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CHAPTER 286

Cost of Treating Patients

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INTRODUCTION

Studies of the cost of epilepsy or cost-effectiveness evaluations of care for epilepsy were, until recently, quite rare, and the methods and data used in both the new and older studies are difficult to compare (12,14). The information presented in this chapter is designed to assist persons who are not economists in understanding the concepts and methods used to estimate the costs of illness and the specific problems of their application to epilepsy.

Also presented are the findings of several recent examples of studies of the costs of epilepsy, with brief comments on the data and methods of estimation. The issues discussed include the more serious methodologic obstacles to estimating the costs of epilepsy, the problem of separating the costs of epilepsy from the costs of coexisting illnesses, the problems associated with sampling and sample sizes, and the confusion that exists among many clinicians about the concepts on which cost estimates are based. For the chapter to accomplish its purpose, agreement on the meaning of several concepts and terms is required.

CONCEPTS AND DEFINITIONS

Definition of Epilepsy

Most studies of the costs of epilepsy have included persons with single seizures. Many epileptologists do not classify cases of single seizures as epilepsy, especially if the seizures are febrile seizures of early childhood or are precipitated by specific factors, such as certain drugs. The inclusion of cases of single seizures in cost studies creates a cost distribution that includes a large number of persons with low health care costs and near-zero indirect costs, and a small number of persons for whom epilepsy creates substantial losses. The common practice of using average total costs from these distributions to represent individual costs of epilepsy is deceptive, because the means are poor measures of the costs to either group.

Definitions of Costs

Terms such as *costs* have common usages that differ from the definitions used by economists. This section provides some definitions that are needed to understand the concepts on which cost estimates are based.

Opportunity costs are the costs of resources (i.e., capital, labor, and natural resources) measured in terms of the value of the outputs that could have been produced by using the same resources in their next most efficient employment. If, for example, a physician spends 1 hour treating a person with epilepsy, the opportunity cost of that hour is the value of an hour-long treatment of other patients by the same physician. Empiric cost estimates rarely measure opportunity costs, relying instead on payments to providers, which are the values recovered by accounting systems.

Social cost can be considered a synonym for *opportunity cost*. *Private costs*, the amounts paid by individuals or organizations for goods or services, may or may not equal social costs. If, for example, government subsidizes health insurance, then consumers' payments for health care (their private cost) are less than the social costs of producing their care.

Direct costs are the costs of goods and services used to treat epilepsy. Payments for health care and rehabilitation services are likely to be the largest components of the direct costs of epilepsy.

Indirect costs are the value of the outputs that would be produced in the home or the work place if individuals' productivity had not been limited by epilepsy. The indi-

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rect costs of epilepsy include wages losses, losses of household production, and the costs of care provided to a person with epilepsy by members of the household.

Transfer payments is a term most frequently applied to public assistance payments, but its meaning is more general. A transfer payment is an exchange of ownership of a resource that does not change the stock of resources. As social costs are incurred only when resources are consumed, a transfer payment is not a social cost. Many clinicians mistakenly count transfer payments as costs in their studies of epilepsy (4).

Many estimates of the costs of epilepsy are used by *cost-benefit* or *cost-effectiveness analyses* (*CEA*) of alternative methods of care. *Cost-benefit analysis* values the benefits produced by an activity and compares the benefits with their costs of production. The benefit-to-cost ratio is then compared with the ratios for alternative activities, which need not produce similar outcomes. Thus, the benefit-to-cost ratio for an antiepileptic drug could be compared with the benefit-to-cost ratio for construction of a football stadium. *Cost-effectiveness analysis* compares the efficiency of alternative methods of producing equivalent outcomes. It has become conventional to define the outputs in CEA in natural units (years of life, days of work loss) rather than in monetary values. Cost-effective analysis is a variant of cost-benefit analysis and, like cost-benefit analysis, it refers to efficiency criteria without regard to equity, ethics, or distribution of income. The most *efficient* use of funds could, for example, be for programs that benefit wealthy adults but deny funds to hungry children.[1]

The literature on the economic aspects of epilepsy is marked by very sharp differences in methodology. The next section describes methods for estimating the costs of epilepsy, discusses the need to separate the effects of epilepsy from the effects of coexisting illnesses, and describes some methods of achieving that objective.

METHODS

The literature on the estimation of the costs of illness is extensive. The most recent discussions of the topic and of the execution of cost-effectiveness studies are the 1995 report of the Task Force on Principles for Economic Analysis of Health Care Technology (the "Task Force") (16) and the report of the government panel on cost effectiveness in health and medicine (8).

Estimating Direct Costs

The estimation of the health care costs for epilepsy requires information on payments to health care providers and the ability to distinguish care required for epilepsy from the treatment of other illnesses that may affect persons with epilepsy. The allocation of the costs of care between epilepsy and coexisting illnesses is discussed in a subsequent section.

The only other obstacle to the estimation of the direct costs of epilepsy is the need to obtain data on payments to providers rather than the charges that providers submit for their services. It is often difficult to identify the amount paid for health care, because providers bill charges that are subject to different discounts for different payers. Payments data, which are the most direct measure of costs, do not reflect coinsurance or deductible payments by consumers, but they can be adjusted to represent full costs. It is unfortunate that so little use is made of the information on the costs of epilepsy contained in large insurance claims data sets. Information on the indirect costs of epilepsy, which are considered next, is much more limited than the data on direct costs

Estimating Indirect Costs

Wage Losses

The wage losses associated with epilepsy are often estimated by comparing the employment of persons with epilepsy with average employment rates and assuming that the difference represents the effects of epilepsy on employment and earnings. This approach ignores the fact that the employment and earnings of persons with health impairments are influenced by disability insurance benefits (a disincentive to work), labor market conditions, and employer discrimination in addition to the effects of illness (Butler 1995; Baldwin and Johnson 1995; 1,6).[2] Studies of attitudes towards persons with disabilities agree that attitudes towards persons with epilepsy are extremely negative (Baldwin and Johnson 1993). Employers base hiring decisions on their judgments of workers' productivity. Those judgments may be biased because of prejudicial attitudes. No one has estimated the contribution of discrimination to the indirect costs of epilepsy, but it is likely to be significant. The omission of the costs of discrimination can bias the benefits, stated in terms of reductions in indirect costs, that improve productivity but do not change patterns of discrimination. One study that considers socioeconomic influences on work decisions among persons with epilepsy

[1]Louise Russell has pointed out that in practice, CEA are unlikely to yield such a result, as they do not value outcomes according to willingness-to-pay criteria (Louise Russell, *personal communication*).

[2]The most important work disincentives for persons with epilepsy may be caused by the health insurance coverage provided by programs such as Social Security Disability Insurance (SSDI) and Supplemental Security Income (SSI). Persons with epilepsy have difficulty obtaining individual health insurance and are often excluded from employment-related insurance coverage or required to pay higher premiums than their fellow workers (2,5). The lifetime health insurance given to SSDI and SSI beneficiaries is of such great value to persons with epilepsy that it may be a stronger disincentive to work than disability benefit payments..

has found that they are as important as the direct effects of seizures on the employment and earnings of persons with epilepsy (13).

The omission of these influences can bias evaluations of the effectiveness of care or drugs as well as estimates of the costs of epilepsy. Although the data needed to control for these influences may be difficult to obtain, it is important that estimates of the indirect costs of epilepsy recognize the potential for error when the effects of these characteristics are omitted.

Another element of indirect costs that should be considered is the loss of non-wage compensation or employers' payments for fringe benefits. The compensation of labor is commonly referred to as *wages*, but non-wage compensation represents as much as 30–40% of workers' real earnings and should be included in the calculation of indirect costs. No study of the costs of epilepsy has, to the knowledge of the authors, included losses of non-wage compensation. The omission of these losses, all else being equal, substantially understates the actual indirect costs of epilepsy.[3] The understatement is partially offset in those studies that measure private rather than social costs by the equally common practice of measuring wage losses in terms of gross wages rather than after-tax wages.

Losses of Household Production

Although household production is not included in the calculation of gross national product, it is a large segment of real output. the authors are aware of only one estimate of losses of household production attributable to epilepsy.[4] Illness-related losses of household production are usually estimated either by valuing lost work time in the home based on the wage that a homemaker could earn in the labor force, or by valuing commodities (e.g., meals, laundry) done without because of epilepsy based on the prices paid for similar commodities in the market.[5]

The wage-based method: The rationale for valuing household work at a wage rate is the concept that wages are the opportunity cost of time spent in household work. If, for example, a person can earn an hourly wage that exceeds the hourly wage paid to hire homemakers, the individual is expected to work for wages rather than working at home. The comparative-wage rationale is subject to a number of qualifications, but it is a good representation of the average effect of wage opportunities on the choice between working at home and working in the labor force.[6]

The "replacement" method: The rationale for valuing household work at the cost of purchasing similar goods and services is the assumption that the outputs of household production are substitutes for similar commodities that would be purchased in household replacement. Meal •4• preparation, for example, would be valued at the prices charged by restaurants for similar meals. The replacement-value approach suffers from two biases that somewhat offset each other. One is the assumption that householders are as efficient as market producers, and the other is the failure to consider the economics of jointly producing products at home that must be purchased separately in the market. The use of cab fares as the proxy for the value of transporting children, for example, ignores the fact that many errands are often performed in a single trip by a homemaker.

The Effects of Multiple Conditions

The most serious obstacle to the estimation of both the direct and indirect costs of epilepsy is the need to factor out the costs of other illnesses that affect persons with epilepsy. People with epilepsy are often affected by structural tumors, stroke, significant diffuse brain injuries, and other conditions that may be more costly than epilepsy itself.

The prevalence of multiple illnesses is high among persons with epilepsy, but most estimates of the costs of epilepsy attribute the costs of care and losses of productivity to epilepsy alone. This practice overstates the costs of epilepsy, and thereby overstates the potential benefits of interventions that reduce the incidence or severity of seizures. A reduction in the need for physician office visits to monitor the effects of antiepileptic drugs has, for example, little impact on health care costs if the office visits are still needed for the treatment of coexisting illnesses. Similarly, if a person's ability to work is limited by the combined effects of seizures and other illnesses, reducing seizures may not increase functional capacity sufficiently to eliminate the limitation. Thus, attributing the loss of output to epilepsy is inappropriate.

Generally, persons with two or more illnesses experience twice as many days of restricted activity as persons with only one condition (17). Among persons with epilepsy only, the prevalence of work impairments is one-fifth as great as the prevalence of work impairments among persons with epilepsy and one or more other ill- •5•
nesses (Rodin et al. 1977).

[3]Because some coverages pay benefits for disability and health care, the estimation of losses of non-wage compensation can be difficult. The method of including losses of non-wage compensation in estimates of cost of illness is discussed by Weiler et al. (18).

[4]Begley et al. (3) estimate household production losses for a set of hypothetical patients. The results are discussed in a subsequent section of this chapter.

[5]Methods of calculating losses in household production are reviewed in King and Smith (9; part IV).

[6]Qualifications include the possibility that household production permits the consumption of benefits—such as spending time with young children, a flexible work schedule, and freedom from supervision—that are not reflected in the value of outputs produced at home but that influence the choice between work for wages and work at home.

The problem of identifying the costs of epilepsy is particularly difficult in the case of persons with epilepsy and conditions, such as mental retardation, that sometimes require institutional care. As noted by Cockerell (4), 35% of the costs attributed to epilepsy by the 1978 U.S. study (5) represented the cost of institutional care for persons with mental handicaps and epilepsy.

The authors suggest that studies of epilepsy cannot effectively distinguish among the costs of multiple conditions for persons in institutional care. Cost estimates for persons who require institutional care might be best presented as a separate cost category accompanied by some reasoned speculation about the division of costs among the multiple conditions.

There are no completely adequate methods for separating the costs of epilepsy from the costs of coexisting conditions. Three methods of analyzing group differences in costs are described in the next section, and the merits and limitations of each are briefly discussed. Complete discussions of the methods are found elsewhere.

Cost Estimates and Comparison Groups

The three methods to be discussed are control group-experimental group comparisons, attribution of costs by procedure (CPT) or diagnosis (ICD-9) codes, and decomposition of cost differentials. These methods provide an approach to separating the costs of epilepsy from the costs of other illnesses, but they yield approximations that are to be used cautiously.

The control group-experimental group method is used quite often in the study of the clinical interventions, such as comparisons of antiepileptic drugs. Two groups of patients are selected who are as nearly identical as possible in respect to all relevant characteristics; each group receives a different intervention, or one is treated and one is given a placebo, and the differences between the outcomes for each group, such as the average frequency or severity of seizures, are used to measure the effects of the intervention.

The use of control group-experimental group comparisons to distinguish the costs of epilepsy from the costs of coexisting illnesses is extremely difficult, because the factors that influence costs are so numerous that the group of cases from which controls are to be drawn may be too large to be practical. The determinants of lost work days, for example, include workers' tenure with the firm in which they are employed at the onset of an illness, the ratio of disability benefits to potential wages, membership in a labor union, age, education, ethnicity, sex, and the physical demands of their usual work. Thus, an ideal match requires a sample of control cases of sufficient size to permit matching on at least nine characteristics, in addition to controlling for differences between groups in the nature of the multiple conditions affecting persons in each group. No study of epilepsy has ever matched a control and experimental group on such a large number of characteristics. The largest of the clinical studies of epilepsy, the Veterans Administration cooperative study of antiepileptic drugs, is based on 622 persons who were randomly assigned to a group with their predominant seizure type. The matching characteristics of patients in each of four drug treatment groups were age, sex, education, IQ, and cause of seizures. Approximately 198 patients remained in the study until its conclusion (10).

The diagnosis-procedure method requires a judgment to be made as to which of the health care procedures supplied to a person with a diagnosis of epilepsy are required to treat the effects of epilepsy, and which would be required in the absence of epilepsy. It is possible, for example, to use ICD-9 codes to identify treatments that are attributable to the effects of epilepsy.[7] The costs of treatment can be allocated among these ICD-9 codes by reference to the information supplied on insurance claim files. The information can be supplemented by classification schemes based on CPT codes to distinguish further between procedures used to treat epilepsy and procedures used to treat other conditions that affect persons with epilepsy. The validity of the method depends on the accuracy of the ICD-9 and CPT coding by health care providers. Retrospective studies are limited by the common practice of coding only the primary diagnosis for claims data. In a prospective study, in which physicians were aware of the importance of complete coding, the coding could include the fifth digit and physicians would code every existing condition.

The decomposition method analyzes group differences in outcomes, such as the average cost of care, by comparing the means and coefficients obtained from estimating the same multivariate equation separately for an experimental and a control group.[8] In the context of this

[7]An illustrative example, suggested by Dr. Robert Fisher of the Barrow Neurological Institute, includes the following categories:

Partial epilepsy, without generalization: ICD-9 345.4 psychomotor epilepsy, limbic epilepsy *or* ICD-9 345.5 focal motor epilepsy, partial epilepsy, jacksonian epilepsy, *not* ICD-9 345.9 convulsions or ICD-9 345.1 tonic-clonic epilepsy.

Partial epilepsy, with generalization: ICD-9 345.4 psychomotor epilepsy, limbic epilepsy *or* ICD-9 345.5 focal motor epilepsy, partial epilepsy, jacksonian epilepsy *and* ICD-9 345.9 convulsions *and* ICD-9 345.1 tonic-clonic epilepsy.

Generalized tonic-clonic epilepsy without specified partial onset: ICD-9 345.1 tonic-clonic epilepsy *or* ICD-9 345.9 convulsions *not* ICD-9 345.4 psychomotor epilepsy, limbic epilepsy *not* ICD-9 345.5 focal motor epilepsy, partial epilepsy, jacksonian epilepsy.

Absence epilepsy: ICD-9 345.0 absence epilepsy, *petit mal* epilepsy.

Pediatric "minor motor" epilepsies: ICD-9 345.0 akinetic (not atonic) epilepsy *or* ICD-9 345.6 salaam seizure, infantile spasm.

Status epilepticus: ICD-9 345.2 *petit mal* status or ICD-9 345.3 *grand mal* status epilepticus *or* ICD-9 345.7 psychomotor status, focal motor status.

[8]The technique, which has never been applied to the costs of epilepsy, has been used for nearly 40 years to study wage discrimination against minority workers. Its first application to health care costs is a comparison between the health care costs of workers compensation and the costs of similar workers who have conventional health insurance (Johnson et al. 1993). *Psychogenic seizures, nonepileptic seizures*: ICD-9 300.11 hysterical convulsion, pseudoseizure, psychogenic seizure.

•6•

discussion, one group could be composed of persons with epilepsy only, and a second group of persons with multiple conditions.

The first step is to express average total cost as a function of the determinants of costs of care. A very simplified example equation could be written as follows:

$$C_i = \alpha + \beta X_i + \mu_i$$

where C_i = health care costs for the ith individual;
I = eo (epilepsy only), m (multiple conditions);
X = a vector of individual characteristics (X_i) that affect the costs of care, such as frequency and severity of seizures, type of seizures;
α,β = parameters to be estimated;
μ = a zero mean, standard normal residual term.

Equations of this type would be estimated separately for each of the comparison groups. The differences between the average total costs for each group are then separated into a part attributable to differences between the average characteristics of patients in the two groups and a part attributed to the costs of conditions other than epilepsy and an unexplained residual. Thus,

$$\bar{C}_{eo} - \bar{C}_m = (\bar{X}_m - \bar{X}_{eo})(\beta_{eo}) + (\bar{X}_{eo})(\beta_m - \beta_{eo})$$

The first term on the right side represents the part of the between-groups difference in average total costs that can be attributed to differences between characteristics of patients in the two groups that can influence costs. The second term on the right side represents the maximal portion of the cost differential that can be attributed to conditions other than epilepsy. The weakness of the decomposition method is that "costs attributable to multiple conditions" is a residual term that is biased if the estimating equations omit significant determinants of costs.[9]

Many of the studies reviewed in the next section fail to separate the costs of epilepsy from the costs of coexisting conditions. Until such distinctions are made, the estimated costs of epilepsy are, all else being equal, likely to be higher than the true costs.

THE COSTS OF EPILEPSY

Recent studies of the costs of epilepsy are reviewed in this section. The articles reviewed include three types of estimates, the first of which is based on representative samples of persons with epilepsy. The second type of study, called *small-group studies*, is based on patients from a particular clinical practice rather than a representative sample of persons with epilepsy. The third type of study, *collage estimates*, combines hypothetical regimens of care for hypothetical patients with cost data from secondary sources.

Estimates from Representative Samples

Representative samples of reasonable size of persons with epilepsy are difficult to obtain from general-purpose surveys, such as the National Health Interview Survey or the National Medical Expenditures Survey (NMES), because prevalence rates for epilepsy are quite low. Representative samples of persons with epilepsy have been created in the UK, and researchers have combined interview data for the sample cases with the subjects' health records to study the costs of epilepsy (4). The estimates are in terms of annual social costs, and the authors are careful to exclude transfer payments from estimated costs. Losses of household productivity are not considered.[10] The results are probably the best estimates of the costs of epilepsy. Indirect costs, of which lost work days and excess mortality are the primary components, are approximately $2.9 billion annually, more than eight times the annual costs of medical care ($374 million).[11] The average annual cost per person is estimated to be $9874, of which approximately $871 is for noninstitutional medical care. If the costs of institutional care and special schooling are excluded, the average annual cost per person is $7216.[12] Maintaining these exclusions, the authors divide the sample into active cases (one or more seizures in the preceding 24 months) and inactive cases (no seizures in the preceding 24 months).

The average annual total cost is $9159 for active cases and $3583 for inactive cases. The largest single contributor to total cost is the wage loss associated with unemployment. The unemployment rate for active cases (28%) is more than triple the unemployment rate for inactive cases, and wage losses represent most of the total cost for the active group.

[9]The same criticism applies to control group-experimental group comparisons, which control for relatively few of the determinants of costs.

[10]The duration for losses resulting from excess mortality is specified as the period from average age at death (assumed to be age 38) to the end of work life (assumed to occur at age 65). Age 65 is not a good measure of the end of work life for most age cohorts in the United States. It may be more accurate for the UK. The annual cost of excess mortality is represented by the present value of excess mortality in the base year. The nominal discount rate of 6% corresponds to the rate used by the government of the UK. The effective discount rate is more than twice as high as the 3% real rate recommended by the government panel, because constant, nominal, average wages are used to estimate losses of productivity—that is, it is assumed that wages do not increase with inflation or with increases in the productivity of labor.

[11]All estimates from this study are converted to 1995 dollars using the Medical Care Component of the CPI-U, November 1995, to make them approximately comparable with the estimates of Murray et al. (MEDTAP) (11). The MEDTAP report, which states that its estimates are in 1995 dollars, was published on November 30, 1995. The authors' assumption may not, therefore, exactly match the MEDTAP calculation, but the differences are likely to be small. Dollar values are obtained by using the ratio of 1.5 dollars to 1 pound sterling, which is used in the original study. The estimates described in this section are converted from the original results of Cockerell (4; Table 5, page 254).

[12]Information is not provided on the proportion of indirect costs attributable to persons requiring institutional care or special schooling.

The results demonstrate that there are at least three very different groups of persons with epilepsy in terms of costs incurred. The highest per-person costs are for those who require institutional care or special schooling. These people typically suffer from multiple conditions, which, as has been discussed, makes it nearly impossible to identify the costs that are strictly attributable to epilepsy. The second group, Cockerell's "active" patients, has the next-highest per-person costs, and it is the largest contributor to total costs because the group is large relative to the number of institutionalized persons. The largest component of costs for active patients is the loss of wage income. Although not measured by the study, losses of household productivity among the active patients are also likely to be large relative to direct costs. The third and lowest-cost group comprises persons without seizures, whose costs are primarily expenditures for drugs.

The results emphasize the need to separate cost estimates for epilepsy by severity into at least the three categories that have been described. Summary cost estimates combining the three groups are deceptive, because they obscure substantial differences in costs between the groups and do not adequately represent important differences in the factors contributing to the costs.

The only other estimate of the costs of epilepsy incorporating data from a national sample uses self-reported data from persons with epilepsy selected from the 1987 NMES. It seems likely that the data do not represent the U.S. population of persons with epilepsy.[13]

The epilepsy sample is also quite small, and the sample weights attached to each case are very large. The sample consists of 113 adults and 15 children who are used to represent a population of 926,000 persons, so the average case represents more than 7235 persons[14] (11). When the authors classify the data by severity, persons with severe epilepsy are represented by 13 adults and 4 children, and nearly two thirds of the average cost of care for adults in the severe class is attributable to one surgery (11; see Table 10). It is difficult, therefore, to place much confidence in the cost estimates.

The estimated annual cost of health care for persons with epilepsy for the year 1995 is $1508 per patient, or a total of $2.2 billion for the prevalent population. Lifetime direct costs of care for epilepsy are estimated at $15,416 per person, or a population total of slightly more than $14 billion.[15]

The NMES estimates are compared with estimates derived from an alternative method, the physician panel study, that combines hypothetical patient profiles and expert judgments concerning their treatment with secondary data on unit costs of health care services. It is, in the terminology of this chapter, a collage estimate. The physician panel data are assumed to represent an incident population of 118,630 persons and a prevalent population of approximately 1.5 million persons. The panel estimate for annual average lifetime cost is lower than the NMES estimate, but the estimates of total costs are only slightly different ($13.7 billion vs. $14.2 billion) because the population to which the panel estimates apply is larger than the NMES population (1.5 million vs. 926,000)

The MEDTAP results are interesting because they are •8• the only estimates for the United States that are derived from a national survey. The extension of the results to larger populations is, however, suspect, because the NMES data on persons with epilepsy may not be a representative sample. In addition, the small size of the data set implies that the confidence intervals surrounding the estimates, if representative, are large.

The studies of the next group have the advantage of direct measures of costs and services, but they are limited by small study populations that are not representative of larger populations.

Small-Group Studies

Swingler et al. (15) estimated health care costs and indirect costs (represented by transfer payments) for 300 patients of a specialized epilepsy service in the UK. The data sources are a survey of the patients and their clinical records. Health care costs are defined from the perspective of a specialist clinic (private rather than social costs), but the analysis and conclusions are phrased in terms of a social perspective.

Slightly more than one third of the patients worked in nonsheltered employment. Another third were not employed because of the effects of epilepsy. Most of the remaining patients were housewives or students.[16] Transfer payments, used as a proxy for indirect costs, account for three quarters of the total costs. Some of the transfers are not attributable to the effects of epilepsy, but even so, the estimates understate indirect costs because they exclude the costs of excess mortality and losses of household production.

The study of Famulari (7) of 160 patients of an epilepsy clinic is unique for its application of labor market models and econometric techniques to the analysis of the costs of epilepsy. She constructs a measure of severity based on seizure characteristics and includes it as a control variable in education, employment, and wage functions whose parameters are estimated from the data. She finds that higher levels of severity reduce educational attainment and the likelihood of employment and in-

[13]The primary question is whether a subsample of persons with an illness of low prevalence, such as epilepsy, can be representative when it is drawn from a national sample that did not oversample the population to obtain epilepsy cases. The question can be resolved only by an analysis of the sample design.

[14]The sample weights are likely to vary widely between cases, but the information is not reported in the study.

[15]The lifetime estimates reported here are based on a 6% discount rate. The values are in 1995 dollars.

[16]There are overlapping categories. See Swingler et al. (15; page 117).

crease losses of potential wages. The hourly wage penalty for employed persons with epilepsy ranges from $2.17 for persons with less severe seizures to $3.98 for persons with more severe seizures. Because the data are restricted to one clinic, the results cannot be used to estimate the costs of epilepsy for larger populations.[17]

The lack of a representative sample of persons with epilepsy in the United States has forced researchers to attempt estimates of the costs of epilepsy for the nation by combining secondary data with expert opinions concerning the treatment of persons with epilepsy. Their approach, referred to here as *collage estimates*, has some merit for global estimates, but in the authors' opinion, the estimates are not appropriate measures of costs for CEA of competing methods of treatment.

Collage Estimates

The collage approach asks experts to describe the treatments that would be appropriate for hypothetical patients with characteristics thought to be representative of different groups of persons with epilepsy. The costs of care are then estimated by applying unit prices from a variety of secondary data sources to the assumed treatments, and then summing the costs over a hypothetical population of persons with epilepsy. The validity of the results depends on the validity of the assumptions and the degree of error introduced by the use of cost data from noncomparable sources. Those who would use the collage study estimates need to evaluate carefully the methods used to elicit expert opinions, the comparability of the unit cost data, and the assumptions concerning the relationship between the hypothetical patients and the populations they are assumed to represent. The information required for a reasonable evaluation of the validity of the cost estimates should be requested from the authors if it is not included in the published studies.

The best of the collage estimates is the study of Begley et al. (3) of a 1-year cohort of hypothetical patients with epilepsy. The study carefully describes its methods and the limits that apply to the interpretation of its results. The perspective is that of society, the reference year is 1990, and the duration is the predicted lifetimes of the cohort members.[18]

Expert opinion is used to define the health care required by six groups of hypothetical patients whose composition is derived from epidemiologic data. The patients are assumed to represent an incident population of 147,000 persons with onset of epilepsy in 1990. The costs of health care services and pharmaceuticals are obtained from several sources, ranging from annual provider surveys by the American Hospital Association to surgery charge data from the Baylor College of Medicine. The direct lifetime costs of care are estimated to equal an average of $11,328 per person, or approximately $1.6 billion for the incident population.

The effect of epilepsy on employment is estimated by extrapolating data for patients in Rochester, Minnesota, to the six groups. Unemployment rates for persons with epilepsy were assumed to be some multiple of national labor market rates. The wage rates used to value time lost from work were based on age-sex earnings profiles for the United States. Indirect costs, which included estimated losses of household productivity, are much larger than direct costs, totaling $18,543 per person, or $2.8 billion for the incident population. The indirect costs for persons who are institutionalized are excluded from the estimates because the authors thought it too difficult to distinguish the effects of epilepsy from the effects of coexisting conditions.[19]

The cost distributions are very skewed, with the average costs per patient for the relatively small number of patients having intractable and frequent seizures equal to more than thirty-five times the comparable costs for patients in remission after initial diagnosis. The skewness is an important consideration in the use of average costs from collage studies to evaluate the benefits of interventions, such as drug therapy. Average cost data from skewed distributions, as in this study, may not represent the average cost of epilepsy for either the low-cost patients or the patients with more severe forms of epilepsy.[20]

SUMMARY AND CONCLUSIONS

One of the most important characteristics of the estimates that have been reviewed is that none of the studies are comparable in terms of the selection of a sample of cases, the cost elements that are included, or the methods of estimation. Until studies adopt more comparable methods, the true costs of epilepsy must remain unknown, and the users of cost data for the evaluation of treatments and drugs will continue to face the risk of making inappropriate decisions because of flawed information on costs.

The studies of the costs of epilepsy differ so much in method and scope that comparisons of their estimates are not very useful. Each study does, however, increase our knowledge of the costs of epilepsy and helps define con-

[17]Professor Famulari is conducting a study of the indirect costs of epilepsy based on a sample of 1387 persons with epilepsy who are patients at one of 18 epilepsy centers in the United States. The results of the study, sponsored by the Epilepsy Foundation of America, will be available in 1997 (M. Famulari, *personal communication*).

[18]Present values are calculated using a real discount rate of 6%. See equation 1 in Begley et al. (3; pages 1239 and 1240 in the results section). No rationale is presented for the choice of a very high real discount rate.

[19]See Begley et al. (3; page 1242). The costs of care for institutionalized persons are, however, included in the estimates of direct costs.

[20]Professor Begley is directing a longitudinal study of the medical costs of epilepsy for the Epilepsy Foundation of America. The estimates of direct costs are to be combined with the estimates of indirect costs from Professor Famulari (described in footnote 17) to provide an estimate of the costs of epilepsy in 1995 (C.E. Begley, *personal communication*).

ditions that must be satisfied to obtain better estimates in the future. Several propositions are consistent with the results of most of the studies that have been reviewed, as follows:

1. The population of persons with epilepsy comprises a relatively large number of persons whose costs are relatively low and a relatively small group whose costs are high. The seizures of many persons who are included in the cost studies would not be classified by many epileptologists as epilepsy.

2. Many members of the high-cost group have serious illnesses in addition to epilepsy. Within this group, the highest costs are incurred for persons who have multiple conditions and who are institutionalized. In both situations, it is often impossible to separate the costs of epilepsy from the costs of coexisting conditions. Unless a separation is possible, the costs should not be attributed to the effects of epilepsy.

3. The indirect costs of epilepsy, of which wage losses are the most important part, are several times as large as the costs of health care. The importance of indirect costs implies that cost benefit or CEA of interventions, such as antiepileptic drugs or surgery, must include the effect of the intervention on employment and earnings or risk inappropriate conclusions.

The methodologic obstacles to the estimation of the costs of epilepsy are substantial, but the single most important barrier to a convincing estimate for the United States is the absence of a nationally representative sample of persons with epilepsy. Because persons with epilepsy are a relatively small part of the population of persons with disabilities, it seems unlikely that funding for such a survey will be obtained. The use of health insurance claims data may, in the interim, provide opportunities for much more extensive studies of the direct costs of epilepsy. The potential utility of health insurance claims data for the study of the costs of epilepsy has not been adequately explored.

REFERENCES

1. Baldwin ML, Johnson WG, Butler RJ. The error of using returns to work to measure the outcomes of health care. *Am J Ind Med* 1996;6: 63–641. •11•
2. Batavia AI. Health care reform and people with disabilities. *Health Aff* 1993; •12•
3. Begley CE, Annegers JF, Lairson DR, et al. Cost of epilepsy in the United States: a model based on incidence and prognosis. *Epilepsia* 1994:6:1230–1243.
4. Cockerell DC, Hart YM, Josemir WAS, et al. The cost of epilepsy in the United Kingdom: an estimation based on the results of two population-based studies. *Epilepsy Res* 1994;18:249–260.
5. Commission for the Control of Epilepsy and its Consequences. *Economic Cost of Epilepsy. Plan for Nationwide Action on Epilepsy.* Washington, DC: USDHEW publication No (NIH)78-279; 1978:117–148 (vol 4).
6. Deyo RA, Tsui-Wu Y. Functional disability due to back pain. *Arthritis Rheum* 1987:30:1247–1253.
7. Famulari M. The effects of a disability on labor market performance: the case of epilepsy. *South Econ J* 1992;1072–1087. •13•
8. Gold MR, Siegel JE, Russell LB, Weinstein MG. *Cost Effectiveness in Health and Medicine.* New York: Oxford University Press; 1996.
9. King EM, Smith JP. *Computing Economic Loss in Cases of Wrongful Death.* Rand: The Institute for Civil Justice; 1988. •14•
10. Mattson RH. Comparison of carbamazepine, phenobarbital, phenytoin, and primidone in partial and secondarily generalized tonic-clonic seizures. *N Engl J Med* 1985;3:145–151.
11. Murray M, Rentz A, Halpern M. *Cost of Illness of Epilepsy.* MEDTAP International Report prepared for Batelle Memorial Institute, to be transmitted to Centers for Disease Control and Prevention, November 30, 1995.
12. National Foundation for Brain Research. *The Costs of Disorders of the Brain.* Washington, DC, 1992.
13. Rodin E, Rennick P, Dennerell R, Lin Y. Vocational and educational problems of epileptic patients. *Epilepsia* 1972;13:149–160.
14. Smith GT. Epilepsy and health economics. In: Chadwick D, ed. *Quality of Life and Quality of Care in Epilepsy.* London: Royal Society of Medicine; 1990:32–37 (*Round Table Series* 23).
15. Swingler RJ, Davidson DLW, Roberts RC, et al. The cost of epilepsy in patients attending a specialist epilepsy service. *Seizure* 1994:3: 115–120.
16. Task Force on Principles for Economic Analysis of Health Care Technology. Economic analysis of health care technology. A report on principles. *Ann Intern Med* 1995;122:59–60.
17. Trupin L, Rice DP. Health status, medical care use, and number of disabling conditions in the United States. *Disability Statistics,* June 1995 (abst No 9).
18. Weiler PC, Hiatt HH, Newhouse JP, Johnson WG, Brennan TA, Leape LL. *A Measure of Malpractice.* Cambridge, MA: Harvard University Press; 1993.

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CHAPTER 286

AQ1 AU: Please add city and zip code for author Johnson and zip code for author Nuwer.
AQ2 AU: Please add date of communication with Louise Russell in footnote 1.
AQ3 AU: Butler 1995, Baldwin and Johnson 1995, and Baldwin and Johnson 1993 should be added to reference list and corresponding numbers inserted here ("This approach ignores the fact that the employment ...").
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AQ5 AU: Rodin et al. 1977 should be added to reference list and the corresponding number inserted here ("Among persons with epilepsy only, ...").
AQ6 AU: Johnson et al. 1993 should be added to reference list and corresponding number inserted in footnote 8.
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