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The societal cost of Huntington's disease: are we underestimating the burden?

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Background and purpose: Approximately 9000 people in the UK are affected by Huntington's disease (HD). People with HD require ongoing health and social care support. There is a knowledge gap about costs of health and social care use associated with HD in the UK. This paper estimates the economic cost in the UK.

Methods: Data on UK patients for the year 2013 were extracted from the European Huntington's Disease Network REGISTRY study, a full clinical dataset, including the full medical history and medication history for patients with HD. National unit costs for the price year 2013 were applied to health and social care services.

Results: Data were available for 131 people. The mean annual cost per person with HD was £21 605. The largest proportion of this cost (65%) was due to informal care (£14 085).

Conclusions: Informal care was the largest driver of costs across all stages of HD; thus there is a need to also consider the needs of carers when planning services for people with HD.

Introduction

Huntington's disease (HD) is an inherited neurological disease, characterized by a progressive deterioration of movement, cognition and behaviour. Approximately 12 in every 100 000 people in the UK are affected by HD [1]. Research has been conducted into the economic cost of other neurodegenerative conditions [2,3]. A European study of service use associated with HD has been undertaken [4], but costs were not estimated. One US study of the cost of HD was identified [5]. An estimate of the economic cost of HD in the UK is presented.

Methods

Data

The European Huntington's Disease Network (EHDN) REGISTRY database [6] includes demographics,

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medical history and longitudinal information on patients with HD across Europe. Health and social care use is recorded using a Client Service Receipt Inventory. It contains information on hospital and residential services, primary and community care, diagnostic tests, informal care, aids and adaptations to the home.

Patients enrolled in the EHDN REGISTRY observational study provide informed, written consent for their anonymized data to be used for research purposes. Ethical approval was obtained from Bangor University School of Healthcare Sciences Ethics Committee (ref. 2015-15652). Data for UK patients from 1 January 2013 to 31 December 2013 were analysed. Patients were categorized by total functional capacity (TFC) score. A TFC score of 11–13 is classified as stage I (earliest), 7–10 is stage II, 3–6 is stage III, 1–2 is stage IV and a score of 0 is stage V (latest).

Costing service use

National unit costs for the price year 2013 were applied to the service use data by multiplying the frequency of contacts by a unit cost [7–9]. The

REGISTRY database does not record the length of a contact; therefore a fixed length was assumed for each visit, e.g. 12 min for a general practitioner visit. Medication was costed by multiplying dose and duration of a medication course by its unit cost [9]. Informal care included personal care, help inside the home and help outside the home. The cost of informal care was estimated using the hourly rate of a home care worker as a replacement service (£24 per hour) [7]. Descriptive statistics are used to present the mean, standard deviation and median costs of service use, by TFC score.

Results

Client Service Receipt Inventory data were available for 131 people (10% of REGISTRY patients in 2013). Our sample contained similar proportions of patients in each TFC stage compared to the larger REGISTRY sample; however, our sample had no stage V patients (3% of the REGISTRY sample were stage V). The mean age was 50 years (range 18–78). The sample contained 72 females and 59 males.

Health and social care costs

Mean annual costs per person in stage I were £2250, rising to £89 760 in stage IV. The average cost across all stages was £21 605 per year (Table 1). This equates to £195 million per year in the UK.

Discussion

Average US costs have been estimated to be \$4947 (£3150 in 2013 pounds) per person with HD in the early stage rising to \$22 582 (£14 378 in 2013 pounds) in the late stage for commercial patients, and from \$3257 (£2074 in 2013 pounds) in early stage Medicaid patients to \$37 495 (£23 873 in 2013 pounds) in late stage Medicaid patients [5]. In the US study more Medicaid patients were in long-term care compared to commercial patients, thus driving the cost differences; the authors noted that this may be due to Medicaid patients being at a later disease stage or because data on long-term care for the commercial patients is less complete [5]. In our study, the average annual cost per person was £21 605, which is comparable to the cost of dementia in the UK (£32 242 per person) [10]. Looking at disease severity, our range was greater than in Divino *et al.* [5], at £2250 per person in the earliest stage of HD and £89 760 in the later stages. However, there are substantial differences between the US and UK healthcare systems and the categories of costs included in the respective studies; therefore our findings are only applicable to UK patients.

Table 1 Mean (SD) and median annual costs

Category	All stages (n = 131)			Stage I: TFC 1–3 (n = 43)			Stage II: TFC 7–10 (n = 37)			Stage III: TFC 3–6 (n = 38)			Stage IV: TFC 1–2 (n = 13)		
	Mean	SD	Median	Mean	SD	Median	Mean	SD	Median	Mean	SD	Median	Mean	SD	Median
Primary/ community care	£2680.24	£5768.72	£886.00	£763.35	£758.57	£588.00	£1709.89	£1813.60	£1156.00	£3813.74	£6752.50	£1106.00	£8469.23	£12 312.59	£2112.00
Hospital/ residential care	£2436.76	£9455.52	£0	£99.40	£160.93	£0	£1800.27	£6783.42	£0	£2236.31	£6673.06	£238.00	£12 565.56	£23 686.39	£314.00
Investigations/ diagnostic tests	£19.96	£87.30	£0	£5.16	£17.30	£0	£23.35	£75.19	£0	£35.42	£141.14	£0	£14.15	£38.99	£0
Medication	£851.42	£1675.51	£204.40	£306.53	£883.76	£18.25	£730.14	£1262.71	£261.78	£838.97	£823.49	£623.27	£2976.83	£3847.27	£1653.45
Informal care	£14 085.24	£30 391.87	£0	£1073.86	£5558.46	£0	£4941.41	£8561.39	£18 096.00	£21 051.79	£19 847.84	£18 096.00	£62 784.00	£69 831.12	£38 688.00
Adaptations/ aids	£1531.46	£4837.66	£0	£1.77	£11.59	£0	£2662.70	£7424.02	£0	£1675.68	£4186.72	£0	£2949.85	£4422.71	£162.00
Total	£21 605.08	£36 373.51	£3515.35	£2250.06	£5922.11	£736.95	£11 867.76	£15 749.52	£4944.58	£29 671.91	£23 597.03	£26 814.41	£89 759.62	£69 829.63	£110 939.60

As would be expected with a degenerative condition, costs increased by disease severity, presenting the case for investing in care and support for people in the earlier stages to keep them functioning well. The appropriate time to introduce and subsequently increase social care packages to support people with HD and their informal carers is likely to vary between individuals, but our study has identified that increased social care support is likely to be needed during stages III and IV of HD. Informal care was the largest driver of costs; thus there is a need to consider the needs of carers as well as people with HD. While the study population was representative of the larger REGISTRY sample in terms of TFC stratification, it is not possible to say whether service use data were missing at random, and therefore whether there is inherent bias in our cost estimates due to under-reporting of service use. In particular, our study had no people in stage V. REGISTRY data are collected by interviews in outpatient visits, which may lead to under-representation of people living in long-term care. As such, the estimated costs of HD presented here may be even higher in practice.

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Disclosure of conflicts of interest

The authors declare no financial or other conflicts of interest.

Supporting Information

Additional Supporting Information may be found in the online version of this article:

Data S1. List of EHDN investigators.

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