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Original Research

Costs of treatment of adult patients with cystic fibrosis in Poland and internationally

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ABSTRACT

Objectives: Despite its low prevalence, cystic fibrosis (CF) may have a considerable impact on healthcare system expenditures in terms of direct healthcare costs and lost productivity. This study was aimed at calculation of costs associated with CF treatment in Poland, as well as at comparison of average costs of treatment of CF patients in selected countries, taking into account the purchasing power parity.

Study design: Retrospective study.

Methods: The researchers undertook a retrospective study of adult patients with CF taking into account the broadest social perspective possible. Medical and non-medical direct costs as well as indirect costs were calculated. CF costs estimated by researchers from other countries over the last 15 years were also compared.

Results: Total annual treatment cost per one CF patient in Poland was on average EUR 19,581.08. Costs of treatment of CF patients over the last 15 years varied between the countries and ranged from EUR 23,330.82 in Bulgaria to EUR 68,696.42 in the United States.

Conclusions: CF is an international problem. The data in this study could be the baseline for integrated and harmonised approaches for periodical assessment of the future impact of new public policies and interventions for rare diseases at the national and international levels.

Introduction

In European Union (EU) countries, any disease affecting fewer than five people out of 10,000 is considered rare. That number may seem small, but it translates into approximately 246,000 people throughout the EU's 28 member countries.[1]

One of the key rare diseases is cystic fibrosis (CF), with a prevalence in Europe is estimated at one case per 8000–10,000 people.[2] American researchers report its incidence in the United States between 1:1900 and 1:3700.[3] In 2008, Farrell[4] estimated the incidence of CF in the EU countries at between 1:1353 in Ireland and 1:25,000 in Finland. Polish register of patients with CF kept by the Polish Cystic Fibrosis Society lists over 1300 patients, of which around 300 are adults.[5] Not all CF patients are listed in the register as registration is optional. The mean age of a CF patient was 20.6 years in 2014.[6]

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Despite its low prevalence, CF may have a considerable impact on healthcare system expenditures in terms of direct healthcare costs and lost productivity as orphan drugs are frequently very expensive and CF symptoms tend to appear at a young age.[7–10]

The aim of a cost of illness study is to identify and measure all the costs of a particular disease, including the direct, indirect and intangible dimensions. The output, expressed in monetary terms, is an estimate of the total burden of a particular disease to society.[11] Knowledge of the costs of an illness can help policymakers to decide which diseases need to be addressed first by healthcare and prevention policy. Additionally, these studies can indicate for which diseases cures would be valuable in reducing the burden of disease. For specific stakeholders, such as the federal government, cost of illness studies can show the financial impact a disease has on public budget.[11] Numerous cost of illness studies have been conducted over the past decades across a range of diseases, however, few have addressed rare diseases, in particular CF. In this context, this study was aimed at calculation of costs associated with CF treatment in Poland as well as at comparison of average costs of treatment of CF patients in selected countries, taking into account the purchasing power parity.

Methods

Literature review

We reviewed relevant literature on costs of CF from the last 15 years. The following databases were browsed: MEDLINE, Web of Science and EMBASE. The search words had to be part of the title or abstract. The search terms used were 'cystic fibrosis' AND 'cost of illness'; 'cystic fibrosis' AND 'economic evaluation'; 'cystic fibrosis' AND 'cost study' and 'cystic fibrosis' AND 'cost analysis'. Language was restricted to English. No other limitations were specified. A search on the references cited in the reviewed papers was also performed.

Inclusion criteria for publications in this review were as follows: 1) analysis of cost for adult population (>18 years of age); 2) average value of total CF treatment costs estimated in the study; and 3) cost analysis conducted between 2000 and 2015 (15 years). International references were included in the conducted comparative analysis mainly on the basis of the fact that the calculated costs applied to CF treatment only. This prevented overestimation of the costs. The last search was performed on January 12, 2016.

To compare costs calculated in different years, the calculation was discounted at 5% annual rate. To take into account the differences in price levels between countries, costs were adjusted for purchasing power parities in 2013.[12]

Study population

The researchers undertook a retrospective study, taking into account the broadest social perspective possible. Having analysed medical documentation of the patients, they included a group of 46 adult (>18 years of age) patients (28 female and 18 male) treated in 2013 by a team of pulmonologists from the CF clinic at the pulmonology ward at one of the Poznań hospitals. The mean age of the patients in the study group was 27.4 years, where the youngest subject was 19 and the oldest was 42. A vast majority of the study subjects were not residents of Poznań ($n = 42$). Only four subjects resided in Poznań (Table 1).

Study inclusion criteria were as follows: a) age above 18 years; b) CF diagnosed according to International Statistical Classification of Diseases and Related Health Problems 10th Revision (ICD-10) classification; and c) being under continuous pulmonologist's supervision between January 2013 and December 2013. The patients' data were obtained from case history forms, doctors' request cards and medical records. A number of different information about the patients was collected, including their present therapy as well as demographic (e.g. age, sex) and clinical details (e.g. the number of outpatient visits, hospitalization, pharmacotherapy etc.). Time horizon of the study was 1 calendar year (January 1, 2013 until December 31, 2013). The study was approved by the Bioethics Committee of the University of Medical Sciences.

Calculation of costs

Direct (medical and non-medical) and indirect costs of CF were calculated. The following types of direct costs were taken into account: hospitalization, outpatient visits, pharmacotherapy, diagnostic tests and transportation. Indirect costs were associated with presenteeism. Indirect costs of CF were estimated using the human capital approach. As a financial measure of human capital, gross added value per person employed in the national economy was used.[13]

The data required for the analysis were obtained from: the patients' case histories, doctors' request cards, the patients' hospital discharge summaries and data collected by the Central Statistical Office. Pharmacotherapy costs were calculated based on wholesale drugs price list. Laboratory test prices were calculated based on the hospital laboratory's price list. Outpatient visit price was calculated based on the price list for medical procedures in place at the hospital, under the agreement with the National Health Fund. Hospital

Table 1 – Study population characteristics.

Variable	Value
Number of patients (*n*)	46
Women/men (*n*)	28/18
Mean age in years (±SD)	27.4 (8.01)
Youngest/oldest, in years (±SD)	19 (7.2)/42 (10.01)
Poznań residents/non-Poznań residents (*n*)	4/42
Education (*n*)	
Higher	18
Secondary	23
Vocational	4
Primary	1
Source of income (*n*)	
Supported by parents	11
Full-time job	8
Disability pension awarded due to CF	27
Average number of days at the hospital ward (±SD)	27.8 (10.01)
Average number of outpatient visits (±SD)	2.74 (1.48)

SD, standard deviation; CF, cystic fibrosis.