoneme tasks. The group had the most difficulty with the rhyme assessments. Table 33 summarises these results.

Measure	Score	% Correct
Sentence Structure	10.2 (4.8)	46.4
Word Structure	6.7 (4.8)	27.9
Expressive Vocabulary	14.2 (9.1)	35.5
Concepts & Following Directions	6 (3.9)	27.3
Recalling Sentences	5 (5.8)	13.5
Basic Concepts	12 (4.5)	66.6
Word Classes	12 (10.8)	30.0
Welsh Receptive Vocabulary	22.8 (6.7)	61.6
Syllable	7 (2.4)	35.0
Rhyme	5 (3.2)	25.0
Phoneme	10 (5.8)	50.0

Table 33. Summary of the results for the DSB control group. *Note:* Mean scores are reported with standard deviations in parenthesis.

Control Group - TDB

The TDB control group had an average raw score on the KBIT of 11.84 (with an average IQ equivalent of 94.96 according to the KBIT manual). The group's digit span was an average of 3.4, and the average age of first word was 12.3 months. The group displayed higher receptive language compared to expressive language with 39.5/62 (63.7%) and 49.2/101 (48.7%) respectively. Higher abilities were found for Language Content than Language Structure with an average of 47.56/80 (59.45%) for Language Content and 44.28/83 (53.35%) for Language Structure. Performance on the subtest showed that the mean scores were above an age equivalent of 3;0 and were in line with age expectancy for the population (average age 4;3). The average for the Welsh receptive vocabulary assessment was 22.5/37. For PA, the group had an average score of 26.5/60 overall in English and 26.6/60 in Welsh and had the highest performance on the phoneme tasks. The group had the most difficulty with the rhyme assessments. A summary of the results is presented in Table 34.

Measure	Score	% Correct
Sentence Structure	13.2 (6.0)	60.0
Word Structure	12.3 (5.9)	44.1
Expressive Vocabulary	18.7 (9.5)	46.8
Concepts & Following Directions	11.2 (5.6)	50.9
Recalling Sentences	17.1 (10.3)	46.2
Basic Concepts	15.2 (3.9)	84.4
Word Classes	22.1 (11.2)	55.3
Welsh Receptive Vocabulary	22.5 (8.3)	60.8
Syllable	9.7 (4.3)	48.5
Rhyme	6.2 (5.1)	31.0
Phoneme	10.6 (6.5)	53.0

Table 34. Summary of the results for the TDB control group. *Note:* Mean scores are reported with standard deviations in parenthesis.

Control Group - DSM

The average raw score on the KBIT for the DSM control group was 10.9 (average IQ equivalent of 58.2). The majority of children in this group were considered to have moderate learning difficulties (11/15) and the remainder had severe learning difficulties (4/11). They had a mean digit span of 2.9 and the average age of first word was 21.7 months. They also displayed higher receptive language abilities than expressive language on the CELF with 32.3/62 (52.1%) and 36.9/101 (36.5%) respectively. Language Content was higher also in the DSM control group than Language Structure with an average of 43.9/80 (54.9%) for Language Content and 26.9/83 (32.4%) for Language Structure. Performance was above an age equivalent of 3 for all subtests with age equivalents as follows: sentence structure, 3;1, word structure, 3;2, expressive vocabulary 4;1, concepts and following directions, 3;3, recalling sentences, 3;5, basic concepts, 3;7 and word classes below an age equivalent of 4. For the PA assessments, the control group had an average score of 25.1/60 overall and also had the highest performance on the phoneme tasks. The group had the most difficulty with the rhyme assessments. A summary of the results is presented in Table 35. A summary of results for core, receptive and expressive language in English in comparison to the three control groups is presented in Figure 16 and the results for the Welsh receptive vocabulary task are presented in Figure 17. A summary of the results for the PA assessments is presented in Figure 18.

Measure	Score	% Correct
Sentence Structure	9.9 (6.3)	45.0
Word Structure	9.2 (5.7)	38.3
Expressive Vocabulary	19.9 (8.7)	49.8
Concepts & Following Directions	8.3 (4.3)	37.7
Recalling Sentences	7.8 (8.5)	21.1
Basic Concepts	14 (4.3)	77.8
Word Classes	16.9 (11.2)	42.3
Welsh Receptive Vocabulary	-	-
Syllable	8.1 (5.0)	40.5
Rhyme	7.2 (4.7)	36.0
Phoneme	9.9 (6.2)	49.5

Table 35. Summary of the results for the DSM control group. *Note:* Mean scores are reported with standard deviations in parenthesis

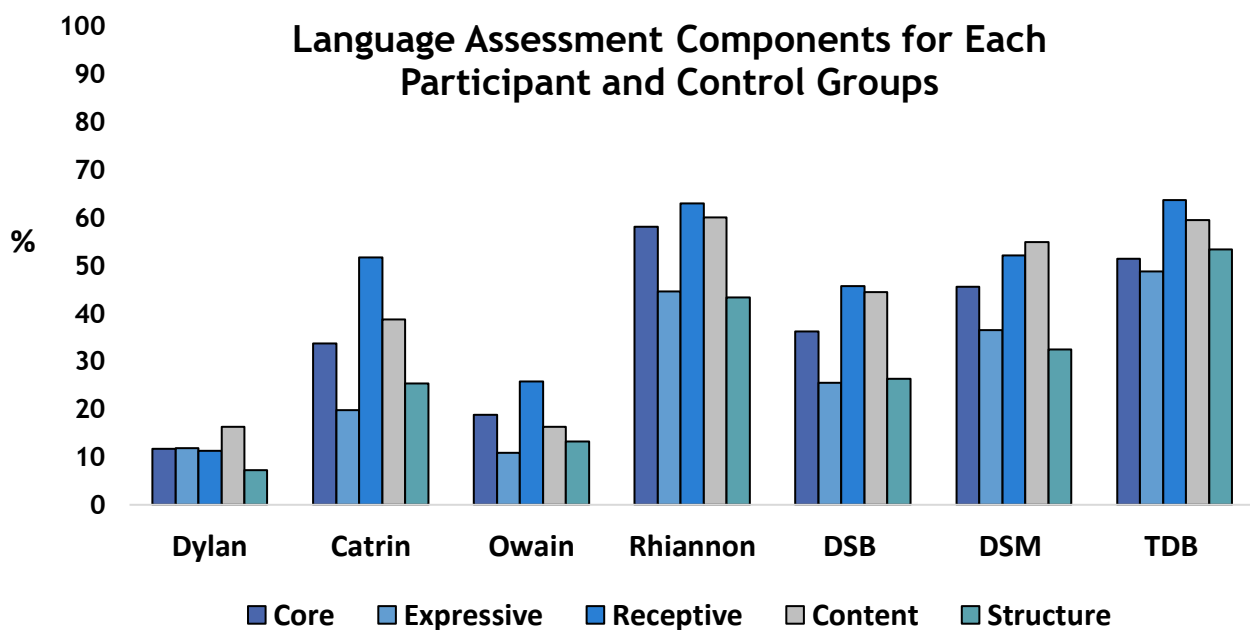


Figure 16. A summary of the case study results for English language assessments on the CELF in comparison to the control groups (reported in %).

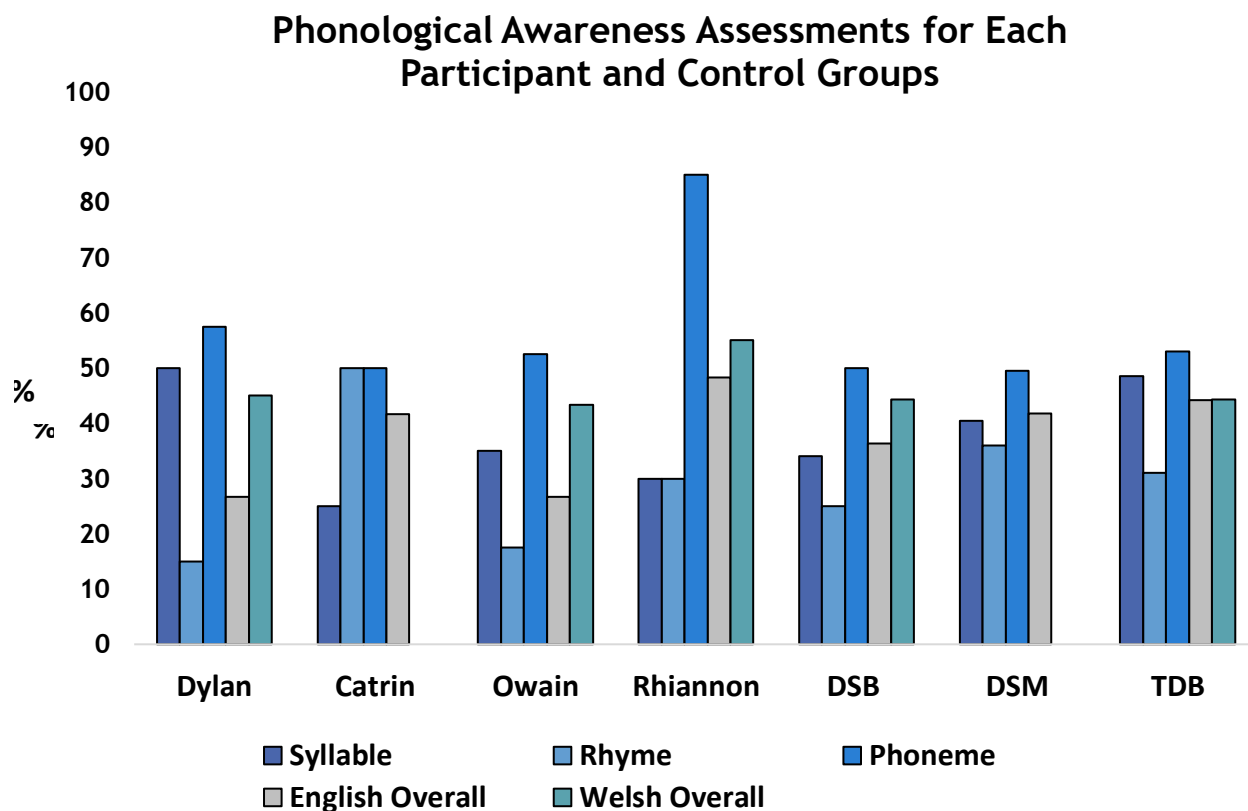


Figure 17. A summary of the case study results for Syllable, Rhyme, Phoneme and Overall Phonological awareness in comparison to the control groups.

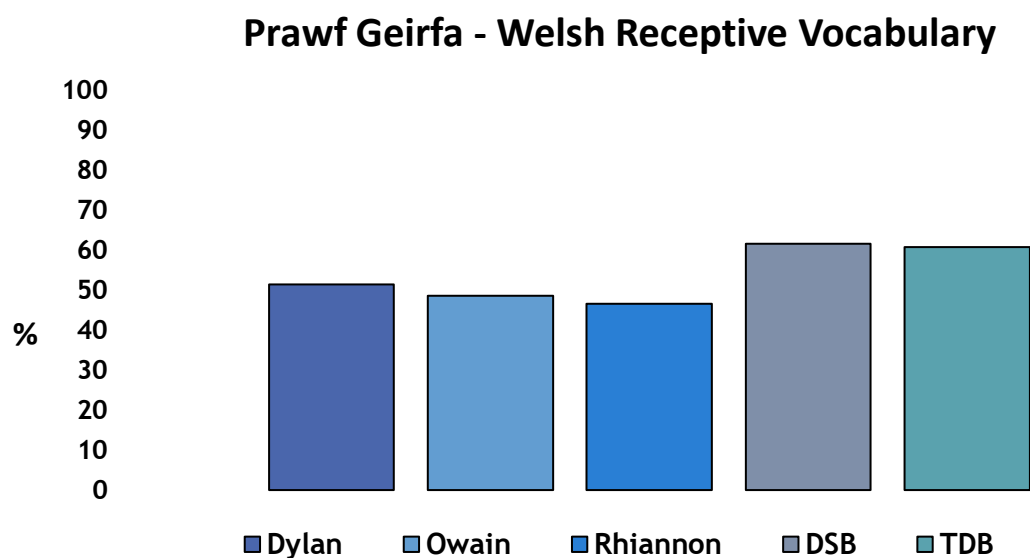


Figure 18. A summary of the case study results for Welsh receptive Language as measured by the Prawf Geirfa in comparison to the control groups.

7.3.2 Statistical Analyses - Revised Standardised Difference Tests

Dylan

Dylan's age of first word was reported to be around 12 months of age. This was significantly earlier than the average for the DSB group but not significantly different from the other two control groups (DSB, $p < .001$, DSM, $p = .415$ and TDB, $p = .930$). As is often typical in children with a dual DS-ASD diagnosis, Dylan performed significantly lower than the TDB group for all the English language measures. In comparing his performance to the two groups of children with DS he performed significantly lower than both the bilingual and monolingual groups of children with DS on measures of English receptive language. For all other measures, there were no significant differences in performance, however, he did score below that of all control groups for measures of non-verbal cognitive ability, all English language assessments, and Welsh receptive vocabulary. Similar performance was found for all measures of PA in comparison to the three control groups with no significant differences emerging. See Table 36 for a summary of Dylan's performance in comparison to the control groups which displays significant effects and point estimates of performance and 95% confidence intervals.

Catrin

Age of first word for Catrin was reported to be the latest out of all case study participants at 48 months of age. This was significantly later than all three control groups (DSB, $p < .001$, DSM, $p < .05$ and TDB, $p < .001$). Catrin's score on the non-verbal mental age assessment was lower than all three control groups but there was no significant difference found. A similar finding was evident for her performance on the English language assessments where her performance was lower on all components of the CELF, however, she did not perform significantly lower than any group. In terms of PA, she scored slightly lower on the syllable component which did not reach significance between any group. For all other elements of PA, her performance was comparable to that of the other groups with no significant differences. Table 37 summarises Catrin's performance in comparison to the control groups which displays point estimates of performance and 95% confidence intervals.

Owain

Owain's age of first word was reported to be around 24 months of age. This was significantly later than the average for the TDB group but not significantly different from the other two control groups of children with DS (DSB, $p = .727$, DSM, $p = .845$ and TDB, $p < .01$). Owain displayed the largest cognitive impairment and his performance on the non-verbal cognitive assessment was significantly lower than the TDB control group. His language abilities were lower than all control groups for English and Welsh language assessment and this was significantly so for English language content in comparison to the TDB group ($p < .05$). Rhyme performance on the PA assessment was lower than the control groups but not significantly so. All other PA measures were similar to that of the control groups. See table 38 for a

summary of Owain's performance in comparison to the control groups which displays significant effects and point estimates of performance and 95% confidence intervals.

Rhiannon

The age of first word for Rhiannon was reported to be around 15 months of age. This was significantly earlier than the DSB control group but not significantly different from the other control groups (DSB, $p < .01$, DSM, $p = .571$ and TDB, $p = .403$). In contrast to the other case study participants, Rhiannon did not perform below the three control groups for NV cognitive ability or any of the English language measures. In comparison to the two DS groups, Rhiannon's scores were higher for all English language assessments and were often higher than the TDB control group also. Although her performance was higher than the control groups, this only approached significance and did not reach significance for any measure. Rhiannon's Welsh receptive language score was slightly lower than the control groups but again this was not significant. Her performance on the PA tasks was similar to that of the control groups for syllable and rhyme but she scored higher than all control groups again for the phoneme measures but not significantly. Rhiannon's performance in comparison to the control groups is presented in Table 39 with point estimates of performance and 95% confidence intervals.

Measure (Max Score)	Dylan	DSB (n=10)	Point Estimate (95% CI)	DSM (n=15)	Point Estimate (95% CI)	TDB (n=25)	Point Estimate (95% CI)
Non-Verbal Ability							
<i>KBIT (46)</i>	4	9.6 (6.6)	22.1 (6.0-45.9)	10.9 (5.2)	11.0 (2.2-27.0)	11.8 (4.7)	5.8 (1.2-14.8)
English Assessments							
<i>Core (86)</i>	10	31.1 (17.1)	13.5 (2.0-35.4)	39.1 (17.8)	6.8 (0.8-21.1)	44.2 (17.1) *	3.1 (0.4-9.5)
<i>Receptive (62)</i>	7	28.3 (10.6) *	4.44 (0.1-18.9)	32.27 (13.5) *	4.55 (0.3-15.6)	39.5 (13.9) *	1.67 (0.1-6.5)
<i>Expressive (101)</i>	12	25.8 (18.9)	25.2 (7.8-49.6)	36.9 (20.0)	12.4 (2.8-29.2)	49.2 (22.2)	5.7 (1.1-14.6)
<i>Content (80)</i>	13	35.5 (18.7)	14.0 (2.2-36.1)	43.9 (19.5)	7.4 (0.9-21.1)	47.6 (16.8) *	2.7 (0.3-8.8)
<i>Structure (83)</i>	6	21.8 (14.0)	15.5 (2.8-38.0)	26.9 (18.3)	14.4 (3.7-31.8)	44.3 (19.3) *	3.2 (0.4-9.7)
Welsh Assessment							
<i>Receptive (37)</i>	19	22.8 (6.7)	30.0 (11.0-55.0)	-	-	22.5 (8.3)	32.9 (19.3-48.3)
Phonological Awareness							
<i>Syllable (20)</i>	10	7 (2.4)	86.8 (65.2-98.1)	8.1 (5.0)	64.2 (44.2-81.5)	9.7 (4.3)	52.7 (37.3-67.8)
<i>Rhyme (20)</i>	3	5 (3.2)	28.4 (9.8-53.0)	7.2 (4.7)	20.0 (6.9-39.1)	6.2 (5.1)	27.2 (14.6-42.3)
<i>Phoneme (20)</i>	11.5	10 (5.8)	59.5 (35.2-81.2)	9.9 (6.2)	59.7 (39.7-77.9)	10.6 (6.5)	56.8 (41.3-71.6)
<i>Overall (60)</i>	24.5	24.1 (7.5)	52.0 (28.5-75.0)	25.1 (14.4)	48.3 (29.1-67.8)	26.6 (13.9)	44.1 (29.3-59.6)

Point Estimate indicated the percentage of the population in the control group estimated to perform below Dylan's score. *Bold indicates a significant effect with $p < 0.05$.

Table 36. Dylan's performance on all measures in comparison to the control groups which shows any significant effects with point estimates and 95% confidence intervals.

Measure (Max Score)	Catrin	DSB (n=10)	Point Estimate (95% CI)	DSM (n=15)	Point Estimate (95% CI)	TDB (n=25)	Point Estimate (95% CI)
Non-Verbal Ability							
<i>KBIT (46)</i>	5	9.6 (6.6)	26.2 (8.4-50.6)	10.9 (5.2)	14.5 (3.8-32.0)	11.8 (4.7)	8.4 (2.3-19.1)
English Assessments							
<i>Core (86)</i>	29	31.1 (17.1)	45.5 (22.9-69.3)	39.1 (17.8)	29.6 (13.4-49.6)	44.2 (17.1)	19.6 (8.8-33.9)
<i>Receptive (62)</i>	32	28.3 (10.6)	62.7 (38.3-83.8)	32.27 (13.5)	49.2 (29.8-68.8)	39.5 (13.9)	30.1 (17.0-45.4)
<i>Expressive (101)</i>	20	25.8 (18.9)	38.8 (17.6-63.0)	36.9 (20.0)	21.3 (7.7-40.5)	49.2 (22.2)	10.4 (3.2-22.0)
<i>Content (80)</i>	31	35.5 (18.7)	41.2 (19.2-65.4)	43.9 (19.5)	26.6 (11.3-46.5)	47.6 (16.8)	17.1 (7.2-30.7)
<i>Structure (83)</i>	21	21.8 (14.0)	47.8 (24.9-71.5)	26.9 (18.3)	38.0 (20.1-58.1)	44.3 (19.3)	12.4 (4.3-24.8)
Phonological Awareness							
<i>Syllable (20)</i>	5	7 (2.4)	22.4 (6.2-46.5)	8.1 (5.0)	27.9 (12.2-47.9)	9.7 (4.3)	14.7 (5.7-28.0)
<i>Rhyme (20)</i>	10	5 (3.2)	91.5 (72.6-99.4)	7.2 (4.7)	71.4 (51.4-87.3)	6.2 (5.1)	76.4 (61.5-88.2)
<i>Phoneme (20)</i>	10	10 (5.8)	50.1 (26.9-73.3)	9.9 (6.2)	50.6 (31.4-70.0)	10.6 (6.5)	45.5 (30.5-61.0)
<i>Overall (60)</i>	25	24.1 (7.5)	54.4 (30.5-77.0)	25.1 (14.4)	49.7 (30.4-69.1)	26.6 (13.9)	45.5 (30.5-61.0)

Point Estimate indicated the percentage of the population in the control group estimated to perform below Catrin's score. *Bold indicates a significant effect with $p < 0.05$.

Table 37. Catrin's performance on all measures in comparison to the control groups which shows any significant effects with point estimates and 95% confidence intervals.

Measure (Max Score)	Owain	DSB (n=10)	Point Estimate (95% CI)	DSM (n=15)	Point Estimate (95% CI)	TDB (n=25)	Point Estimate (95% CI)
Non-Verbal Ability							
<i>KBIT (46)</i>	3	9.6 (6.6)	18.3 (4.0-41.5)	10.9 (5.2)	8.2 (1.2-22.5)	11.8 (4.7) *	4.0 (0.6-11.4)
English Assessments							
<i>Core (86)</i>	17	31.1 (17.1)	22.6 (6.3-46.6)	39.1 (17.8)	12.5 (2.8-29.3)	44.2 (17.1)	6.6 (1.5-16.1)
<i>Receptive (62)</i>	16	28.3 (10.6)	14.8 (2.5-37.2)	32.27 (13.5)	13.1 (3.1-30.1)	39.5 (13.9)	5.5 (1.1-14.4)
<i>Expressive (101)</i>	11	25.8 (18.9)	23.7 (7.0-47.8)	36.9 (20.0)	11.5 (2.4-27.8)	49.2 (22.2)	5.2 (1.0-13.8)
<i>Content (80)</i>	13	35.5 (18.7)	14.0 (2.2-36.1)	43.9 (19.5)	7.4 (0.9-21.1)	47.6 (16.8) *	2.7 (0.3-8.8)
<i>Structure (83)</i>	11	21.8 (14.0)	24.0 (7.1-48.5)	26.9 (18.3)	20.7 (7.3-40.0)	44.3 (19.3)	5.2 (1.0-13.7)
Welsh Assessment							
<i>Receptive (37)</i>	18	22.8 (6.7)	25.6 (8.1-50.0)	-	-	22.5 (8.3)	28.8 (15.9-44.1)
Phonological Awareness							
<i>Syllable (20)</i>	7	7 (2.4)	50.0 (27.0-73.3)	8.1 (5.0)	41.7 (23.3-61.6)	9.7 (4.3)	27.2 (14.6-42.3)
<i>Rhyme (20)</i>	3.5	5 (3.2)	33.2 (13.3-57.9)	7.2 (4.7)	23.0 (8.8-42.4)	6.2 (5.1)	30.4 (17.4-45.9)
<i>Phoneme (20)</i>	10.5	10 (5.8)	53.2 (29.6-76.0)	9.9 (6.2)	53.7 (34.1-72.6)	10.6 (6.5)	49.2 (34.1-64.6)
<i>Overall (60)</i>	16	24.1 (7.5)	16.5 (3.1-39.2)	25.1 (14.4)	27.5 (11.9-47.2)	26.6 (13.9)	23.1 (11.4-37.8)

Point Estimate indicated the percentage of the population in the control group estimated to perform below Owain's score. *Bold indicates a significant effect with $p < 0.05$.

Table 38. Owain's performance on all measures in comparison to the control groups which shows any significant effects with point estimates and 95% confidence intervals.

Measure (Max Score)	Rhiannon	DSB (n=10)	Point Estimate (95% CI)	DSM (n=15)	Point Estimate (95% CI)	TDB (n=25)	Point Estimate (95% CI)
Non-Verbal Ability							
<i>KBIT (46)</i>	20	9.6 (6.6)	91.7 (72.9-99.4)	10.9 (5.2)	94.4 (82.3-99.5)	11.8 (4.7)	94.9 (86.5-99.1)
English Assessments							
<i>Core (86)</i>	50	31.1 (17.1)	84.0 (61.3-97.0)	39.1 (17.8)	71.9 (52.0-87.6)	44.2 (17.1)	62.9 (47.4-77.0)
<i>Receptive (62)</i>	39	28.3 (10.6)	82.0 (58.7-96.1)	32.27 (13.5)	68.2 (48.1-84.9)	39.5 (13.9)	48.6 (33.4-63.9)
<i>Expressive (101)</i>	45	25.8 (18.9)	82.1 (59.0-96.2)	36.9 (20.0)	65.0 (44.9-82.3)	49.2 (22.2)	42.7 (28.0-58.2)
<i>Content (80)</i>	48	35.5 (18.7)	73.0 (48.5-91.0)	43.9 (19.5)	57.9 (38.1-76.3)	47.6 (16.8)	50.9 (35.7-66.1)
<i>Structure (83)</i>	36	21.8 (14.0)	82.0 (58.7-96.2)	26.9 (18.3)	68.2 (48.2-84.7)	44.3 (19.3)	33.9 (20.1-49.4)
Welsh Assessment							
<i>Receptive (37)</i>	18	22.8 (6.7)	25.6 (8.1-50.0)	-	-	22.5 (8.3)	28.8 (15.9-44.1)
Phonological Awareness							
<i>Syllable (20)</i>	6	7 (2.4)	35.0 (14.7-59.7)	8.1 (5.0)	34.5 (17.3-54.6)	9.7 (4.3)	20.4 (9.4-34.7)
<i>Rhyme (20)</i>	6	5 (3.2)	61.4 (37.0-82.7)	7.2 (4.7)	40.4 (22.2-60.4)	6.2 (5.1)	48.5 (33.4-63.7)
<i>Phoneme (20)</i>	17	10 (5.8)	86.0 (64.2-97.8)	9.9 (6.2)	85.7 (68.4-96.3)	10.6 (6.5)	88.4 (76.3-96.2)
<i>Overall (60)</i>	29	24.1 (7.5)	72.6 (48.0-90.8)	25.1 (14.4)	60.1 (40.2-78.3)	26.6 (13.9)	56.7 (41.1-71.5)

Point Estimate indicated the percentage of the population in the control group estimated to perform below Rhiannon's score. *Bold indicates a significant effect with $p < 0.05$.

Table 39. Rhiannon's performance on all measures in comparison to the control groups which shows any significant effects with point estimates and 95% confidence intervals.

7.4 Discussion

This research aimed to investigate the language profiles of children with a dual DS-ASD diagnosis who had received exposure to both Welsh and English. Given that children with ASD and DS display language impairments and both have a unique language phenotype, the purpose of these case studies was to highlight how these profiles interact in children who have received varying language input to more than one language. Four children were identified as having a confirmed or suspected (for one participant) diagnosis of DS and ASD. Cognitive and linguistic assessments were administered in English and also in Welsh for those that were still exposed to Welsh (three out of the four). Performance of each participant was compared to three control groups, Welsh-English bilingual children with DS, Welsh-English TD children and English monolingual children with DS.

7.4.1 Summary of Main Findings

In terms of the prevalence rate in the current sample of children with DS recruited for this research, 12.5% had a confirmed or expected dual DS-ASD diagnosis. Previous research estimates that prevalence of ASD in children with DS is around 16-18% when measured on a standardised checklist for autism or through using a social communication questionnaire (DiGuseppi et al., 2010; Richards et al., 2015). The prevalence rate in the current sample, although slightly lower than DiGuseppi and colleagues estimation, is in line with reports that ASD prevalence is considerably higher than the general population (with recent estimates between 1.1%-2.24% in the general population; Brugha et al., 2012; Zablotsky, et al., 2015).

Cognitive Abilities in Bilinguals with DS-ASD

In the current sample, one case study participant (Owain) had a significantly lower NV cognitive ability score than that of the TDB control group, although he was also the oldest case study participant at the time of data collection (16;8). No other significant differences were found between the DS-ASD case study participants and the three control groups. In contrast, Rhiannon was the youngest case study participant and she displayed the highest NV cognitive ability. This was also above the average of the two DS control groups (estimated to be higher than 91-94% of the control groups, although this did not reach statistical significance). This displays the variability in the cognitive profiles of children with DS, however, for this case specifically, the higher cognitive performance displayed by Rhiannon was likely due to the fact that she had mosaic DS as opposed to a full trisomy 21.

Overall, three out of the four children with a dual-diagnosis had lower cognitive profiles than the mean of each of the three control groups. On average, the two control groups of children with DS had a CA of 9;4, however, all of the children in the case study analysis were older than this, with the exception of Rhiannon who was younger (6;8). Given that three of the children in the case study were older, if the cognitive profile for children with DS was the same as those with DS-ASD, it would be expected that these would be similar

to or even slightly above the DS groups. Consequently, this finding is in line with previous findings and was expected according to the literature that documents lower general cognitive functioning in children with a dual DS-ASD diagnosis (Capone et al., 2005) in comparison to children with DS without ASD.

Language Abilities in Bilinguals with DS-ASD

In terms of language profiles, two of the DS-ASD case study participants showed significantly lower performance compared to the control groups. The significant effects emerged for Dylan who displayed lower performance on the English receptive vocabulary component compared to all three control groups. In addition, he also had lower performance on all other English language components compared to the TDB group, except for the tasks assessing expressive language. No significant differences were reported for his Welsh receptive language abilities. Significant differences were also found for Owain compared to the TDB control group only for English language content. No significant differences emerged between any case study participant and the three control groups for any PA measure.

In considering the age of first word, one DS-ASD participant had an age of first word that was significantly later than all groups (Catrin), however, one had a significantly earlier age of first word compared to the DSB group (Rhiannon). The final two case study participants had an age of first word that was comparable to the two control groups of children with DS and the TDB group. This finding is in contrast to what was reported by Warner and colleagues who did find a significant delay in the onset of the first word by around 6 months compared to those with DS alone. This may be due to the fact that Warner and colleagues employed a large-scale group design whereby across group comparisons were undertaken with large sample sizes. It is plausible that this finding would be replicated if a group analysis was employed as opposed to a case study approach, however, the aim of this study was to compare the four unique profiles of children with DS-ASD. Although single case study comparisons with control groups may be less likely to find significant results, this approach was taken to compare and contrast these language profiles due to the varying profiles in both DS and ASD and gain an insight into the uniqueness of the cases studied.

On the other hand, bilingualism may have influenced the age of first word for the children in this multiple case study analysis. The three children who were reported to have a first word at the expected 24 months of age or even before this had continued with bilingual exposure. In typical populations, bilingual children are reported to have their first word around the same time as monolinguals and within the typical time expected (Genesee, Paradis & Crago, 2004). As this was not a group study and no monolingual children with DS-ASD were included, it is unclear how bilingualism may have impacted on the age of first word. Nevertheless, in comparison to the control groups and other studies of children with DS-ASD, bilingualism did not seem to negatively impact the children with DS-ASD in this study. Furthermore, some of the children with DS-ASD had an earlier age of first word in comparison to the groups of children with DS, particularly Dylan who was reported to have an age of first

word at 12 months and Rhiannon at 15 months. Both of these children reached this milestone significantly earlier than the DSB control group. Further research is needed to examine the role of bilingualism on early language milestones in children with DS and children with more complex language impairments such as those with DS-ASD.

In general, the children with DS-ASD displayed lower performance on all assessments on average compared to the three control groups. An exception to this was again found for Rhiannon who showed greater performance than the DS group's averages for all language assessments in English. This is again likely to be a result of mosaic DS as opposed to trisomy 21, which was the sub-type that all other participants in the case studies and DS control groups had. Children with mosaic DS also typically do not experience language impairments to the same extent as children with trisomy 21 (Korbel et al., 2009; Papavassiliou et al., 2015; Zhao et al., 2015), however, limited information is available regarding the phenotype of mosaic DS in comparison to trisomy 21, particularly in relation to language. Furthermore, although only based on limited research, the degree of mosaicism (i.e. the number of cells containing the additional copy of the 21st chromosome) has also been found to correlate with the severity of cognitive impairment (Papavassiliou et al., 2015).

The finding that most of the case study participants with DS-ASD in the current study had lower English language abilities than that of all control groups is also in line with research conducted by Warner et al. (2014). These researchers also found that the children in their study who had DS-ASD performed lower on language assessments than children with DS only. In comparing the number of children at the single word level, there were significantly more children in the DS-ASD group. Furthermore, in the current study, there was a greater range of abilities in the children with DS-ASD than those with DS only. This was true for both developmental age and language abilities, however, the children's language abilities generally reflected their developmental level. For example, Dylan displayed the most substantial cognitive impairments and language impairments in English as he scored significantly lower than the TDB control group on all English assessments and significantly lower than the DSB control group for receptive language in English. In contrast, his Welsh language abilities were considerably stronger, indicating that he was Welsh dominant, as there were no differences between his Welsh receptive vocabulary and any other group. Owain was also reported to be Welsh dominant by parent report of current and lifetime exposure to Welsh and English. This was also reflected in the finding that his Welsh receptive vocabulary was not significantly different from the control groups, but he did perform significantly lower on the language content assessments on the CELF in English. Catrin performed lower than the control groups but not significantly so and Rhiannon demonstrated superior performance to the control groups but again this was not a significant effect.

It is interesting to note that both of the males with DS-ASD displayed the largest impairments in relation to language compared to the two females. Although it is only speculative to suggest that males may exhibit greater impairments if they have a dual DS-ASD diagnosis, this proposal may reflect the fact that ASD appears to affect males differently to

females (Mandy et al., 2012). Prior investigations of males and females with ASD have proposed that females with ASD generally do not experience language impairments to such an extent as males (Sturrock et al., 2019) and that ASD expresses itself differently in females leading to it often going undetected in females due to differences in the indicators of ASD and diagnostic criteria which are more accurate in detecting ASD in males (Goldman, 2013; Mandy et al., 2012). Further research would again be needed to identify if this was also the case in females with both DS and ASD in a larger sample.

The variability in performance justified the case by case approach that was taken with regards to this population specifically and can help to identify the individual differences that exist within these populations. Given that bilingualism is also a highly variable concept with a range of variables to consider, case analyses such as these can provide valuable insights into these fairly unique circumstances. Across these multiple-case studies, patterns did, however, emerge for all children with DS-ASD in terms of the PA assessments. Specifically, all children displayed the greatest impairments in the tasks which measured rhyme awareness and rhyme production. In contrast, all of the children with DS-ASD displayed the strongest abilities in the phoneme tasks. This was also the case for the three control groups, suggesting that the profile in children with DS (both bilingual and monolingual) and those with a dual DS-ASD displayed the same patterns of strengths and weaknesses. These difficulties appeared to also be evident in the younger TD children, suggesting that similar profiles are found across these populations, whereby these difficulties appear to persist for children with DS and children with DS-ASD to an even greater extent. These impairments were evident for all domains of PA which raises concerns about the development of reading and spelling in these populations, given that PA is reported to be strongly associated with reading and spelling attainment in those who are TD (Bradley, & Bryant, 1985; Kolinsky, Cary, & Morais, 1987; Bruck & Genesee, 1995) and also children with DS (Fletcher & Buckley, 2002; Hulme et al., 2012), although, to date, no research has addressed the association in children with DS-ASD.

Profiles of English language abilities also patterned across the four case study participants in terms of receptive versus expressive language. Receptive language abilities in English were consistently higher than spoken or expressive abilities in all case study participants, besides Dylan who performed similarly for both elements. This was also the case for the three control groups with them all displaying greater performance for the sub-tests which assessed receptive language. The gap between expressive and receptive language was also evident in the two control groups of children with DS in comparison to the TDB control group. This was also found for three of the case study children with DS-ASD who displayed substantially greater performance on the tasks assessing their receptive language abilities as measured on the CELF (see section 7.3.1). This was not the case for Dylan, however, his scores on the CELF were generally low on all sub-tasks showing that his English language development was considerably below that of all three control groups and also some of the children with DS-ASD. For the current population of children with DS-ASD, language development appeared to be more impaired than PA in comparison to the control groups.

This was also true for the DS control groups who displayed more difficulties with the CELF and Prawf Geirfa (for the bilinguals) than they did the PA tasks in comparison to the TDB control group. This may be a reflection of the fact that the PA tasks were specifically designed for the populations under study and were adapted in line with prior recommendations from previous research with children who have DS (see section 4.1.2 for a summary of these adaptations).

On the whole, from this multiple case study approach, it appears that the children with DS-ASD showed the typical profiles of development in terms of cognition, language and PA that would be expected in children with this particular dual diagnosis. The children who had continued to receive exposure to two languages did not appear to display any greater difficulties to that of the DS control groups, however, there was evidence that they displayed greater impairments in general in language development, particularly in comparison to TDB controls. This would be expected given the research that suggests that children with DS-ASD encounter additional impairments in their cognitive and linguistic development (Warner et al., 2014). Similar profiles were found in comparison to the DS control groups, suggesting that those with DS-ASD show similar trajectories of development to those with DS alone. This preliminary study consequently suggests that the literature which documents no additional adversity as a result of bilingualism for those with either DS or ASD can be applied to children with DS-ASD when considered on a case-by-case basis. The analysis of individual profiles showed that the children with DS-ASD had difficulties with language in general but that this was true for both of their languages. Finally, abilities in each language seemed to be commensurate with exposure to each language.

7.4.2 Limitations and Implications

The first limitation of this research is the small sample of children with DS-ASD who have received exposure to both English and Welsh. This is a reflection of the small number of children with DS who have been exposed to two languages and an even smaller number who have a dual DS-ASD diagnosis. This case study approach consequently offers a unique insight into the cognitive and linguistic profiles of these individuals in comparison to the three control groups with considerably larger sample sizes (with 50 children in the control groups overall). Given the case study approach taken, it is acknowledged that it is not possible to make definitive conclusions or make any generalisations. Although the findings should be considered preliminary, the case studies do provide a useful starting point in understanding bilingual language development in children with fairly severe developmental disabilities and substantial intellectual impairments. The small sample size is in line with, and a reflection of the prevalence of children with DS who also have an additional diagnosis of ASD. Alternatively, it is also possible that this sample may be even smaller than the population of children with DS as a whole as it may be harder to identify and to diagnose children with a dual-diagnosis if they are bilingual, particularly due to the lack of standardised bilingual assessments. A further possibility is that the small sample could be a reflection of the fact that children with DS who also have ASD may be advised to limit input to a single language

even more so than children with DS alone, given the cognitive and language impairments that are usually apparent in this population. Clinicians and researchers have expressed caution in making these dual-diagnoses too early (Buckley, 2005; Howlin, 2000) and have raised concerns with the appropriateness of diagnostic tools designed to assess ASD in children with other co-morbid diagnoses. In bilingual children, the use of these diagnostic tools may become even more unsuitable, given that they do not consider the amount of input children have received to each language, the consistency of input or the several other factors which are known to influence language outcomes in bilinguals.

Secondly, there were three control groups in which each case study participant was compared to. The study design would have potentially benefited from an ASD control group in order to tease apart differences in the development of children with both DS-ASD in comparison to the DS control group and an ASD control group. This was not possible for the current study as the focus was primarily on understanding bilingualism in children with DS and consequent to these aims, the control groups were bilinguals and monolinguals with DS. Future research that employs these types of designs should endeavour to recruit control groups of children with both DS and ASD as the literature documents a specific language phenotype for children with ASD. A control group of children with ASD would then assist in identifying if the children with DS-ASD were experiencing language impairments that are more in line with the DS or the ASD bilingual profile. Similarly, it was difficult to match the children in the case studies due to their unique cognitive and linguistic profiles. Although the three control groups were matched on NVMA, the control group children were more closely matched on chronological age with each other. Two of the children with DS-ASD had very low cognitive development which is likely due to the additional ASD diagnosis.

One final limitation of this study is the limited number of available standardised tests and the suitability of these assessments for the target population. This was especially the case for the Welsh language assessments as there is not currently any Welsh language assessment that has an equivalent English version. The single standardised language assessment in Welsh is the *Prawf Geirfa*, which was utilised in this study, however, this is designed for 7 to 11-year-olds and only has normed scores for children falling within this age range. Consequently, bespoke PA tasks needed to be designed in order to assess PA in Welsh, with an equivalent English version. Ideally, future research should use measures that have corresponding equivalents in different languages when possible in order to examine both languages of bilingual children with standardised measures. Nevertheless, this research aimed to undertake comprehensive language assessments in English and also obtained measures of Welsh receptive language and PA in Welsh. Given that Welsh is considered a minority language, this approach was deemed the most suitable given the lack of standardised assessment in Welsh to date.

One important implication arising from this work is that this research is the first to document children with DS-ASD and their bilingual capabilities. Given that concerns have been raised about the capacity of children with either DS or ASD to be bilingual, this novel

study has shown that even children with dual-diagnosis and substantial cognitive and linguistic impairments can be successful and functional bilinguals under the correct circumstances. Although one family did opt to restrict language input to English as they were advised to do so and did not speak Welsh themselves, the other three families had continued to expose their children to both Welsh and English from an early age and all were developing language abilities in line with their developmental age and in line with the amount of input they had received to each language. These case studies highlight the importance of considering each child on a case-by-case basis and ensuring that families and educators receive evidence-informed information so that each family can make an informed decision regarding language use as opposed to assumptions that children with complex DDs will not be able to develop skills in two languages. Policies should consequently reflect this approach and clinical practice (i.e. interventions and speech and language therapy) should be tailored to suit the case-by-case approach taken for each child in line with their linguistic needs.

7.4.3 Conclusion

In summary, this multiple single-case study has highlighted the unique language profiles of four children with a dual DS-ASD diagnosis who had varying experiences in acquiring Welsh and English. This novel piece of research has provided a new understanding of bilingual language development in an under-researched population who often display substantial impairments in cognitive and linguistic functioning. Findings show that these children had varied but considerable impairments in relation to their cognitive development and in their language abilities which is in line with research that shows that children with a dual DS-ASD diagnosis exhibit considerable challenges within these domains. Bilingualism did not appear to have any negative impact on the development of Welsh for any of the children and only one child with DS-ASD performed lower than both DS control groups for one English language measure. This was likely to be a reflection of the fact that he was Welsh dominant and only had limited input to English. All children appeared to be developing their language in line with expectations of language input and cognitive ability.

This is the first study of its kind to document bilingualism in children with DS-ASD to date, showing that bilingualism in this population is possible. Importantly, similar profiles to that of the monolinguals and bilinguals with DS were found, however, the impairments and delays were greater in the majority of the children with DS-ASD. This highlights the need for timely assessment and interventions which are appropriate for bilingual children with complex DDs. The case studies presented here may assist in understanding how bilingualism impacts development in these children and that the role of language input may be more important in children with DS-ASD. This factor should be carefully considered when an additional diagnosis of ASD is made, ensuring that full assessments of behaviour, social skills and language are conducted in all languages children are exposed to. Further research is needed to better understand this dual diagnosis which will assist in ensuring that children are supported in the best way in order to enable suitable cognitive and linguistic development.

Chapter 8: Discussion

This thesis sought to expand the current understanding of bilingualism in children with Down syndrome (DS). As children with DS often exhibit substantial language impairments and delays, some have questioned the capabilities of children with a developmental disability (DD), including children with DS, in acquiring two (or more) languages. This proposal, which is also referred to as the Cumulative Effects Hypothesis (Orgassa & Weerman, 2008) suggests that the cumulation of a language disorder and bilingualism will result in further language difficulties. Furthermore, as discussed in section 2.3, previous research reports that families have received advice to restrict language to a singular language with the view that this would lead to more successful language outcomes (Kay-Raining Bird et al., 2012; Kremer-Sadlik, 2005). The scope of this thesis was to explore this issue within the Welsh language context specifically, given that Wales is an officially bilingual country and that there are policies and provisions in place to support the Welsh language and promote bilingualism, particularly in young children (see sections 1.1.1 and 1.1.2.). As well as documenting the language profiles of bilingual children with DS in comparison to control groups of monolingual children with DS and comparable typically developing (TD) children, specific areas of linguistic development that are known to cause difficulties in those with DS have been targeted in this thesis. Profiles of children with more substantial and complex impairments as also presented in children with a dual DS and ASD diagnosis.

Preliminary evidence has been reported in studies examining bilingual language development, primarily in French-English speaking children with DS in a group of studies conducted by Kay-Raining Bird and colleagues (Kay-Raining Bird, Cleave, Trudeau, et al., 2005; Trudeau, Kay-Raining Bird, et al., 2011; Cleave et al., 2014; Kay-Raining Bird, Trudeau et al., 2016). Furthermore, as presented in Chapter 2, researchers have now explored bilingualism in a range of other populations, including children with Autism Spectrum Disorder (ASD) and Developmental Language Disorder (DLD). These studies, on the whole, report that children who have been exposed to a second language (at least as simultaneous bilinguals) show comparable language skills as matched monolingual children, with some studies even reporting differences in favour of the bilingual children in areas such as adaptive functioning (Valicenti-McDermott et al., 2013) and social interaction (Hambly & Fombonne, 2012). Research concerning children with DS specifically is, however lacking, particularly in comparison to the emerging literature in children with other DDs.

As children with DS display a particular language phenotype (as discussed in section 3.1.1-3.2.6), understanding this phenotype in children with DS who are acquiring two languages is crucial for enhancing the understanding of bilingualism in this population so that evidence-informed recommendations can be provided. At the same time, this research has important practical implications with the potential to impact educational provisions, policies, professional practice as well as furthering theoretical understanding. This research has provided a novel contribution to the field whilst expanding current understanding of

bilingualism in children with DS, filling current gaps in the literature that exists whilst providing important new information that has substantial practical implications. Together, the findings presented in this thesis have the potential to have a substantial impact in relation to policy, clinical assessment and intervention, professional recommendation and educational provisions within the context of Wales. This chapter will consider the findings highlighted in each chapter whilst addressing any limitations and further considerations that have arisen as a result of this research. At the end of this chapter, avenues of impact and future research will be discussed in relation to the areas mentioned above.

8.1 Summary of Main Findings

The primary aim of this research was to establish the linguistic profiles of Welsh-English bilingual children with DS in order to identify if there was any evidence to support the apparent claim that children with this DD are disadvantaged in any way by being exposed to a second language. Findings reveal that children with DS who are raised in bilingual environments show commensurate levels of English language abilities as English monolingual children with DS. Although no evidence of a bilingual advantage was found for these children (as might be expected for some aspects of development as discussed in section 1.1.4), the finding that they do not exhibit any further delays or impairments supports the proposal that children with DS are capable of acquiring two languages. These abilities are appropriate for their developmental ages and degree of language exposure, meaning that these children can flourish within bilingual families and educational systems.

Furthermore, similar profiles of strengths and weaknesses were observed in the profiles of all children with DS, independent of whether they had been raised in a bilingual or monolingual environment. Evidence was found for language and communication impairments in the children with DS compared to the developmentally matched TD children, as expected, but these impairments were no greater for children who received significant exposure to a second language (namely English or Welsh). Additionally, case studies of four children with a dual DS and ASD diagnosis (DS-ASD) showed that even children with multiple and fairly profound DDs were also capable of developing measurable abilities with no evidence to suggest that they were negatively impacted (in terms of language outcomes) by bilingual exposure. Table 40 and 41 summarises the findings of this thesis for the various aspects of language that were investigated in this research.

Measure	DS V TD	<i>p</i>	Bil V Mon	<i>p</i>	Interaction	<i>p</i>
CELF						
Core	DS < TD	.019	Bil = Mon	.094	No	.994
Receptive	DS < TD	.015	Bil = Mon	.390	No	.776
Expressive	DS < TD	.001	Bil = Mon	.101	No	.783
Content	DS < TD	.024	Bil = Mon	.119	No	.910
Structure	DS < TD	<.000	Bil = Mon	.302	No	.984
Phonological Awareness						
Syllable	DS = TD	.336	Bil = Mon	.078	No	.967
Rhyme	DS = TD	.229	Bil = Mon	.935	No	.283
Phoneme	DS = TD	.927	Bil = Mon	.58	No	.637
Overall	DS = TD	.478	Bil = Mon	.735	No	.568
Working Memory						
Digit Span	DS = TD	.066	Bil = Mon	.310	No	.387
Prawf Geirfa						
Welsh Receptive	DS = TD	.927	NA	-	NA	-

Table 40. Summary of results for language measures across language status and diagnosis. *Note:* Data in this table relates to the larger group matched sample. Bold indicate significant effects. (Bil = Bilingual, Mon = Monolingual)

Measure	DS V TD	<i>p</i>	Bil V Mon	<i>p</i>	Interaction	<i>p</i>
CELF						
Core	DS < TD	<.001	Bil = Mon	.131	No	.596
Receptive	DS < TD	<.001	Bil = Mon	.691	No	.387
Expressive	DS < TD	<.001	Bil = Mon	.135	No	.413
Content	DS < TD	<.001	Bil = Mon	.078	No	.811
Structure	DS < TD	<.001	Bil = Mon	.259	No	.896
Phonological Awareness						
Syllable	DS < TD	<.001	Bil = Mon	.664	No	.140
Rhyme	DS < TD	<.001	Bil = Mon	.106	No	.760
Phoneme	DS < TD	<.001	Bil = Mon	.352	No	.448
Overall	DS < TD	<.001	Bil = Mon	.987	No	.300
Working Memory						
Digit Span	DS < TD	<.001	Bil = Mon	.782	No	.349
Prawf Geirfa						
Welsh Receptive	DS < TD	.022	NA	-	NA	-

Table 41. Summary of results for language measures across language status and diagnosis. *Note:* Data in this table relates to the larger group matched sample and accounts for differences in chronological age. Bold indicate significant effects. (Bil = Bilingual, Mon = Monolingual).

8.1.1 Expressive and Receptive Language Abilities in Bilinguals with DS

This research aimed to build a language profile of expressive and receptive abilities for bilingual children with DS in order to examine the claim that bilingualism may impact on language acquisition in children who are experiencing language delays and impairments. Assessments comprised of standardised measures to assess language development in English, including assessments of expressive and receptive language abilities, language content and language structure. These were conducted alongside assessments of receptive vocabulary in Welsh for the bilingual groups. The language abilities of the bilingual children with DS were then contrasted to that of the three control groups after carefully matching for non-verbal mental age (NVMA), socioeconomic status and gender. The control groups were: monolinguals with DS, TD bilinguals and TD monolinguals.

Findings show that after accounting for developmental and chronological ages, the bilingual children with DS performed comparably to the monolingual children with DS. There were no differences in the language abilities of these groups after undertaking appropriate statistical analyses for measures of language structure, core, expressive, receptive language and language content. At the same time, the bilingual children with DS also performed comparably to the TD control group on the assessment of Welsh receptive vocabulary. The finding that bilingualism did not have a detrimental impact on language abilities extends and substantiates previous claims made by researchers in Canada who have explored language abilities predominantly in French-English bilinguals with DS (Kay-Raining Bird et al., 2005; Feltmate & Bird, 2008b; Cleave, Kay-Raining Bird & Trudeau, 2014). These studies also report that bilinguals with DS did not have any lower language abilities in their L1 compared to monolinguals with DS. The current research strengthens these findings by Kay-Raining Bird and colleagues and expands on them by documenting language outcomes in a novel linguistic context. In the current study, many of the bilingual children were acquiring a minority language (i.e. Welsh) as their first language and the majority language (English) as their second or were balanced bilinguals. In previous studies, the children were dominant in the majority community language (English) and were most often acquiring French (also a majority language) as their second or joint first language.

Additionally, the population under study in the current research were chronologically older than the children in Kay-Raining Bird et al.'s study (2005). The children in the current research had an average chronological age of 9;6 in comparison to an average age of 6;1. This study has furthered understanding of bilingualism in children with DS by providing information on the language profiles of children at a later stage in language development and children at a higher developmental stage. Additionally, this is the first group study of bilingualism in children with DS where all the children were acquiring the same two languages. A further finding that was replicated in the current research was that of a specific language phenotype in children with DS, whereby the children with DS showed marked language impairments in comparison to the TD control groups. This was expected and coincides with previous research that documents specific difficulties with various aspects of

language for those with DS. This was particularly the case for aspects such as speech production and expressive morphosyntax, which is again in line with previous literature (Andreou & Katsarou, 2013; Chapman et al., 1998; Martin et al., 2009). In the current study, the areas which appeared to be most challenging were the assessments of word structure and the recalling sentences subtest of the CELF-P, both of which measure expressive language and expressive morphosyntax.

Results from this thesis also substantiate previous claims of a specific language profile in those with DS when it comes to expressive versus receptive language abilities (Chapman et al., 1998), whereby receptive language abilities are usually higher than expressive abilities. Although expressive language in Welsh was not directly assessed in this study due to a lack of appropriate measures, English receptive and expressive language scores for both groups of children with DS (and also the four case studies for children with DS-ASD) found that receptive language abilities were consistently higher than expressive language skills. This was specific to the DS groups as the TD children often showed equivalent expressive and receptive skills. The finding that both the monolinguals and bilinguals with DS showed this profile suggests that this general profile of development can be extended to bilinguals with DS also. It is anticipated that this profile would also be apparent in Welsh, however, further research would be needed to substantiate this proposal.

Furthermore, the findings in Chapter 4 also explored the recommendations that parents had received concerning bilingual exposure and any advice received in relation to the language of instruction. Previous research suggests that parents or caregivers of children with a DD may be advised to limit language input to a singular language with the hope that this would result in better outcomes (Kremer-Sadlik, 2005; Kay-Raining Bird et al., 2012; Marinova-Todd et al., 2016b). The findings of this study, suggest that these concerns are not substantiated by the empirical evidence presented as the bilinguals did not show any evidence of being disadvantaged in either their L1 or their L2. Additionally, if bilingualism did have any negative impact on language outcomes, it would be expected that these would be most observed in aspects of linguistic development that are specifically impaired. Given that no additional challenges were documented in areas such as expressive language and morphosyntax, this further strengthens the conclusion that those with DS should not be recommended to avoid bilingualism, and can flourish within Welsh-medium settings.

In the current sample, findings suggest that the majority of parents who have children with DS do not receive any advice or information relating to bilingualism, with only a few reporting positive or negative recommendations of bilingual language exposure. This may be due to several factors. Firstly, as the Welsh language holds an official status and recent government legislation suggests that bilingualism should be provided “as far as reasonably practicable” and “according to the needs and wishes of the pupil and the parents” (see section 1.1.2; Welsh Government: Special Educational Needs Code of Practice - Paragraph 1.7, 1993), practitioners may be cautious in making recommendations that conflict with these policies. Secondly, some families may feel that bilingualism is a necessity (as proposed by De Houwer,

1999) to them and their families in order to communicate in a natural home environment and may not have considered restricting language input to English only if Welsh is their native language. Of concern is the large number of families who reported not receiving any information or guidance at all on this topic. Although the Royal College of Speech and Language Therapists (RCSLT) recently acknowledged that families should “never be advised to abandon home language in favour of the majority language (typically English, Welsh or Gaelic) under any circumstances” (Bilingualism-Guidance Key Points: RCSLT, 2019, p.26), this policy does not stipulate that families should be supported and encouraged to use two languages with their children if they wish (e.g. in the case of English speaking families wishing for their children to attend Welsh-medium schools). In order to ensure that evidence-informed recommendations are made in the future, it is important that the findings of this research and similar research are communicated with relevant organizations and individuals where appropriate to inform the advice that families receive.

Finally, in this chapter, analyses were also undertaken to explore the extent to which factors such as chronological age (CA), working memory (WM), current language input and parental socioeconomic status (SES) related to language outcomes in each of the four groups. These analyses aimed to explore the relatively large individual differences that are often observed in the language and cognitive outcomes of children with DS that were also observed in this research (Roberts et al., 2007; Tsao & Kindelberger, 2009). These analyses found that for both TD and DS groups, the significant predictors of language outcomes were WM, NVMA, CA and PA. These accounted for 81% and 90% of the variability in language abilities in these populations respectively. For both the DS groups, WM was the strongest predictor variable, whereas, for the TD groups, NVMA was the strongest predictor. This also coincides with previous findings that document that NVMA plays less of a role in determining language outcomes in DS (Abbeduto et al., 2003) relative to WM. This research has extended this finding to bilinguals with DS. This may have important clinical implications which are discussed later in section 8.3.1.

8.1.2 Phonological Awareness in Bilinguals with DS

Previous research suggests that PA appears to be disproportionately affected in those with DS (Cossu, Rossini & Marshall, 1993; Fletcher & Buckley, 2002; Næss et al., 2015; Næss, 2016). At the same time, researchers have identified that some studies report that TD children who have been exposed to two languages outperform monolingual children on tasks which tap into PA (Campbell & Sais, 1995; Bialystok et al., 2003; Chen et al., 2004; Canbay, 2011). As a result, this chapter aimed to examine the PA abilities in bilinguals with DS in order to explore the possibility that these bilingual benefits may also emerge for this population. Bilingual children with DS were assessed on six bespoke tasks of PA in English as well as tasks that were designed to be equivalent measures in Welsh. The three control groups described above also completed these tasks to enable between-group comparisons, with the tasks designed in line

with recommendations from previous research in making the tasks suitable for use with children with DS and young TD children.

Findings reveal that for PA as a whole, there were no significant differences between the bilinguals and monolinguals. This was true for both the DS groups and the TD groups. The trajectory of development of PA in bilinguals with DS appeared to coincide with the profiles of the monolinguals with DS as both groups displayed specific difficulties with rhyme level assessments. Similarly to the findings above, an effect of diagnosis was found in that the two groups of children with DS performed lower than the TD children after controlling for CA, as anticipated. This finding supports the results of the only other group study that has investigated PA in TD children and children with DS that matched for NVMA (Boudreau, 2002). In their study, Boudreau and colleagues reported that the TD children performed significantly better than the DS groups for rhyme judgement specifically, a finding that was replicated in the current research. Several other studies have documented a deficit in rhyme judgement and production in those with DS (Cardoso-Martins et al., 2002; Kennedy & Flynn, 2003; Hulme et al., 2012). Possible explanations for this include a specific weakness in attending to word endings (Snowling et al., 2002) which may also assist in partly explaining other deficits observed in those with DS, such as difficulties with morphosyntax (which also rely on attending to word endings).

The only evidence of a bilingual advantage was that the TD bilinguals outperformed the TD monolinguals on the syllable awareness sub-task. Although there was no evidence to suggest that the bilinguals with DS displayed any advantages on these aspects of PA, importantly, there was no evidence of any disadvantage to these aspects. Given that children with DS are reported to have substantial difficulties in the development of this skill, the fact that no further disadvantages were documented substantiates the findings in the previous section (i.e. bilingualism does not further impede language acquisition in those with DS.) As the development of PA plays a crucial role in the later development of reading and spelling (Fletcher & Buckley, 2002), the finding that bilinguals do not result in any further difficulties in this area also has important implications on educational and clinical practice which are presented later. Although no clear evidence of a bilingual advantage was found (besides for one sub-task for the TD groups), as noted in section 6.4.1, an “absence of evidence is not evidence of absence” (Altman & Bland, 1995). Some possible theoretical explanations as to why this bilingual advantage may not have been observed in this specific context are discussed in section 8.3.3.

One further factor that should be considered in relation to phonological awareness development for children with DS and TD children is the role of literacy instruction. The participants recruited for this research ranged in ages as the children with DS were matched on NVMA to the TD children. Consequently, the TD children were significantly younger than the children with DS and some had only just begun schooling with very limited literacy instruction. In contrast, the children with DS ranged in ages of between 5-16 years old. Many of the children with DS would have been receiving literacy instruction through their

schooling for several years, meaning that they may have been at an advantage over the TD children. Although the results do not evidence this suggestion, this may partly explain why the children with DS performed comparably to the TD children. As children with DS are reported to have a specific weakness in the development of PA, the finding that there was no significant difference between the TD children and the children with DS is somewhat surprising. The role of literacy instruction and the longer duration of schooling that the children with DS had, may therefore partly explain why this was not the case (also see section 8.2. below).

8.1.3 Bilingualism in Children Dually-Diagnosed with DS-ASD

Chapter 6 documented the language profiles of children with DS and ASD who displayed complex and unique developmental profiles and also received varying exposure to Welsh and English. Prior research has evidenced that children with DS have a higher prevalence rate of ASD than the general population (DiGuseppi et al., 2010; Richards et al., 2015). Given the worldwide increase of bilingualism and the unique language phenotype observed in children with DS-ASD (section 7.1.2; Molloy et al., 2009; Warner et al., 2014), research that identifies how bilingualism impacts on children with this specific dual diagnosis is needed. No single piece of research to date has reported on bilingualism in children with DS-ASD. In the current research, the language profiles of four children with DS-ASD were documented who had received varying exposure to both English and Welsh across their lifetimes. These profiles were descriptively compared to each case study participant, alongside statistical analyses to identify differences between the three control groups: bilinguals with DS, monolinguals with DS and TD bilinguals.

An overall prevalence rate for DS-ASD in the children with DS who were recruited for the research was 12.5%. This coincides with previous estimates which suggest a prevalence of between 5-37.7%, with most recent and reliable estimations suggesting 16-18% (DiGuseppi et al., 2010). This finding suggests that DS-ASD was observed in the bilinguals in the current study at similar rates to that of monolinguals with DS, and is also concurrent with reports that the prevalence is higher in children with DS compared to the general population which is reported to be around 1.1-2.24% (Zablotsky et al., 2015). Also in line with previous literature is the finding that the children with DS-ASD showed lower general cognitive development (as measured by the NV subtest of the KBIT-II) on the whole compared to the other groups of children with DS, although only one case study had a significantly lower performance for the NVMA assessment. This has been documented previously by Capone and colleagues (2005) who reported that 87% of the children with DS-ASD had severe-profound intellectual impairments. The exception to this was the participant with DS-ASD in the current study who displayed an unusually high cognitive profile, however, she did have mosaic DS as opposed to a full trisomy 21 which may explain why this was the case. Researchers have previously reported that children with mosaic DS do not tend to have as substantial cognitive impairments (Korbel et al., 2009; Zhao et al., 2015) and the difficulties

that these children do have tend to be in line with the degree of mosaicism (i.e. the number of cells containing the additional 21st chromosome; Papavassiliou et al., 2015).

In terms of early language abilities, on average the participants in the case study had an age of first word that was similar to that of the control groups of children with DS at around 24 months, with some being earlier (12 months) and others being later (48 months). For expressive and receptive language abilities, the majority of the case study participants displayed equivalent language abilities in English and Welsh as the three control groups, suggesting that bilingualism did not have a negative impact on language acquisition for this sample of children with DS-ASD. The exception to this was the first case study participant, whereby he displayed similar levels of English language abilities of the two DS groups, but consistently performed lower than the developmentally matched TD control group. This may be due to the fact that he had the lowest cognitive ability or a reflection of the fact that he was Welsh dominant. For Welsh vocabulary, he did not perform significantly lower than any other group. For the final case study participant with DS-ASD (mosaic DS), she consistently performed higher than control groups on cognitive and linguistic assessments in English and performed similarly to the control groups for her Welsh receptive vocabulary.

For measures of PA, the case study participants displayed similar levels of abilities to all three control groups with no significant differences emerging for any participant for any measure of PA. At the same time, similar profiles were found for the children with DS-ASD and the monolinguals and bilinguals with DS. A specific deficit in being able to identify and produce rhyming word pairs was documented, which also further extends previous reports that this area is challenging for children with DS (Cardoso-Martins et al., 2002; Kennedy & Flynn, 2003; Hulme et al., 2012). This finding suggests that this is also the case for the children with DS-ASD who are bilingual. Similar patterns were also found in terms of expressive and receptive language abilities for the DS control groups and the case study participants with DS-ASD. Receptive language abilities were higher for all case study participants than expressive language (besides participant 1 who displayed similar expressive and receptive language), which was parallel to that of the children with a single DS diagnosis.

On the whole, similar profiles were found for the bilingually exposed children with DS-ASD compared to the monolinguals with DS, suggesting that this additional diagnosis did not limit their ability to acquire the two languages, for those who had received consistent and ongoing exposure to both languages (for three out of the four participants). It is worth highlighting that it is difficult to meaningfully interpret the findings of these case study participants and that the results are not generalisable to all children with DS-ASD due to the heterogeneity of the participants. This reflects the approach taken in this chapter whereby the children were presented in a case by case approach in order to highlight their individual circumstances. This is especially true for Catrin as she has mosaic DS and therefore this highlights the unique circumstances regarding her cognitive and linguistic phenotype and her experiences regarding bilingualism.

It was clear that further language and communication impairments were evident by the finding that two of the case study participants performed significantly lower than the TDB group for some measures. The case by case approach highlighted the variability in cognitive and linguistic outcomes for children with DS-ASD, even more so than for children with DS only. Some children may thrive in bilingual environments whereas others (such as the participant who was reported to have limited speech in English or Welsh at age 4 and subsequently discontinued bilingual exposure) may encounter more difficulties with language in general, meaning that learning two languages would also be difficult. These case studies have contributed valuable information that documents that some children with DS-ASD are capable of being bilingual given the correct circumstances and support. These findings should be considered as preliminary and explorative, and it is possible that bilingualism will be more appropriate and successful for some children with DS-ASD than others.

8.2 General Discussion

The primary finding presented in this thesis is that there was no evidence to suggest that bilingualism resulted in any disadvantages for children with DS. There were no differences observed for important elements of language for the bilingual children with DS in this study versus children with DS who had been raised in a monolingual environment. These measures included assessments of receptive and expressive speech, language structure and content, and PA. Taken together, the findings presented throughout chapters 4-6 have provided a unique and novel insight into the language profiles of Welsh-English bilingual children with DS who had a range of backgrounds with different levels of intellectual abilities. This thesis also included a case study analysis of four children with dual DS-ASD diagnosis who received bilingual exposure, an area of research which was previously completely absent in the literature. As with all children, adequate exposure to both Welsh and English is necessary to develop abilities in both languages and this was highlighted by the finding that lifetime input to Welsh was significantly associated with Welsh receptive vocabulary abilities in the bilinguals with DS and the TD bilinguals. This is in line with literature that reports that the role of input and SES are both important predictors in young Welsh-English bilingual children (Gathercole, Kennedy & Thomas, 2016).

This thesis has highlighted that children with DS in Wales can and do become bilingual and can be fully included in mainstream Welsh-medium schools. Prior research has also documented that children with DS can have success with bilingualism (Kay-Raining Bird, et al., 2005; Feltmate & Kay-Raining Bird, 2008; Kay-Raining Bird, Genesee & Verhoeven, 2016b), however, prior research had only documented children attending schools through the majority language. The current research has extended on previous literature by exploring bilingualism within a novel population where the language status is considered vulnerable (Moseley, 2012). There was a fairly large range of ages and consequently a large range of variation in the children's language abilities, particularly for those children with both DS and ASD. This has been reported for children with DS previously (Oliver & Buckley, 1994; Roberts

et al., 2005), it is worth considering these individual differences in future research. In a study conducted by Zampini and D’Odorico (2009) that explored vocabulary and gesture development in children with DS, the child who had the highest level of development made the greatest amount of gains throughout the research. This highlights that the developmental progression may not be at the same pace for all children and it is likely that this is also the case for bilingual children with DS.

In line with the findings of this research and previous research that generally reports that bilingualism is not detrimental to children with a DD, parents of a child with a DD should be provided with information about bilingualism and the expected outcomes. If parents are recommended to avoid bilingual exposure, then they themselves may internalise the belief that two languages would be too big of a challenge for their child and that it would lead to negative consequences on language development. This may have further repercussions on families who may already be in a challenging position. Families may believe that they have to decide between their child learning a different native language (in some cases) or their child having greater language impairments if they are learning two languages. As discussed later, this view should be amended so that bilingualism for children with a DD is not seen as a problem or an additional challenge but that bilingualism can be a useful resource as it can be for TD children and adults. If children with a DD have access to bilingualism, it is reported that this can assist with emotional development (Howard et al., 2019). Identifying the expected outcomes for bilingual children with DS is also important so that any child who is experiencing language impairments that are not in line with this profile can be identified and receive appropriate and targeted support.

One further interesting point to note is that the children with DS performed comparably to the TD children on measures of PA before controlling for CA, however, for the expressive and receptive language assessments, they did not. When the children with DS were matched on NVMA to the TD children, they performed equally as well on the PA assessments. Consequently, this finding suggests that PA develops independently to the other areas of language that were investigated in this thesis including grammar and vocabulary. This contrasts with previous research which suggests that PA develops alongside vocabulary, at least in TD children (Singh et al., 2012). This also provides support for the modular approach to language which suggests that there are various ‘modules’ to language as opposed to it being a single entity (Perovic, 2006). Alternatively, it could be the case that the specific assessments used to measure PA led to these findings, however, this was not replicated in the groups of TD children as they performed comparably on the language measures and the PA measures. As the PA assessments were specifically adapted for use with children with DS, this may have provided a more accurate assessment of PA than previous studies who have suggested a specific weakness in the development of PA (Cossu, Rossini & Marshall, 1993; Fletcher & Buckley, 2002).

As highlighted at the outset of this thesis in section 1.1, bilingualism is not a concept that is easily definable or concrete. This is particularly true for young children and children

with a DD who may still be in the process of acquiring their languages. One clear distinction that is usually made is that of simultaneous versus sequential bilinguals. All children in this study were considered as simultaneous or early sequential bilinguals, given prior definitions and distinctions (Paradis et al., 2011). There may, however, be subtle differences in the language outcomes of children who acquire Welsh and English from birth at home and those who are introduced to Welsh slightly later with English as the home language (or vice versa). Further research is required to investigate if this is the case (as discussed in section 8.5). As bilingualism is multifaceted, several important avenues are related to supporting the language development of children with DS, as highlighted by Ware and colleagues (2015). These include clinical assessment, appropriate training, appropriate academic progress, parental advice, language development and policy implications. Many of these facets have been explored in this thesis and are address further in the following section.

8.3 Implications and Impact

This research has been guided by the absence of information in the literature and the research that documents a discrepancy between clinical practice and research evidence in the field of bilingualism in children with DDs. As a result, there are several substantial and important implications arising from this work. The implications expand across three key areas: clinical practice and recommendations, educational provisions and policy, and theoretical implications. These are discussed throughout the following sections.

8.3.1 Implications for Clinical Practice

The first aspect of this thesis that has the potential to have important implication relates to professional recommendations and clinical practice. As highlighted in the literature review in Chapter 2, it appears that parents/caregivers of children with ASD or DS may receive recommendations to restrict language input to the majority community language only, with the view that restricting language to a single language would lead to better language outcomes. In direct conflict to this, is the research that has been conducted which empirically compares the language outcomes of bilinguals and monolinguals with these DDs that frequently converge on their findings that bilingualism does not lead to any further language delays or impairments. The results from this thesis replicate these findings with no evidence of any detrimental impact of bilingualism for Welsh-English bilinguals with DS compared to monolinguals with DS and also TD bilinguals and monolinguals after considering NVMA, CA and SES. Recommendations provided to these parents were also briefly explored in Chapter 4 which suggested that in Wales, practitioners do not appear to provide supportive or negative views of bilingualism on the whole. The majority of parents of children with DS reported that they did not receive any information on the subject, meaning that these parents may have been required to make their own judgements without any information or guidance as to how their children would progress in a bilingual setting.

Given the results presented in this thesis, parents of children with DS should not be advised by clinicians to avoid bilingual exposure as this is not an evidence-informed recommendation. Families who seek information and advice about bilingualism, whether this is as a home language or as a language learnt in an immersion setting, parents or guardians should receive information in relation to the outcomes of this research and previous research. The converging evidence in this thesis and previous studies suggests that children with DS (Kay-Raining Bird et al., 2005), ASD (Hambly & Fombonne, 2009; Drysdale et al., 2015; Kay-Raining Bird, et al., 2016c) and DLD (Paradis & Crago, 2000; Paradis et al., 2003) can all successfully become bilingual with no detrimental impact on linguistic abilities under the correct circumstances. Therefore, clear guidelines need to be provided to clinicians so that these can be provided to families who have these concerns in the future. Practitioners such as speech and language therapists are often in direct contact with these families and the most likely clinicians to be in a position to provide recommendations. Positive changes have been made in line with research to date with recent guidelines provided by the RCSLT which state that “Parent(s) and carers should never be advised to abandon home language in favour of the majority language (typically English, Welsh or Gaelic) under any circumstances. Such an approach is not supported by the evidence base and leads to poor speech, language and communication outcomes for the child, and poor social outcomes for the child, family and wider community” (Bilingualism-Guidance Key Points: RCSLT, 2019, p.26).

Although these guidelines are a promising step in aligning key guidance with the empirical literature, these guidelines may (and possibly inadvertently) suggest that parents from English speaking families should not be encouraged to place their children in a Welsh-medium educational setting as this could also be considered as abandoning the home language. The RCSLTs also further states that it “recognises that bilingualism in a child, young person or adult is an advantage.” Although the guidelines do not yet state that children with a DD should have the right to access bilingual services, the fact that speech and language therapists (who are governed by the RCSLT) are now required to encourage bilingualism for families whose native language is different to that of the majority community language (English) is a positive change which has been made as a direct result of the growing body of research in this field. It appears that these recommendations now appreciate that the impact of removing a home L1 may not be appropriate and could potentially result in worse outcomes if this means that the child is not receiving a rich language input at home (due to parents using a non-native language only) combined with a sudden shift in language use in the home. Removing the home language has been reported to reduce the quantity of language input in some circumstances (Ijalba, 2016), to result in parents feeling less comfortable in communicating with their child (Hampton et al., 2017) and may also have implications on social development (Howard et al., 2019) as well as impacting early parent-interaction styles (Hudry et al., 2017).

One further Implication arising from this thesis that is relevant for clinical practice relates to the way that language development is assessed and monitored in bilingual clients

during SLT. It may be difficult for clinicians to disentangle language disorders in bilingual children as they will have a unique language history which may influence their development in each language depending on the languages being acquired, each child's exposure to each of those languages including the context, duration and consistency of language exposure. This is further complicated by the lack of standardized assessments that are suitable and normed for bilingual children, as these are often standardised on monolingual populations. Assessment criteria and cut off points for language impairments may inadvertently overestimate the number of bilingual children presenting with language disorders. Related to this is the importance of evaluating all the languages that a child is in the process of acquiring. Language disorders (if present) will be apparent in all the languages that a child has been exposed to, and assessing all languages will provide a greater insight as to whether or not there is any evidence of an underlying language impairment in bi/multilingual children. This may, of course, present pragmatic challenges in that clinicians may not have the resources available to assess all the languages of each child (or of course not have suitable therapists to deliver assessments in other languages). It is essential that clinicians are at least aware of this factor so that the actual abilities of each child are not under-estimated by bilingualism.

Finally, the results from this thesis may have implications on the language used in SLT. Generally, SLT would be delivered in English, being the majority community language in the UK. Given that the findings of this research suggest that bilingualism is not detrimental to language development, SLT may be more beneficial if this is available in both of a child's languages (i.e. English and Welsh). This may be particularly beneficial if there is a possibility that this would lead to transfer effects, thus enhancing linguistic abilities. Although there may be a limited number of qualified speech and language therapists who are capable of delivering SLT in Welsh, this may be particularly useful for younger children who may only have limited exposure to English. Furthermore, there is some evidence to suggest that SLT may be more effective if it is initially delivered in a child's first language, at least for a bilingual child with ASD (Seung, Siddiqi & Elder, 2006). As language and communication interventions are reported to be more successful if they are introduced earlier in children with DS (Aparicio & Balaña, 2002), clinicians should endeavour to implement any bilingual interventions as early as possible. Although more research is required to evaluate the outcomes of bilingual interventions for children with DS, it is pertinent to consider bilingual SLT services for bilingual children with DS where possible, particularly for those children who are first language Welsh speakers.

8.3.2 Educational and Policy Implications

The second area that this thesis has the potential to impact relates to educational provisions and policy guidelines for bilingual children with DS in Wales. Currently, there are no clear guidelines for parents and schools as to the inclusion of children with various DDs, such as DS and ASD within bilingual services. The Welsh Government has policies which state that provisions should be available in Welsh or English "in line with the children's linguistic needs

as an integral principle of inclusive practice” (Welsh Government: Inclusion and Pupil Support, 2017), and that bilingual or Welsh services should be available for children with a SEN “as far as reasonably practicable” (Welsh Government: Special Educational Needs Code of Practice - Paragraph 1.7, 1993). A further recent review of the use of Welsh in education suggested that the Welsh government has previously noted that there is a lack of specialist support through the medium of Welsh for families of children with a SEN (A rapid review of the Welsh in education strategic plan; Welsh Government, 2017). Although these policies seem to suggest that children with SEN could be included in Welsh-medium or bilingual services, they do not make this message very clear. More specifically, the statement “as far as reasonably practicable” could be considered very subjectively as what one family considers as reasonably practicable may not be in line with what schools believe fit this criterion. Furthermore, none of these policies differentiate between the nature of the SEN that is being referred to. Future policies should consider the findings from research relating to bilingualism, such as those presented in this thesis in order to adapt current policies so that these clearly state how schools should approach families with specific DDs in relation to including them in bilingual or Welsh-medium schools.

Additionally, these policies generally seem to refer to families from Welsh-speaking backgrounds who have children with a SEN. Consequently, English-speaking families who wish for their children to access Welsh-medium education (i.e. elective bilingualism) may not be supported in their decisions due to a lack of guidelines for these families specifically. There are many possible reasons why families may want their children to attend immersive nurseries and schools, such as giving them the same opportunities to learn the Welsh language and to be included alongside siblings and friends. In addition, current policies either do not state whether children with more complex and severe DDs should also receive the same guidance compared to children who may have a mild SEN. Previous research suggests that parents of children with severe learning difficulties may be advised to avoid bilingual exposure even more so than other children with a learning disability or DD and it would not be likely that they would be included in bilingual opportunities to the same extent according to a recent study conducted by Valenzuela et al. (2016).

The severity of cognitive and language impairments leads to a separate concern, which is that these children are more likely to be placed in SEN schools if mainstream schooling is not appropriate to meet their needs. In Wales, there are very limited Welsh-medium services available within SEN schools with only two schools in the country who report that they facilitate Welsh-medium provisions. This results in some families not having the option to educate their children through the medium of Welsh if they are not located near one of these schools, which is in direct conflict to their statement which states that “no one should be denied opportunities for Welsh-medium education or learning Welsh as a language because of their race, ethnicity, disability, gender, sexual orientation, age or religion” (Welsh Government, 2014, p.12).

In order to ensure that all families have the option to choose Welsh-medium education, there needs to be an increase in Welsh-medium provisions for those with more profound needs, such as those with DS-ASD. This may be especially beneficial for children from Welsh-speaking homes as these children may otherwise be at risk of L1 attrition if these children do not have adequate input to Welsh (if they have to attend English-medium schools) given that Welsh is a minority language. At the same time, clearer policies need to be put in place to support schools and parents in choosing appropriate educational provisions for these children. Finally, research has documented that there may even be benefits to the children themselves if they are included in Welsh-medium schools (Howard, Katsos & Gibson, 2019), whereby those attending Welsh-medium schools were reported to be more socially active and had a more favourable view of bilingualism. Given that the findings in this thesis report that children with DS and in some cases children with DS-ASD are not disadvantaged by bilingual exposure, policies should reflect this and take into consideration the research-evidence to date in this field.

8.3.3 Theoretical Implications

Finally, several important theoretical implications can be drawn from the research presented in this thesis. The first relates to bilingual capacities for children with DDs, and more specifically children with DS. As children with DS show a specific language profile with a range of language impairments and delays, this has consequently led to uncertainty as to whether these children will be impacted by bilingualism. Prior research suggests that some hold the belief that bilingualism will have a detrimental impact on this population with the viewpoint being if one language is hard then two languages would be too hard. The findings presented in this thesis show that this does not seem to be the case for Welsh-English bilingual children with DS. Additionally, if bilingualism did impact negatively on language development, it would be anticipated that this would be most evident in aspects of development which are specifically compromised in children with DS. The finding that no differences emerged for aspects such as expressive language and PA support this view. It appears that similarly to TD children, bilingualism does not negatively impact on several aspects of language development (Fabiano-Smith & Barlow, 2010) after an initial smaller vocabulary in each of a child's languages when considered separately (Bialystok et al., 2010). The current findings show that bilingualism does not compound language impairments that are observed in children with DS, and contributes to previous research which also documents this finding in children with other DDs. Therefore, this research does not support the proposal of a Cumulative Effects Hypothesis (Orgassa & Weerman, 2008), that is, that the cumulation of language impairments and bilingualism would result in further difficulties delays in language acquisition.

Secondly, the findings from this thesis have theoretical implications concerning theories of PA and research that relates to the bilingual advantage hypothesis (see Chapter 5). Prior research has proposed several theories in an attempt to explain the finding that children who have been exposed to two languages show enhanced performance on tasks

assessing PA (Bialystok, Majumder & Martin, 2003; Verhoeven, 2007). In this thesis, only very limited evidence to support the proposal of a bilingual benefit was found, which was for one task (syllable detection) and for the TD children only. In considering PA as a single ability, no bilingual advantage was observed for either the TD children or the children with DS. One possible explanation for this lack of effect is provided by Cross-Language Transfer Theory. (Durgunoğlu et al., 1993). Within this theory, it is proposed that positive transfer (i.e. acceleration) will only occur if the L1 being acquired is phonologically and orthographically more complex than the L2 (Loizou & Stuart, 2003), but not the other way around. As English is more complex than Welsh, particularly in relation to orthographic transparency, in Welsh-English bilinguals, English would need to be the L1 for these benefits to be observed, according to this theory. In this thesis, the majority of the bilingual children were acquiring Welsh as their first language in the home, meaning that positive transfer would not be expected.

Furthermore, the only other study to document bilingualism and PA abilities in an individual with DS was provided in a single-subject case study (Burgoyne et al., 2016). In this research, biliteracy and various measures of PA were undertaken to explore the impact of bilingualism, however, no bilingual benefit was reported either. In this case, the 7-year-old was acquiring Russian as her first language and English as a second language. Although she displayed proficient abilities in both languages which were in line with expectations given her developmental age and with comparable abilities as the control groups, with performance on the PA measures not at any higher than the monolingual control group with DS. It is possible that the bilingual benefit does not transfer to this population (i.e. children with DS) due to differences in processing mechanisms, or it is possible that in this case, as English was also being acquired as the second (and more phonologically and orthographically complex) language a positive transfer would not be anticipated in this situation according to this theory either. Moreover, according to the Lexical Restructuring Hypothesis (Metsala & Walley, 1998), the bilingual advantage emerges as a result of increased vocabulary and an increase in potential competing words which leads to more fine-grained representations and the development of PA. This also requires the two languages to be phonologically similar to each other for these benefits to emerge. This theory also offers a possible explanation as to why the bilingual advantage was not documented in this case study or this thesis, given the differences in grapheme to phoneme consistency.

A further interesting finding that has possible theoretical implications is that the children with DS showed larger differences between syllable, phoneme and rhyme scores (whereby rhyme scores were substantially lower, significantly so for phoneme and rhyme components) in comparison to the TD control group (see Chapter 5). This supports previous findings which suggest that children with DS have a specific weakness in rhyme abilities (Cardoso-Martins et al., 2002; Kennedy & Flynn, 2003; Hulme et al., 2012), which may be due to a specific deficit in attending items that are in word-final position (as discussed in section 6.4.1; Næss, 2016). Furthermore, this suggestion may also assist in partly explaining some of

the deficits which have also been observed in expressive and receptive morphosyntax. This also appeared to be the case for the children with DS-ASD reported on in Chapter 6. In the current research, fairly large differences were also found in the four children with DS-ASD in terms of differences between each component of PA. Although these patterns are only based on numerical differences as opposed to any statistical significance, it is intriguing that this profile of development across the various components of PA in bilinguals also appears to have been extended to the children in this study who have both DS and ASD.

Finally, the findings reported in this thesis may also contribute to the proposal that the linguistic situation in Wales specifically, is the reason why no bilingual benefit emerged in the current research in terms of PA. Researchers have failed to replicate the apparent bilingual advantages for executive function and metalinguistic abilities in large samples of Welsh-English bilinguals (Gathercole et al., 2014). The explanation given to the lack of effect in Gathercole and colleague's study was because Welsh and English are often used interchangeably within bilingual communities amongst bilingual speakers. This has led to the proposal that both Welsh and English are 'on-line' in these bilingual speakers. This means that the constant monitoring, inhibiting and differentiating between the languages is not required to the same extent as for other bilinguals in different contexts. Although Spencer and Hanley (2003) reported enhanced phoneme awareness amongst Welsh learners in their study, they reported that these findings were due to the orthographic transparency of Welsh as opposed to bilingualism. The children in Spencer and Hanley's study who were learning Welsh only had limited exposure to English as the children considered as 'Welsh speakers' came from Welsh-speaking homes and attended Welsh-medium schools. Consequently, the results from this thesis support the proposal that the unique linguistic setting in Wales means that positive effects of bilingualism on PA and other cognitive functions are only observed in bilinguals who are in circumstances where they are required to separate and differentiate between their languages frequently.

8.4 Limitations

This research is the first empirical group study to investigate bilingual language development in children with DS to be conducted in the UK. The research took a novel approach in relation to the linguistic context that the research was situated (i.e. Wales with Welsh-English bilinguals) and as a result, there are many potential challenges and factors that needed to be considered. Whilst every effort was made to identify and eliminate any possible confounding variables before and during the research, some factors will undoubtedly need to be considered when interpreting the results and drawing conclusions. The first and most obvious limitation to this research is the relatively small number of participants recruited for the research (with a total sample size of 77 and 32 of which being children with DS). Although these participant numbers are fairly small, this is a reflection of the fact that populations of bilingual children with DS are small and the research also specified that they had to be Welsh-English bilinguals, making the target sample even smaller. Furthermore, the criteria were

fairly strict in order to obtain a robust match between groups which further explains the relatively small sample.

The population of bilinguals with DS in Wales may be small as a result of parents being advised to limit language input to English only (given that English is the majority community language in Wales), meaning that the lack of information on this topic may have reduced the number of bilingual children with DS in Wales as a whole. It is worth noting that although the sample size is relatively small, this is the largest study of bilingual children with DS to date with the largest sample size of bilinguals with DS previously being eight children (Kay-Raining Bird et al., 2005). Prior research of children with DS has also employed groups of bilingual children who do not all share the same two languages, meaning that these findings are more difficult to generalise. Further studies investigating bilingualism in other DDs also generally have small sample sizes due to the nature of the target population and the range of languages that often exist within societies, making studies which specify the combination of languages having smaller samples (Feltmate & Kay-Raining Bird, 2008b; Hambly & Fombonne, 2009; Cleave et al., 2014). Future studies employing larger sample sizes will be advantageous to substantiate findings from the current research and that of previous research.

A further consideration of this research is that because of the small number of Welsh-English bilingual children with DS, a relatively large age range of children were required to obtain an adequate sample size (with children with DS being between 5-16 in the full sample). As noted above, this is a reflection of the fact that the population of bilinguals with DS is small and consequently the inclusion of this age range was required in order to obtain a sample size large enough to undertake statistical analyses. Although this range is large compared to research with TD children, studies with children who have a DD often have larger age ranges (although developmental ages did not have a large range). The use of statistical analyses was considered the most appropriate way to answer the main research questions in this thesis (i.e. to identify if there any meaningful differences between bilinguals and monolinguals with DS). As such, group analyses were required which meant that the age range of children included had to be relatively larger. Although this meant that individual differences cannot be identified as clearly, this approach to including children with a range of abilities led to a relatively representative sample of children with DS. Furthermore, the groups were matched for chronological age and developmental age meaning that although the groups did include children between 5-16 (with the individually matched sample between 5-14), as a whole, the groups were fairly homogeneous and both had very similar mean ages (with an average for the bilinguals with DS being 9;6 and the monolinguals with DS being 9;4 on average).

In addition, bilingualism is a multifaceted heterogeneous concept which is often dynamic and unique to each individual. This may further complicate the issue of using group studies as large individual differences may be masked by employing group analyses. Although it is important to identify that the samples included a range of Welsh-English bilingual children (with the majority being Welsh dominant or balanced bilinguals) with varying Welsh exposure, this sample subsequently reflected the nature of bilingualism in Wales. As Welsh is

a minority language and considered as 'vulnerable' according to the UNESCO criteria (see Chapter 1; Moseley, 2012), this means that exposure to Welsh is often dependent on language transmission through family and/or the school context. Although the consistency of exposure is dependent on several factors, specific criteria were in place to ensure the bilingual and monolingual status of the children. The criteria included specifying that the children had to be exposed to both Welsh and English before age three (in line with previous studies of simultaneous bilinguals; Paradis et al., 2011) and the children in the individual sample were exposed to both languages before the age of two. In addition, information was collected concerning the context of language exposure, the amount of current and lifetime exposure to each language, the language of response and also any inconsistencies in exposure. This resulted in participants who were clearly defined as bilingual or monolingual with no uncertainty of the children being bilingual or monolingual.

A further potential limitation of this research is the issue of available assessments in Welsh and English, and their suitability for use with children with DS. There is currently a dearth of language assessments that are standardised for use with Welsh-English bilingual children, with the *Prawf Geirfa Cymraeg: Fersiwn 7-11* (The Welsh Vocabulary Test: Version 7-11; Gathercole & Thomas, 2007) being the only exception. In addition, many of the standardised English assessments and the *Prawf Geirfa* had to be used with raw scores converted to standardised z-scores as opposed to using the standard scores provided within each respective manual. This was because the children with DS were outside of the age range for these assessments. The lack of equivalent assessments in Welsh and English also meant that specially designed tasks needed to be designed in order to assess PA within this population specifically. Furthermore, although there are numerous standardised English language assessments, these are usually designed for English monolinguals and consequently, the norms may not be appropriate to use with bilingual children, particularly if English is being acquired as a second language.

This is an issue for researchers and clinicians working with bilingual children and adults across all languages, as assessments that are designed for monolinguals may overestimate language impairments or may not be sensitive enough to detect language-specific difficulties. Although this is an issue, the fact that the bilinguals performed equally as well as the monolinguals for expressive and receptive language (Chapter 4) and PA (Chapter 5), if anything, strengthens the findings and conclusions. The majority of English language assessments used are designed for English monolinguals, and as a result, the finding that the bilingual children displayed similar English abilities substantiates the claim that the bilingual children are not disadvantaged by the addition of a second language. In the future, assessments that are designed or have norms available for bilingual children should be developed and employed wherever possible in order to overcome this issue.

Finally, some additional factors may have impacted language outcomes in the populations under study. For example, the hearing status of the children over their lifetime may have impacted their language abilities. Although efforts were taken to ensure that the

children had normal (or corrected to normal hearing) with no more than mild hearing loss being part of the inclusion criteria, actual measures of hearing status were not obtained during this study. Additionally, many of the children with DS wore hearing aids and so many may have had limited hearing at early stages in development. This factor is often an issue for research that assesses language in children with DS, however, in order to ensure that this does not impact on results, actual hearing status assessments may assist in ensuring that current hearing is at an appropriate level in future studies. The use of Makaton is also frequently used within this population, and although the majority of parents/guardians did state whether or not their child used or was still using Makaton, it was not possible to obtain this information for all children. Consequently, the use of Makaton may have impacted the language development of the children under study in some way. Future research should endeavour to explore this factor in bilingualism research further.

8.5 Future Research Directions

The research undertaken in this thesis has provided an in-depth exploration of the language profiles of Welsh-English bilingual children with DS and provided a valuable starting point for further research in this field. As this is the first piece of research that employs an empirical group study of bilingualism in this population and the first in the UK, many further aspects warrant further investigations. Firstly, longitudinal studies that monitor the progress of language development in bilingual children with DDs, and particularly DS will provide very valuable information as to the trajectory of language development in these bilingual children. This would also allow for a clearer understanding as to the role of individual factors on language outcomes in this population specifically. For example, exploring the role of input and the amount of exposure that is required in each language to result in language growth in both languages. Longitudinal studies are particularly useful in bilingualism research given the diversity and multifaceted nature of bilingualism, the variability which has been documented in the language development of children with DS (Buckley, 2002) and the slower rate of development observed in those with DS (Chapman & Hesketh, 2000). Alongside this, investigations that compare children learning two languages from birth and those who receive bilingual exposure later in development are required to identify if the age of exposure has an impact on language outcomes, as it appears to in TD children.

A further area of research that is needed to further inform language development in bilingual children with DS is to investigate the utility and outcomes of interventions. Intervention studies have been used previously to improve language abilities in monolinguals with DS (Kelly Burgoyne et al., 2012; Davis, 2008; Yoder et al., 2014), bilinguals with SLI/DLD (Ebert et al., 2014) and in a case study of a bilingual child with ASD (Seung, Siddiqi & Elder, 2006). To date, no information has been reported as to the success of similar interventions in bilinguals with DS. Additionally, preliminary reports suggest that cross-language transfer also assists in enhancing language abilities in a study that reported on intervention outcomes for bilingual children with SLI (Ebert et al., 2014). Interventions may be particularly useful for

aspects such as PA, given that this area has been highlighted as a specific area of weakness for these children (Cupples & Iacono, 2000; Snowling, Hulme & Mercer, 2002) and the fact that studies have shown that interventions successfully improve phoneme awareness and letter knowledge in parent-led interventions for monolinguals with DS (Bysterveldt, Gillon & Moran, 2017). As the process of developing PA in bilinguals is believed to be different from monolinguals (i.e. with the possibility of language transfer and interaction between languages), interventions may even be more successful for bilinguals with DS. Further research which directly employs tailored interventions is warranted, including interventions delivered in either the L1, L2, or both languages.

Finally, many further elements require further research in order to understand more about bilingualism in those with DS. For example, all research to date concerns children with DS, however, language impairments often persist in adults with DS. Research that explores bilingualism in adults with DS is warranted. Furthermore, an earlier onset of cognitive decline has been reported in adults with DS (Das et al., 1995; Grieco et al., 2015) and this could also impact on language abilities in adults, for both monolinguals and bilinguals. This may also relate to research that proposes that bilingualism may act as a protective mechanism to delay the onset of Alzheimer's in adults without DS (Bialystok, Craik & Freedman, 2010; Schweizer et al., 2012). Given the earlier onset of Alzheimer's in those with DS, exploring whether any protective mechanism is observed by bilingualism in this population through future research could uncover some important clinical findings. This possibility should be explored in future research.

8.6 General Conclusion

This research aimed to investigate bilingualism in children with DS in order to provide evidence to support or dispute the proposition that the addition of a second language is detrimental to the linguistic outcomes of these children. Prior research suggests that parents or guardians may receive recommendations to avoid bilingual exposure or may themselves believe that two languages would be too challenging, with the logic being if one language is difficult, two languages might be too difficult in this population. The research presented in this thesis conflicts with this suggestion and finds that bilingualism is not an aggravating factor to language outcomes. The findings from this study show that there is no evidence to suggest that bilingual children with DS show any further language delays or impairments in comparison to carefully matched control groups. Commensurate levels of English language abilities were found for bilinguals with DS compared to English monolingual children with DS. Additionally, the bilingual children with DS displayed similar Welsh language abilities to developmentally matched TD Welsh-English bilingual children. As over half of the word is bi/multilingual with over half of the children in the word being raised with two or more languages (Marian & Shook, 2012), there will be numerous families who may have concerns or questions surrounding the suitability of bilingualism if they have a child with a DD. This thesis has a specific focus on Wales given the opportunities that exist for children to become

bilingual and the growing number of Welsh speakers. Moreover, as policies are in place to encourage bilingualism in this context, the linguistic setting is very suitable for bilingualism research. This thesis has consequently provided important information as to the suitability of Welsh-medium provisions for children with DS.

Despite a range of individual variation in monolingual and bilingual children with DS (including in children with a dual ASD and DS diagnosis), this thesis provides evidence to support and substantiate previous literature that children with DS are not negatively impacted by bilingualism for a range of linguistic aspects. These include core, receptive and expressive language as well as aspects of PA in English and Welsh. As stated by Kathryn Kohnert, “monolingualism is not a cure for a language disability” (Kohnert, 2007, p.7) and this summarises the viewpoint of this research based on the finding that bilingualism is not an exacerbating factor for children with language impairments, and more specifically, DS. It is likely that children with language impairments will simply display these impairments in both languages as documented in this thesis, but these impairments will not be greater. Given the findings of this thesis and prior research, and the fact that bilingualism is a natural and necessary feature for many families, future efforts should now be focussed on how best to support and foster bilingualism in these populations.

The findings of this thesis, when taken alongside prior research conducted by Kay-Raining Bird and colleagues (2005), highlights that there are no differences in the fundamental language abilities (i.e. receptive and expressive language) and on PA for children with DS who have received substantial input in a second language compared to monolinguals. This thesis had provided substantial new information as to the language profiles of bilingual children with DS and adds to the growing body of literature that, on the whole, supports bilingualism in children with DDs. Further research in this field is required, particularly concerning the possibility of language transfer, intervention and longitudinal studies as well as any impact of bilingualism on cognitive decline in this population. The findings of this thesis have several crucial implications on clinical practice, professional recommendations, educational provisions and policies in Wales, which have been highlighted in this thesis.

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Appendices List

Item 1: Information sheets (Welsh and English)

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Item 1: Information Sheets



Bangor University Project on Language Development: Information Sheet

This project will examine the overall language development of children who have a range of language backgrounds including children with and without Down syndrome in Wales. Language development will be measured in terms of language comprehension, production and also on a range of tasks that are designed to measure children's awareness of phonological awareness (speech sounds) such as rhyming and alliteration tasks. These skills have been closely linked to reading and spelling abilities. This project will specifically be looking at any differences between Welsh-English bilinguals, and children who only speak one language. There is very limited research to date which explores bilingualism in these populations, and therefore this research aims to increase our knowledge in this field.

The questions that we hope to answer with the research are:

- What are the language abilities of children with Down syndrome compared to typically developing children?
- Are there areas of language development that are more challenging for these individuals?
- How does the language development compare for children who are bilingual compared to those who speak one?

Parents or guardians will be asked to complete a background questionnaire. Children will be tested on a range of standardised and non-standardised tests that will identify their strengths and weaknesses in their language development. Tasks designed to measure children's awareness of speech sounds will also be assessed, such as rhyming and alliteration tasks.

Testing will be conducted during a one-to-one session in a relaxed environment, and your child will be free to stop the tasks at any point if they would like to. The questionnaires and all data collected will be held completely confidentially and anonymously, and any publication of the research will not identify any of the participants. If you are happy for your child to take part, please tick the 'yes' box overleaf and return to the researcher. You are free to withdraw from the research at any time by contacting the researchers on the contact details below.

We hope that this research will provide a new insight into bilingual and monolingual language development in children who are typically developing, and children with Down syndrome. It is hoped that this research will assist families in making their own language choices, as well as implications for clinical practice and education policy in Wales.

This project is funded by the Economic and Social Research Council. If you would like more information about the project, please contact the researchers below:

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**Prosiect Prifysgol Bangor ar Ddatblygiad Iaith:
Taflen Wybodaeth**

Bydd y prosiect yma yn edrych ar ddatblygiad iaith gyffredinol plant sydd ag ystod o gefndiroedd ieithyddol yng Nghymru, gan gynnwys plant sydd â Syndrom Down ac sydd heb. Bydd mesuriadau o ddatblygiad ieithyddol yn cael ei fesur yn nhermau dealltwriaeth, chynhyrchu iaith a hefyd ar ystod o dasgau sydd wedi'u cynllunio i fesur ymwybyddiaeth plant o synau lleferydd, fel odli a thasgau cyflythrennu. Mae'r sgiliau yma wedi cael eu cysylltu'n agos i alluoedd darllen a sillafu cyffredinol. Bydd y prosiect hwn yn edrych yn benodol ar unrhyw wahaniaethau rhwng plant ddwyieithog, a phlant sydd ond yn siarad un iaith. Mae ymchwil yn y maes yma yn gyfyngedig iawn, gan fod dim llawer o ymchwil sy'n archwilio dwyieithrwydd yn y poblogaethau hyn eto.

Dyma rai cwestiynau yr ydym yn gobeithio ei ateb gyda'r ymchwil:

- Beth yw gallu ieithyddol plant â Syndrom Down sy'n ddwyieithog neu sy'n siarad un iaith?
- A oes patrymau o ddatblygiad o fewn y plant hyn, ble mae rhai meysydd yn anoddach i ddatblygu nag eraill?
- Sut mae'r datblygiad iaith yn cymharu ar gyfer plant dwyieithog â'r rhai sy'n siarad un iaith?

Gofynnir i rieni neu gorchuddir i lenwi holiadur cyffredinol. Bydd plant yn cael eu profi ar ystod o brofion safonol ac ansafonol a fydd yn arddangos eu cryfderau a gwendidau yn eu datblygiad ieithyddol. Bydd tasgau ychwanegol yn cael eu cynllunio er mwyn targedu eu hymwybyddiaeth o synau lleferydd, er enghraifft tasgau odli a cyflythrennu.

Bydd y profion yn cael eu cynnal yn ystod sesiwn un-i-un mewn awyrgylch distaw, a bydd eich plentyn yn rhydd i stopio'r tasgau ar unrhyw adeg os hoffent. Bydd yr holiadur a'r holl ddata a gasglwyd yn cael eu storio yn hollol gyfrinachol ac yn ddiennw. Os bydd unrhyw gyhoeddiad o'r ymchwil, ni fydd hwn yn datgelu unrhyw wybodaeth bersonol o'r plant neu'r teuluoedd. Os ydych yn fodlon i'ch plentyn gymryd rhan, ticiwch y 'ie' blwch dros y tudalen a'i ddychwelyd i'r ysgol/meithrinfa. Rydych yn rhydd i dynnu'ch caniatâd o'r ymchwil ar unrhyw adeg drwy gysylltu â'r ymchwilwyr ar y manylion cyswllt isod.

Rydym yn gobeithio y bydd yr ymchwil yn rhoi golwg newydd i mewn i'r datblygiad iaith plant sydd â Syndrom Down sy'n ddwyieithog neu sy'n siarad un iaith. Bydd hefyd yn gobeithio helpu teuluoedd i wneud dewisiadau iaith eu hunain, yn ogystal â goblygiadau i ymarfer clinigol a pholisïau addysg yng Nghymru.

Mae'r prosiect yma wedi cael ei hariannu gan y Cyngor Ymchwil Economaidd a Chymdeithasol. Os hoffech fwy o wybodaeth am y prosiect, cysylltwch â'r ymchwilwyr isod:

Rebecca Ward
Myfyrwraig PhD
Ysgol Ieithyddiaeth ac Iaith Saesneg
Prifysgol Bangor
LL57 2DG.
Rebecca.Ward@bangor.ac.uk

Dr Eirini Sanoudaki
Uwch Ddarlithydd mewn Caffael Iaith
Ysgol Ieithyddiaeth ac Iaith Saesneg
Prifysgol Bangor
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e.sanoudaki@bangor.ac.uk

Item 2: Ethical Approval

College of Arts & Humanities

APPLICATION FOR APPROVAL BY RESEARCH ETHICS COMMITTEE STUDENT RESEARCH PROJECT – DECISION



Reference Number: CAH 75

Name of researcher:	Rebecca Ward
Name of supervisor:	Dr Thora Tenbrink
Research project title:	Bilingual language development in Down syndrome
Submission date:	10.05.17
Decision date:	27.07.17

This proposal was approved by College of Arts and Humanities Ethics Committee, Bangor University.

Signed (on behalf of the committee) by:

Date: 27.07.17
Dr Peter Shapely, Chair of the Ethics Committee

Item 3: Parent/Guardian Consent Form



**Bangor University Project on Language Development
Consent Form**

- I have been briefed to my satisfaction on the research for which I have volunteered, and understand what is required.
- I understand that I have the right to withdraw my child's participation from the research at any point.
- I also understand that my rights to anonymity and confidentiality will be respected.

Would you like your child to take part in this research?

Yes

☐

No

☐

Child's Name:

School:

Parent / Guardian signature

Date: __ / __ / __

Rebecca Ward
PhD Student
School of Linguistics and English Language
Bangor University
LL57 2DG.
Rebecca.Ward@bangor.ac.uk

Dr Eirini Sanoudaki
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School of Linguistics and English Language
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Prosiect Prifysgol Bangor ar Ddatblygiad Iaith
Taflen Caniatâd

- Rwyf wedi derbyn gwybodaeth i fy moddhad ar yr ymchwil yma, a dwi'n deall yr hyn sy'n ofynnol.
- Rwy'n deall bod gennyf yr hawl i dynnu cyfranogiad fy plentyn ar unrhyw adeg.
- Rwyf hefyd yn deall y bydd fy hawl i aros yn ddienw a chyfrinachedd yn cael ei barchu.

A fydddech yn hapus i'ch plentyn gymryd rhan yn yr ymchwil?

Ie

☐

Na

☐

Enw'r Plentyn

Ysgol/Meithrinfa:

Llofnod Rhiant/Gwarchodwyr

Dyddiad: __ / __ / __

Rebecca Ward
Myfyrwraig PhD
Ysgol Ieithyddiaeth ac Iaith Saesneg
Prifysgol Bangor
LL57 2DG.
Rebecca.Ward@bangor.ac.uk

Dr Eirini Sanoudaki
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Item 4: Letter to Parents



Dear Parent or Guardian,

The purpose of this letter is to provide you with information about a research project that is currently underway at Bangor University. We are investigating language development in children who have a range of language backgrounds in Wales in children with and without developmental disabilities. The aim of the project is to increase our understanding of the patterns of development that exist for children learning to speak one or two languages. We hope that this project will contribute to our knowledge of this field and we hope that this research will assist families in making language choices, as well as implications for clinical practice and education policy in Wales.

We are inviting you and your child to take part in this study. If you agree to take part, your child will be assessed on a range of measures of language and general development in a one-to-one environment. The tasks are designed to be game-like and your child will be free to stop taking part at any point if they would like. Confidentiality will be respected throughout the project and any publication of the research will not identify any of the families who have taken part.

We would be very grateful if you could return the consent form and questionnaire to the school as soon as possible. Please keep the information sheet for your records.

If you are happy for your child to take part, please tick the 'yes' box overleaf, fill in the background questionnaire and return to the school. You are free to withdraw from the research at any time by contacting the researchers on the contact details below. If you would like more information about the project or have any questions, please contact the researchers below.

Your sincerely,

Rebecca Ward
PhD Student
School of Linguistics and English Language
Bangor University
LL57 2DG.
rebecca.ward@bangor.ac.uk

Dr Eirini Sanoudaki
Senior Lecturer in Language Acquisition
School of Linguistics and English Language
Bangor University
LL57 2DG.
e.sanoudaki@bangor.ac.uk



Annwyl Rhiant neu Warchodwyr,

Pwrpas y llythyr yma yw rhannu wybodaeth ynglŷn â phrosiect ymchwil sy'n cael ei chynnal ar hyn o bryd ym Mhrifysgol Bangor. Rydym yn astudio datblygiad iaith mewn plant sydd â chefnidiroedd amrywiol o fewn Cymru mewn plant sydd gyda Syndrom Down ac sydd heb. Nod yr ymchwil yw cynyddu ein dealltwriaeth ynglŷn â phatrymau o ddyblygiad sy'n bodoli o fewn plant sydd naill ai yn siarad un neu dwy iaith. Rydym yn gobeithio y bydd yr ymchwil yma yn cyfrannu at ein hadnabyddiaeth o'r maes yma er mwyn helpu teuluoedd gwneud dewisiadau iaith wybodus gan hefyd helpu gyda pholisïau addysg ac ymarfer clinigol yn Gymru.

Rydym yn gwahodd i chi a'ch plentyn i gymryd rhan yn yr ymchwil yma. Os ydych yn hapus i gymryd rhan, bydd eich plentyn yn mynychu sesiynau er mwyn asesu datblygiad cyffredinol a datblygiad iaith yn benodol. Mae'r sesiadau yma wedi cael ei gynllunio i fod fel gemau a bydd hawl gan eich plentyn i stopio'r tasgau os mynnent. Byddwn yn parchu cyfrinachedd trwy gydol i brosiect, ac ni fydd unrhyw cyhoeddi o'r ymchwil yn datgelu unrhyw wybodaeth am y teuluoedd sydd wedi cymryd rhan.

Byddwn yn ddiolchgar iawn os medrwch chi lenwi'r holiadur byr a'r ffurflen caniatâd a ddychwelyd y rhain i'r ysgol cyn gynted ag y medrwch. Plis cadwch y ffurflen wybodaeth am eich cofnodion.

Os ydych yn fodlon i'ch plentyn gymryd rhan, ticiwch y 'ie' blwch dros y tudalen a'i ddychwelyd i'r ysgol gyda'r holiadur. Rydych yn rhydd i dynnu'ch caniatâd o'r ymchwil ar unrhyw adeg drwy gysylltu â'r ymchwilwyr ar y manylion cyswllt isod. Os hoffech fwy o wybodaeth am y prosiect neu os oes gennych chi unrhyw gwestiynau, cysylltwch â'r ymchwilwyr isod:

Yn gywir,

Rebecca Ward
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School of Linguistics and English Language
Bangor University
LL57 2DG.
rebecca.ward@bangor.ac.uk

Dr Eirini Sanoudaki
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School of Linguistics and English Language
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Item 5: Questionnaire for parents of children with DS



PRIFYSGOL
BANGOR
UNIVERSITY

Participant ID: _____

**Bangor University Project on Language Development:
Background Questionnaire**

Information about your child			
Date of birth: __/__/__			
<input type="checkbox"/> Male <input type="checkbox"/> Female			
School: _____			
Does your child have any other diagnosis other than Down syndrome?			
<input type="checkbox"/> No <input type="checkbox"/> Yes <input type="checkbox"/> Suspected but not diagnosed			
If yes, please state _____			
Does your child have a hearing disorder? <input type="checkbox"/> Yes <input type="checkbox"/> No			
If yes, please describe (e.g. cause, severity), and state whether this has been corrected:			

Language Background			
Home Language(s): _____			
Please list what percentage of the time your child is currently exposed to each language:			
Language:			
Percentage: (<i>this should total 100%</i>)			
Please list what percentage of the time your child has been exposed to each language over their lifetime:			
Language:			
Percentage: (<i>this should total 100%</i>)			
If your child speaks a second or third language, what percentage of time does your child chose to respond in each language?			
Language:			
Percentage: (<i>this should total 100%</i>)			
Rate your child's overall expressive speech (production) where: 1= Poor, 5 = Excellent			
<input type="checkbox"/> 1 <input type="checkbox"/> 2 <input type="checkbox"/> 3 <input type="checkbox"/> 4 <input type="checkbox"/> 5			
Rate your child's overall receptive speech (comprehension) where: 1 = Poor, 5 = Excellent			
<input type="checkbox"/> 1 <input type="checkbox"/> 2 <input type="checkbox"/> 3 <input type="checkbox"/> 4 <input type="checkbox"/> 5			

What was the age of your child's first word? _____

Did your child attend nursery? ☐ Yes ☐ No

If so, what was the main language of the nursery? _____

Does your child have a language disorder?

☐ None ☐ Diagnosed ☐ Suspected

If yes, please state _____

If your child speaks a second or third language, approximately what age were they first exposed to each language? _____

Please indicate the consistency and duration of exposure to each language (e.g. constant, any gaps in exposure?) _____

Have you ever received any recommendations regarding multiple language exposure? _____

Information about you

Highest level of parental/guardian education:

- ☐ GCSE's
- ☐ Apprenticeship
- ☐ A Level
- ☐ Bachelor's Degree
- ☐ Masters Level Degree
- ☐ Doctoral Degree
- ☐ Other _____

Highest parental/guardian occupation:

- ☐ Higher managerial and professional occupations
- ☐ Lower managerial and professional occupations
- ☐ Intermediate occupations (clerical, sales, service)
- ☐ Small employers and own account workers
- ☐ Lower supervisory and technical occupations
- ☐ Semi-routine occupations
- ☐ Routine occupations
- ☐ Never worked or long-term unemployed
- ☐ Other _____

If you are happy to be contacted further about this research and its outcomes, please provide your contact details below: _____

Participant ID:



PRIFYSGOL
BANGOR
UNIVERSITY

**Prosiect Prifysgol Bangor ar Ddatblygiad Iaith:
Holiadur Cyffredinol**

Gwybodaeth am eich plentyn

Dyddiad Geni: __/__/__

☐ Bachgen ☐ Merch

Ysgol: _____

A oes gan eich plentyn unrhyw ddiagnosis arall heblaw Syndrom Down?

☐ Nac oes ☐ Oes ☐ Efallai – ond ddim wedi'i ddiagnosio

Os oes, nodwch pa fath: _____

A oes gan eich plentyn anhwylder clyw? ☐ Oes ☐ Nac oes

Os oes, disgrifiwch (e.e. achos, difrifoldeb), a nodwch os yw wedi ei gywiro neu beidio:

Cefndir iaith

Iaith/leithoedd Gartref: _____

Nodwch pa ganran o'r amser mae eich plentyn yn clywed bob iaith ar hyn o bryd:

Iaith:			
Canran: (dylai hyn adio i 100%)			

Nodwch pa ganran o'r amser mae eich plentyn wedi clywed bob iaith ar draws ei bywyd:

Iaith:			
Canran: (dylai hyn adio i 100%)			

Os yw eich plentyn yn siarad ail neu drydedd iaith, pa ganran o'r amser ydyn nhw'n dewis ymateb ym mhob iaith?

Iaith:			
Canran: (dylai hyn adio i 100%)			

Nodwch sgiliau eich plentyn yn nhermau lleferydd cynhyrchu (1 = Gwan, 5 = Rhagorol)

☐ 1 ☐ 2 ☐ 3 ☐ 4 ☐ 5

Nodwch sgiliau eich plentyn yn nhermau lleferydd dealltwriaeth (1 = Gwan, 5 = Rhagorol)

☐ 1 ☐ 2 ☐ 3 ☐ 4 ☐ 5

Beth oedd oedran eich plentyn ar adeg ei air cyntaf? _____

A wnaeth eich plentyn mynd i feithrinfa? ☐ Do ☐ Naddo

Os le, nodwch ym mha iaith _____

Oes gan eich plentyn unrhyw anhwylder iaith?

☐ Dim

☐ Wedi'u diagnosio

☐ Efallai

Os oes, nodwch pa fath: _____

Os yw eich plentyn yn siarad ail neu drydedd iaith, tua pha oedran dechreuon nhw glywed bob iaith? _____

Nodwch gysondeb a hyd o amlygiad i bob iaith (e.e. cyson iawn, bob dydd, unrhyw fylchau mewn clywed bob iaith?) _____

Ydych chi erioed wedi derbyn unrhyw argymhellion ynghylch amlygiad dwyieithog? _____

Gwybodaeth amdanoch chi

Lefel uchaf o addysg rhieni/gwarchodwyr:

- ☐ TGAU
- ☐ Prentisiaeth
- ☐ Lefel A
- ☐ Gradd Israddedig
- ☐ Gradd Lefel Meistr
- ☐ Gradd Doethuriaeth
- ☐ Arall _____

Galwedigaeth uchaf o rieni/gwarchodwyr:

- ☐ Galwedigaeth rheoli a phroffesiynol uwch
- ☐ Galwedigaeth rheoli a phroffesiynol is
- ☐ Swyddi canolradd (e.e. clerigol, gwasanaeth, gwerthu)
- ☐ Cyflogwyr bach a gweithwyr unigol
- ☐ Goruchwylio is a galwedigaethau technegol
- ☐ Swyddi lled-reolaidd
- ☐ Swyddi rheolaidd
- ☐ Erioed wedi gweithio neu ddi-waith yn hir dymor
- ☐ Arall _____

Os ydych yn hapus i ni gysylltu gyda chi ymhellach ar gyfer yr ymchwil a'i ganlyniadau, rhowch eich manylion cyswllt isod: _____

Item 6: Questionnaire for parents of TD children

Participant ID:



**Bangor University Project on Language Development:
Background Questionnaire**

Information about your child

Date of birth: __/__/__

☐ Male ☐ Female

School: _____

Disability: ☐ None ☐ Yes ☐ Suspected but not diagnosed

If yes, please state: _____

Does your child have any hearing disorder? ☐ Yes ☐ No

If yes, please describe (e.g. cause, severity), and state whether this has been corrected:

Language Background

Home Language(s): _____

Please list what percentage of the time your child is currently exposed to each language:

Language:			
Percentage: (<i>this should total 100%</i>)			

Please list what percentage of the time your child has been exposed to each language over their lifetime:

Language:			
Percentage: (<i>this should total 100%</i>)			

If your child speaks a second or third language, what percentage of time does your child chose to respond in each language?

Language:			
Percentage: (<i>this should total 100%</i>)			

Rate your child's overall expressive speech (production) where 1= Poor, 5 = Excellent

☐ 1 ☐ 2 ☐ 3 ☐ 4 ☐ 5

Rate your child's overall receptive speech (comprehension) where 1 = Poor, 5 = Excellent

☐ 1 ☐ 2 ☐ 3 ☐ 4 ☐ 5

What was the age of your child's first word? _____

Did your child attend nursery? ☐ Yes ☐ No

If so, what was the main language of the nursery? _____

Does your child have a language disorder?

☐ None ☐ Diagnosed ☐ Suspected

If yes, please state _____

If your child speaks a second or third language, approximately what age were they first exposed to each language? _____

Please indicate the consistency and duration of exposure to each language (e.g. constant, any gaps in exposure?) _____

Have you ever received any recommendations regarding multiple language exposure? _____

Information about you

Highest level of parental/guardian education:

- ☐ GCSE's
- ☐ Apprenticeship
- ☐ A Level
- ☐ Bachelor's Degree
- ☐ Masters Level Degree
- ☐ Doctoral Degree
- ☐ Other _____

Highest parental/guardian occupation:

- ☐ Higher managerial and professional occupations
- ☐ Lower managerial and professional occupations
- ☐ Intermediate occupations (clerical, sales, service)
- ☐ Small employers and own account workers
- ☐ Lower supervisory and technical occupations
- ☐ Semi-routine occupations
- ☐ Routine occupations
- ☐ Never worked or long-term unemployed
- ☐ Other _____

If you are happy to be contacted further about this research and its outcomes, please provide your contact details below: _____

Participant ID:



Prosiect Prifysgol Bangor ar Ddatblygiad Iaith:
Holiadur Cyffredinol

Gwybodaeth am eich plentyn

Dyddiad Geni: __/__/__

☐ Bachgen ☐ Merch

Ysgol: _____

Anabledd: ☐ Dim ☐ Oes ☐ Efallai –ond ddim wedi’u ddiagnosio

Os oes, nodwch pa fath: _____

A oes gan eich plentyn anhwylder clyw? ☐ Oes ☐ Nac oes

Os oes, disgrifiwch (e.e. achos, difrifoldeb), a nodwch os yw wedi ei gywiro neu beidio:

Cefndir iaith

Iaith/leithoedd Gartref: _____

Nodwch pa ganran o’r amser mae eich plentyn yn clywed bob iaith ar hyn o bryd:

Iaith:			
Canran: (dylai hyn adio i 100%)			

Nodwch pa ganran o’r amser mae eich plentyn wedi clywed bob iaith ar draws ei bywyd:

Iaith:			
Canran: (dylai hyn adio i 100%)			

Os yw eich plentyn yn siarad ail neu drydedd iaith, pa ganran o’r amser ydyn nhw’n dewis ymateb ym mhob iaith?

Iaith:			
Canran: (dylai hyn adio i 100%)			

Nodwch sgiliau eich plentyn yn nhermau lleferydd cynhyrchu (1 = Gwan, 5 = Rhagorol)

☐ 1 ☐ 2 ☐ 3 ☐ 4 ☐ 5

Nodwch sgiliau eich plentyn yn nhermau lleferydd dealltwriaeth (1 = Gwan, 5 = Rhagorol)

☐ 1 ☐ 2 ☐ 3 ☐ 4 ☐ 5

eth oedd oedran eich plentyn ar adeg ei air cyntaf? _____

A wnaeth eich plentyn mynd i feithrinfa? ☐ Do ☐ Naddo

Os ie, nodwch ym mha iaith _____

Oes gan eich plentyn unrhyw anhwylder iaith?

☐ Dim ☐ Wedi'u diagnosio ☐ Efallai

Os oes, nodwch pa fath: _____

Os yw eich plentyn yn siarad ail neu drydedd iaith, tua pha oedran dechreuon nhw glywed bob iaith? _____

Nodwch gysondeb a hyd o amlygiad i bob iaith (e.e. cyson iawn, bob dydd, unrhyw fylchau mewn clywed bob iaith?) _____

Ydych chi erioed wedi derbyn unrhyw argymhellion ynghylch amlygiad dwyieithog? _____

Gwybodaeth amdanoch chi

Lefel uchaf o addysg rhieni/gwarchodwyr:

- ☐ TGAU
- ☐ Prentisiaeth
- ☐ Lefel A
- ☐ Gradd Israddedig
- ☐ Gradd Lefel Meistr
- ☐ Gradd Doethuriaeth
- ☐ Arall _____

Galwedigaeth uchaf o rieni/gwarchodwyr:

- ☐ Galwedigaeth rheoli a phroffesiynol uwch
- ☐ Galwedigaeth rheoli a phroffesiynol is
- ☐ Swyddi canolradd (e.e. clerigol, gwasanaeth, gwerthu)
- ☐ Cyflogwyr bach a gweithwyr unigol
- ☐ Goruchwylio is a galwedigaethau technegol
- ☐ Swyddi lled-reolaidd
- ☐ Swyddi rheolaidd
- ☐ Erioed wedi gweithio neu ddi-waith yn hir dymor
- ☐ Arall _____

Os ydych yn hapus i ni gysylltu gyda chi ymhellach ar gyfer yr ymchwil a'i ganlyniadau, rhowch eich manylion cyswllt isod: _____

Item 8 - Phonological Awareness Stimuli in English and Welsh (Alphabetical)

English Stimuli	English Transcription	Welsh Stimuli	Welsh Transcription
Alligator	/ˈælɪɡətə/	Afal	/aval/
Ant	/ænt/	Aligator	/alɪɡatɔr/
Apple	/ˈæpl/	Arth	/ɑrθ/
Arm	/ɑ:m/	Aur	/aɪr/
Bag	/bæg/	Bag	/bag/
Ball	/bɔ:l/	Bara	/bara/
Balloon	/bəˈlu:n/	Bran	/bra:n/
Bat	/bæt/	Brenhines	/brɛnhi:nɛs/
Bear	/beə/	Brenin	/brɛnɪn/
Bed	/bɛd/	Broga	/brɔga/
Bell	/bɛl/	Bryn	/brɪn/
Bike	/baɪk/	Buwch	/bɪuχ/
Bird	/bɜ:d/	Bwlb	/bɔlb/
Black	/blæk/	Bwyd	/bɔɪd/
Bone	/bəʊn/	Bys	/bi:s/
Book	/bʊk/	Cadair	/kadair/
Bowl	/bəʊl/	Canu	/kani:/
Brick	/brɪk/	Cap	/kap/
Bug	/bʌg/	Car	/kar/
Butterfly	/ˈbʌtəflaɪ/	Cath	/ka:θ/
Cap	/ˈkæp/	Caws	/kaʊs/
Car	/kɑ:/	Ci	/ki:/
Cat	/kæt/	Cloch	/klo:χ/

Caterpillar	/ˈkætəpɪlə/	Clwm	/klɒm/
Cot	/kɒt/	Coed	/kɔɪd/
Coat	/kəʊt/	Côr	/ko:r/
Coin	/kɔɪn/	Cot	/kɒt/
Cow	/kaʊ/	Cwch	/ku:χ/
Crab	/kræb/	Cŵn	/ku:n/
Crow	/krəʊ/	Cwningen	/kʊnɪŋən/
Day	/deɪ/	Cylch	/kɪlχ/
Dice	/daɪs/	Cysgu	/kəsɡi:/
Dinosaur	/ˈdaɪnəʊsɔ:/	Dafad	/davad/
Doctor	/ˈdɒktə/	Dail	/dail/
Dog	/dɒɡ/	Dawnsio	/daʊnʃɔ/
Doll	/dɒl/	Drwm	/drɒm/
Door	/dɔ:/	Drws	/dru:s/
Elephant	/ˈɛlɪfənt/	Dŵr	/du:r/
Eye	/aɪ/	Dwylo	/dʊɪlb/
Fan	/fæn/	Eira	/eɪrə/
Fish	/fɪʃ/	Fan	/væn/
Flag	/flæg/	Ffair	/fair/
Fly	/flaɪ/	Ffôn	/fo:n/
Fork	/fɔ:k/	Gafr	/ga:vr/
Foot	/fʊt/	Giât	/ɡiɑ:t/
Fox	/fɒks/	Glaw	/ɡlaʊ/
Frog	/frɒɡ/	Gwên	/gwe:n/
Gift	/ɡɪft/	Ham	/ham/
Goat	/ɡəʊt/	Haul	/hail/

Glue	/glu:/	Het	/hɛt/
Guitar	/gɪ'ta:/	Jam	/dʒam/
Hair	/heə/	Lindys	/lɪndɪs/
Hat	/hæt/	Llaw	/təʊ /
Hen	/hɛn/	Llew	/tɛu/
Hill	/hɪl/	Llong	/tɔŋ/
Jar	/dʒɑ:/	Llwch	/tu:χ/
Key	/ki:/	Llwy	/tɔi/
King	/kɪŋ/	Llwyd	/tɔɪd/
Kite	/kaɪt/	Llyfr	/təvr/
Lake	/leɪk/	Llygoden	/təgɔdɛn/
Lamb	/læm/	Llyn	/tɪn/
Lamp	/læmp/	Lori	/lɔri:/
Leaf	/li:f/	Malwen	/malwɛn/
Log	/lɒg/	Map	/map/
Map	/mæp/	Mathemateg	/məθematɛg/
Mat	/mæt/	Mefus	/mɛvɪs/
Microscope	/'maɪkrəskəʊp/	Meicrosgop	/mɛɪkrɔsgɔp/
Milk	/mɪlk/	Mêl	/me:l/
Moon	/mu:n/	Mop	/mɔp/
Mosquito	/məs'ki:təʊ/	Môr	/mo:r/
Mountain	/'maʊntɪn/	Mosgito	/mɔsgɪ:tɔ/
Mouse	/maʊs/	mwg	/mu:g/
Nose	/nəʊz/	Nant	/nant/
Pen	/pɛn/	Nos	/no:s/
Pet	/pɛt/	Oen	/ɔɪn/

Photograph	/ˈfəʊtəgræf/	Olew	/ɔləw/
Pig	/pɪg/	Pêl	/pe:l/
Pot	/pɒt/	Pen	/pen/
Rake	/reɪk/	Pont	/pɒnt/
Rat	/ræt/	Pot	/pɒt/
Rain	/reɪn/	Pren	/pren/
Red	/red/	Pysgodyn	/pəsgɒdɪn/
Restaurant	/ˈrɛstrɒnt/	Pysgota	/pəsgɒtə/
Ring	/rɪŋ/	Reis	/reis/
Rock	/rɒk/	Rhedeg	/redɛg/
Roof	/ru:f/	Robin-Goch	/rɒbɪn-go:χ/
Rope	/rəʊp/	Sach	/səχ/
Rose	/rəʊz/	Saer	/sair/
Rug	/rʌg/	Sebra	/sɛbra/
Sea	/si:/	Sêr	/se:r/
Shoe	/ʃu:/	Siop	/ʃɪɒp/
Ski	/ski:/	Tân	/tɑ:n/
Snake	/sneɪk/	Tarw	/tarɔ/
Snow	/snəʊ/	Tedi	/tɛdi:/
Soap	/səʊp/	Tlws	/tlu:s/
Sock	/sɒk/	Trên	/tre:n/
Star	/stɑ:/	Tri	/tri:/
Strawberry	/ˈstrɔ:bəri/	Wal	/wal/
Sun	/sʌn/		
Table	/ˈteɪbl/		
Tap	/tæp/		

Teacher	/ˈti:tʃə/
Ten	/tɛn/
Teeth	/ti:θ/
Tent	/tɛnt/
Toad	/təʊd/
Toes	/təʊz/
Tree	/tri:/
Umbrella	/ʌmˈbrɛlə/
Van	/væn/
Vegetables	/ˈvɛdʒtəb(ə)lz/
Vet	/vɛt/
Village	/ˈvɪlɪdʒ/
Volcano	/vɒlˈkeɪnəʊ/
Zebra	/ˈzɛbrə/

Item 9: Phonological Awareness Statistical Analyses

Tests of Normality

	Kolmogorov–Smirnov ^a			Shapiro–Wilk		
	Statistic	df	Sig.	Statistic	df	Sig.
Letters	.341	218	.000	.718	218	.000
Imagability	.220	218	.000	.751	218	.000
CEG	.305	218	.000	.508	218	.000
Syllables	.445	218	.000	.573	218	.000

a. Lilliefors Significance Correction

Hypothesis Test Summary

	Null Hypothesis	Test	Sig.	Decision
1	The distribution of Letters is the same across categories of Language.	Independent–Samples Mann–Whitney U Test	.410	Retain the null hypothesis.
2	The distribution of Imagability is the same across categories of Language.	Independent–Samples Mann–Whitney U Test	.103	Retain the null hypothesis.
3	The distribution of CEG is the same across categories of Language.	Independent–Samples Mann–Whitney U Test	.143	Retain the null hypothesis.
4	The distribution of Syllables is the same across categories of Language.	Independent–Samples Mann–Whitney U Test	.143	Retain the null hypothesis.

Asymptotic significances are displayed. The significance level is .05.

Item 10: Participant characteristics. *Note:* W = Welsh; E = English.

Ptp	Age	Dev. Age	Gender	Diagnosis	Home Language	Expressive	Receptive	Individually Matched Sample
DB1	8;2	<4;0	F	DS	W & E	3	4	Yes
DB2	9;6	<4;0	M	DS	W & E	2	4	Yes
DB3	13;9	<4;0	M	DS	W & E	2.5	4	Yes
DB4	12;3	4;8	M	DS	English	2	4	Yes
DB5	5;7	<4;0	M	DS	English	1	2	Yes
DB6	11;0	5;8	F	DS	W & E	4	4	Yes
DB7	10;11	5;2	F	DS	W & E	4	4	Yes
DB8	12;10	6;0	F	DS	W & E	3	4	Yes
DB9	5;5	<4;0	F	DS	W & E	1	1	Yes
DB10	5;9	<4;0	F	DS	W & E	3	4	Yes

Table 42. Participants in DSB Group.

Ptp	Age	Dev. Age	Gender	Diagnosis	Home Language	Expressive	Receptive	Individually Matched Sample
DM1	7;4	4;8	F	DS	English	3	3	Yes
DM2	11;10	6;3	F	DS	English	4	5	Yes
DM3	7;11	<4;0	F	DS	English	3	3	No
DM4	9;7	4;10	M	DS	English	3	4	Yes
DM5	7;8	4;4	M	DS	English	2	4	Yes
DM6	16;9	5;0	F	DS	English	4	4	No
DM7	6;0	<4;0	F	DS	English	3	5	Yes
DM8	12;6	<4;0	F	DS & ADHD	English	4	4	Yes
DM9	6;11	<4;0	F	DS	English	2	3	Yes
DM10	8;10	4;4	F	DS	English	3	3	No
DM11	7;7	4;0	M	DS	English	4	4	No
DM12	6;11	<4;0	F	DS	English	3	3	Yes
DM13	9;2	<4;0	M	DS	English	1	2	No
DM14	12;8	<4;0	F	DS	English	3	3	Yes
DM15	11;11	6;9	M	DS	English	4	5	Yes

Table 43. Participants in DSM Group

Ptp	Age	Dev. Age	Gender	Diagnosis	Home Language	Expressive	Receptive	Individually Matched Sample
TB1	5;5	5;0	M	None	English	4	5	Yes
TB2	4;6	5;6	M	None	-	-	-	No
TB3	4;6	5;8	F	None	Welsh	5	5	Yes
TB4	3;9	<4;0	M	None	-	-	-	No
TB5	3;3	4;0	M	None	-	-	-	No
TB6	3;3	<4;0	F	None	W & E	4	4	No
TB7	3;3	<4;0	F	None	W & E	3.5	5	No
TB8	3;9	4;4	F	None	W & E	5	5	No
TB9	3;0	4;4	F	None	English	5	5	No
TB10	7;10	8;3	F	None	W & E	4	4	No
TB11	3;8	<4;0	M	None	W & E	4	5	Yes
TB12	2;11	<4;0	F	None	English	5	5	No
TB13	3;3	<4;0	F	None	W & E	5	5	No
TB14	5;7	4;4	M	None	Welsh	4	5	No
TB15	4;6	<4;0	M	None	W & E	3	3	No
TB16	5;10	4;10	M	None	Welsh	3	3	No
TB17	3;9	<4;0	F	None	W & E	4	4	Yes
TB18	7;10	6;3	M	None	W & E	4	3	Yes
TB19	3;0	<4;0	M	None	W & E	4	4	Yes
TB20	3;8	4;10	M	None	W & E	4	5	No

TB21	3;2	<4;0	F	None	W & E	4	4	Yes
TB22	4;2	4;8	M	None	-	-	-	No
TB23	3;9	4;10	F	None	-	-	-	No
TB24	4;2	4;0	F	None	W & E	5	5	Yes
TB25	4;5	4;0	F	None	Welsh	4	5	Yes

Table 44. Participants in TDB Group

Ptp	Age	Dev. Age	Gender	Diagnosis	Home Language	Expressive	Receptive	Individually Matched Sample
TM1	3;1	4;4	M	None	English	5	5	No
TM2	5;11	7;3	F	None	English	5	5	No
TM3	3;10	4;8	M	None	English	4	5	No
TM4	7;0	6;0	M	None	English	4	4	Yes
TM5	3;9	<4;0	M	None	English	5	4	No
TM6	3;10	4;0	M	None	English	5	5	Yes
TM7	3;1	<4;0	F	None	English	5	5	Yes
TM8	3;2	<4;0	F	None	English	4	4	Yes
TM9	5;11	5;6	F	None	English	5	4	Yes
TM10	5;4	5;0	F	None	English	4	5	Yes
TM11	7;1	5;6	M	None	English	5	5	No
TM12	5;6	5;0	F	None	English	4	5	No
TM13	4;6	<4;0	F	None	English	5	5	No
TM14	3;1	<4;0	F	None	English	5	5	Yes
TM15	3;8	4;0	F	None	English	5	5	Yes
TM16	3;9	5;0	F	None	English	4.5	4.5	No
TM17	3;11	<4;0	M	None	English	3.5	4	Yes
TM18	3;3	<4;0	M	None	English	4	4	Yes

Table 45. Participants in TDM Group