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Economic burden of neural tube defects in Germany

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ABSTRACT

Objective: Failure of closure of the neural tube often leads to serious malformations, including spina bifida, anencephaly and encephalocoele. Despite improvements in medical and surgical treatment, the burden associated with spina bifida is substantial but country-specific data are lacking outside North America. This study aims to improve understanding of the economic implications and burden associated with the morbidity of children and adults with neural tube defects (NTDs) in Germany.

Study design: Retrospective data analysis.

Methods: 2006–2009 German health insurance data of persons with NTDs (spina bifida and encephalocoele) were analysed to determine the economic burden of illness associated with NTDs in Germany. Cases were identified using ICD-10 codes; data included outpatient and inpatient care, rehabilitation, remedies and medical aids, pharmacotherapy use, long-term care and information on sick leave. The analysis was stratified by age group to provide a burden estimate specific to a person's age. To obtain an indicator of incremental burden to the Statutory Health Insurance (SHI), results were compared to the standardized healthcare expenditures according to the German Risk Compensation Scheme (RSA).

Results: Overall, 4141 persons with an ICD code related to NTDs were identified (out of a population of 7.28 million persons screened). The administrative prevalence ranged from 0.54 to 0.58 per 1000 enrollees. Of those, 3952 (95.4%) were diagnosed with spina bifida. The average annual mean healthcare expenditure of persons with spina bifida was €4532 (95% CI = 4375–4689, SD = 9590, Median = 1000), with inpatient care contributing €1358 (30.0%), outpatient care €644 (14.2%), rehabilitation €29 (0.6%), pharmacotherapy €562 (12.4%), and remedies and medical aids €1939 (42.8%). The incremental cost due to spina bifida was substantially higher than the standardized SHI expenditures for all age groups. The difference was highest for persons ≤10 years old (€10,971 vs €2360 for the age group ≤1, €8599 vs €833 for the age group 2–5 years and €10,601 vs €863 for the age group 6–10 years). The difference was smallest for the age group 41–50 years (€2524 vs €1101) and for 71 years and over (€5278 vs €4389).

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Conclusion: Expenditures of persons with spina bifida exceeded the standardized SHI expenditures, indicating a considerable economic burden. The economic burden is continuous throughout the person's life, with high monetary impact and exposure to the healthcare system (especially in early years of life). Efforts should be devoted to improve the prevention of NTDs and provide appropriate support for persons with NTDs, parents, and caregivers—especially in early years.

Introduction

Neural tube defects (NTDs) represent congenital malformations that result from failures in neural tube closure during embryogenesis.[1] They affect the cranial structures (encephalocoele; anencephaly—for which there is no treatment and the prognosis is death) or spinal structures (spina bifida).[2] The aetiology of NTDs is multifactorial with genetic factors and environmental factors playing an important role.[3] There is evidence that about 72% of NTDs could be prevented with periconceptional folate supplementation and folic acid fortification.[4–6] Prevalence of NTDs varies by region, race and ethnicity.[7] The European Surveillance of Congenital Anomalies (EUROCAT) network identified prevalence of 4.71/1.12 per 10,000 births for spina bifida/encephalocoele in Europe during 2005–2010 (including live births, foetal deaths, still births and pregnancy terminations, but excluding chromosomal cases). German registry data from Mainz and Saxony-Anhalt indicate prevalence of 5.68/1.38 per 10,000 births for spina bifida/encephalocoele during 2005–2010.[8] Those born with NTDs tend to suffer from hydrocephalus, bowel and bladder dysfunction, orthopaedic abnormalities (e.g., clubfoot, scoliosis, kyphosis), and specific cognitive impairments, all of which are often associated with the need for life-long medical management.[7] Progress in the medical management has increased the likelihood of surviving to adulthood;[9,10] however, despite improvements, the burden of NTDs remains substantial.[11–13]

Evidence points to a substantial economic burden associated with NTDs, both in terms of direct (healthcare-related) and indirect costs. A considerable burden is typically observed in the first years of life, with higher healthcare expenditures in children and adolescents with NTDs than in those without the disease. High resource utilization continues throughout adulthood, coupled with negative impact on the labour force participation for both persons with NTDs and their parents or caregivers.[14–20]

With most of the evidence coming from North American studies, there is a lack of data describing the burden of NTDs in Europe. The main objective of this study was to improve the understanding of the economic implications and burden associated with the morbidity of children and adults with NTDs in Germany.

Methods

Data source and study population

Data from Techniker Krankenkasse, a large nationwide sickness fund in Germany, were used. The extracted data covered person-specific information for employees, (early) retirees, and their dependents on outpatient and inpatient care, rehabilitation, remedies and medical aids, pharmacotherapy, long-term care, as well as information on incapacity to work (sick leave exceeding three days) for the period 2006–2009. Outpatient care was defined as healthcare delivery by general practitioners and specialists outside hospitals in an ambulatory setting, whereas inpatient care refers to hospitalizations.

The population of Germany is about 82 million and approximately 85% (around 70 million) is covered by SHI. During 2006–2008, the fund annually insured approximately six million individuals, with the number increasing to seven million in 2009. Overall, between 8.7% and 10.5% of the total SHI population was covered by the respective sickness fund in 2006–2009. Techniker Krankenkasse is the second largest sickness fund in Germany. Compared to the SHI average, it insures higher proportions in younger and middle ages and smaller proportions of those in higher ages. In addition, it insures fewer women (48% compared to 53% SHI average); therefore, prevalence of NTDs can deviate. Despite these differences, the tendency of results is considered representative for Germany as a whole because the fund operates on a nationwide level and a substantial proportion of the population was used to study the impact of NTDs without local restrictions. The list of services covered by SHI is equal for all sickness funds, so differences in treatment patterns, resource use and cost due to differences in service level or regional concentration of the insured population are less likely.

As this study was a secondary analysis of pre-existing data and not based on primary research, ethical approval was not sought; however, data usage was approved by the Internal Data Protection Commissioner of the Techniker Krankenkasse and is in line with current data protection acts. All data were anonymized, with personal and other identifying information removed to assure data privacy.

Case identification

The study population was chosen based on ICD-10 codes from either inpatient or outpatient claims during 2006–2009. To be included in the study, the insured had to have at least one of the ICD-10 codes of interest: Q01.- (encephalocoele), Q05.- (spina bifida), Q76.0 (spina bifida occulta). For outpatient claims, only confirmed ('gesicherte') diagnoses were considered. The main analysis focused on persons with spina bifida.

All inclusion criteria were applied to the available claims data from the 2006–2009 period, with data from multiple years of the respective period used for subject identification.