A costs analysis of dental treatment for ectodermal dysplasia

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Ectodermal dysplasia (ED), a hereditary, clinically diverse, genetically heterogeneous group of conditions, is characterized by developmental defects in the tissues of the embryonic ectoderm and its appendages. More than 150 types of ED have been described.[1] They can be inherited through all Mendelian modes of transmission. The best known of the ED conditions—and one for which dental care frequently is the most important aspect of treatment—is the hypohidrotic X-linked form. Anodontia, or severe hypodontia, with conical (peg-shaped) anterior teeth usually is present. Dentures typically are needed at an early age and can be problematic because of poorly developed alveolar ridges.[2] Early dental intervention and continued treatment for many years are required to improve and maintain masticatory function and optimal facial appearance.

Dental treatment for ectodermal dysplasia had a marked financial impact on patients and their families.

The primary goals of dental treatment of patients with ED are enhancing esthetics and improving masticatory function. Optimal treatment typically requires several phases and the involvement of practitioners in several dental specialties to achieve optimal esthetics and function. These types of treatments are predicated on the severity and manifestations of hypodontia and concomitant problems such as malocclusion. Removable prostheses and bonding to reshaped teeth typically are provided while the child still is growing. Once growth is complete, intraosseous dental implant–supported prostheses are the treatment of choice.[3]

Although much has been written about the dental

ABSTRACT

Overview. Dental treatment modalities for ectodermal dysplasia (ED) vary markedly depending on the clinical manifestations, but to date there have been no studies exploring the potential economic impact of ED. On the basis of anecdotal and clinical reports, the authors postulate that costs of dental treatment for this condition can have a substantial financial impact on patients and their families.

Objective. The purpose of the authors' pilot study was to develop an economic model for various treatment modalities for ED with severe hypodontia.

Methods. The authors first used a comprehensive review of the literature and expert consensus to establish a treatment modalities model for ED. Next, they completed chart reviews to validate the model with sample treatment and costs information. Using these data, they then constructed a model of treatment options and associated costs.

Results. The sample included 24 patients with ED who had severe hypodontia. Forty-two percent were female; patients' ages ranged from 4 years, 11 months to 31 years, 1 month. Forty-two percent had dental insurance coverage, while more than one-half paid for services out of pocket. An estimated 84 percent had undergone prosthodontic treatment, 37 percent orthodontic treatment and 19 percent implant surgery. Depending on the age of the patient and types of dental treatment, there was a broad variation in costs. This ranged from $2,038 to $3,298 for those who had received prosthodontic treatment only; it ranged from $12,632 to $41,146 for those who had received a combination of prosthodontic, orthodontic and implant treatment.

Conclusions. Dental treatment for ED had a marked financial impact on patients and their families and varied depending on the type and duration of treatment.

Key Words. Ectodermal dysplasia; hypodontia; costs analysis.

treatment options for patients with ED,[2] no published studies report costs associated with these treatment modalities. Dental care is costly. The estimated annual bill for the restoration of U.S. children's teeth exceeds $2 billion, making dental disease one of the most expensive uncontrolled conditions of childhood.[4] Cost estimates for individual children's dental care based on a review of dental records in an academic setting in 1992 ranged from $170 to $2,212 per child.[5] In an analysis of 1996 Medical Expenditure Panel Survey data, the annual costs for dental care for children in the United States were estimated to be $12 billion.[6] The dollar figure translates into $375 per child, an amount that surpasses the annual national expenditures for treating common childhood respiratory conditions such as asthma.[7] Dental care costs are particularly problematic for children with special health care conditions and needs because they require continued care and incur additional medical costs. Almost one-half of these expenditures are paid out of pocket, and this makes access to adequate dental care difficult—if not impossible—owing to the considerable financial burden.[8]

Various dental treatment modalities for ED have been reported widely, but no studies to date have explored the potential economic impact of this condition on families. Therefore, we undertook an investigation to develop an econometric model that would describe accurately the dental costs from birth through early adulthood for a person affected by ED.

METHODS AND MATERIALS

Process of model development. Model development for this project was a multistep, iterative process of extracting data from the literature and integrating them with expert opinion. The first step in the modeling process was a comprehensive review of the literature regarding ED. The purpose of this review was threefold:

- to obtain sufficient information to develop a model of treatment options for patients with ED;
- to determine the parameters of each treatment modality for inclusion in the model;
- to determine the economic costs of each treatment option.

We used MEDLINE to identify all papers published from 1982 through 2002. Rather than integrating all published papers, such as those from the 1960s and 1970s, we focused on those of the last 20 years to obtain the most current treatment approaches for ED. We then developed a prototypic model. This model incorporated information from our literature review and used principles and techniques of clinical decision making and operations research.[9] After we formulated the initial model, we presented it to a pair of experts for review and consensus. Of particular interest was whether the model would accurately reflect the treatment modalities available and whether the parameters we chose from the literature review were acceptable. We next incorporated the comments from the expert consensus into the iterative process of model development. We developed several scenarios based on different clinical needs and treatment approaches and then estimated costs associated with these scenarios.

Expert input. Expert judgment involving synthesis approaches to estimate probabilities, costs, preference weights and other variables is used often in cost-effectiveness studies.[10] For this study, we called on two experts (A.G. and J.T.W.) to confirm treatment model estimations and estimate values that could not be obtained from the literature review. These experts have extensive experience in treating patients with ED.

Sources of parameters. We derived parameter values from several sources. The literature review during the initial stages of the project yielded the data on diagnosis and treatment options. These were supplemented with information from our experts. The costs of treatment were based on actual charges according to the dental faculty practice of the University of North Carolina (UNC) at Chapel Hill. These fees have been used by government agencies as the usual, customary and reasonable rate.[11] We validated and/or supplemented these data as needed by applying the econometric model to a sample of patients. We defined the total charges for these sample patients as all charges incurred by the patients from treatment. For each treatment choice, we hypothesized that the model would be validated by a patient who had ED and was treated with that modality.

We examined the costs of treatment from the perspective of the health care system rather than from that of society as a whole. Accordingly, we examined direct costs of dental care for treatment of ED. Most patients affected by ED required repeated care that was included in the models, but we did not include costs for pain and suffering that might have occurred as a result of the condition. Because we defined costs from the perspec-