



Cost of illness in autism: a revision of methods

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1) Introduction

ASDEU study (Autism Spectrum Disorders in the European Union) is a project funded by the European Commission whose main objective is to estimate the prevalence of autism in Europe (see more information in www.asdeu.eu).

One of the secondary objectives is to estimate the **economic burden of autism** in 14 European countries, using methods of **cost-of-illness (COI) studies**. There are different types of COI studies. Here we discuss the different types and methods and their validity for autism.

2) Rationale – Main points

Since the first study of the overall costs of autism in UK (Järbrink and Knapp, 2001), several COI studies have been published. There are **three main approaches** to estimate the economic burden of a disease:

1. TOP-DOWN METHOD

- Less expensive
- Not feasible if there are no aggregated data and the experts admit no knowledge about the use of resources related to the disease.
- It relies on the availability and quality of epidemiological evidence → more suitable for highly prevalent diseases.
- No examples of COI studies for autism found.

2. BOTTOM-UP METHOD

- Traditionally more expensive because it needs individual data → direct measurement from questionnaires for patients and/or carers or complete and comprehensive registries (less expensive currently due to new technologies).
- Average costs per person are extrapolated to the whole population using prevalence or incidence data.
- More comprehensive for complex diseases → it doesn't rely on epidemiologic data and it can capture variability related to differences in important demographic or clinical characteristics between patients.
- There are studies with limited samples (Barret et al. 2012; van Steensel et al. 2013) or bigger samples such as an ongoing study in Scotland (The Microsegmentation Study, MacKay et al. 2013) apart from ASDEU Project.

3. MIXED APPROACH

- Combines data from several sources including direct measurements, data from literature, data from national or local databases, experts' opinions, or assumptions.
- Feasible in countries, such as UK, where there is information of use of resources and costs based on statistics, previous studies and other data.
- Some authors have used this approach (Ganz, 2007; Knapp et al. 2009; Buescher et al. 2014; Lavelle et al. 2014). Knapp et al. developed a modular approach which comprised five elements: estimation of prevalence, identification of place of residence, identification of level of functioning and other individual characteristics, estimation of the associated service use patterns, and calculation of cost per individual.

References

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Autism Spectrum Disorders in the European Union

The ASDEU Project	
Design	Cross-sectional study
Target population	Children and adults with autism (at least 6 months since diagnosis) or their relatives/carers answering on behalf of the individual with autism.
Countries	Austria, Belgium, Bulgaria, Denmark, Finland, France, Iceland, Ireland, Italy, Poland, Portugal, Romania, Spain, United Kingdom
Sample size	No predetermined; recruitment by convenience sampling.
Data collection	Internet-based survey, translated into 13 languages and adapted to 14 countries. Anonymous web-based questionnaire collecting data of use of services related to autism (healthcare services, social services, education, housing, etc.) in the last 6 months by people on the autism spectrum and families, and productivity losses related to the autism.
Perspective	Societal perspective: direct costs and indirect costs are included.
Direct costs	<ul style="list-style-type: none"> • Education services • Inpatient hospital services • Residential respite care • Other health and social care services
Productivity losses (indirect costs)	Productivity loss due to cessation or reduction of working time by parent's or other relatives'. It will be measured by means of the human capital method.
Estimation	Prevalence-based estimation: Current annual economic burden related to the prevalence of a disease or condition during a year (time horizon).
Approach	<p>Bottom-up approach: Average cost multiplied by the prevalence; costs are estimated by adding individual costs.</p> <p>Alternative: If it were not possible to launch the survey or the collected data were scarce in some countries, the alternative is a mixed approach combining data obtained from different sources and using the modular approach by Knapp et al.</p>

3) Conclusions

- The **bottom-up approach** have scientific advantages over the top-down approach.
- The methods for estimation of economic burden of autism could differ for different countries **depending on the available information**.
- The **mixed approach** could be an alternative, if there is available information, when the bottom-up approach is not a feasible option.
- In some countries, where there is a scarcity of previous studies and regional and national statistics, **collaboration of people with autism and families** is needed to gather the required data, because estimation based on experts' opinions would not be accurate.
- In **ASDEU Project** we have chosen to gather data directly from people on the autism spectrum and their relatives/carers (**Internet-based survey** for all persons affected by autism and willing to answer).