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Acceptance of epilepsy surgery in the pediatric age — What the parents think and what the doctors can do

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ABSTRACT

Epilepsy surgery (ES) in pediatrics is safe and effective but can be underutilized. Possible barriers could be parental resistance and doctor inertia. We surveyed 138 parents of pediatric patients with epilepsy and found that 25.2% were opposed to this treatment. However, upon completing the questionnaire that contained factual information about ES, 50.4% of the responders stated that they had become more favorable vs. 3.3% more contrary and 46.3% unchanged. Parents of prepubescent patients were most receptive ($p = 0.0343$) and more likely to shift to a more favorable attitude. Thus, pediatric neurologists should not hesitate to discuss ES as soon as indicated, providing all necessary information to increase acceptance. However, among 60 child neurologists surveyed, 60% did not fully comply with guidelines or follow accepted standards of practice, indicating that they may not be apt to provide proper parental guidance.

We conclude that education of both practicing neurologists and parents is needed to facilitate the process.

1. Introduction

Epilepsy surgery (ES) has proved to be beneficial and safe for appropriate candidates with drug-resistant focal epilepsy among adults, especially patients with TLE [1–4]. However, it is well demonstrated that such treatment is underutilized [5–8]. Focal epilepsy in childhood differs from adult epilepsy because the etiology is often congenital and the underlying pathology is poorly definable and frequently extra temporal. A prospective study in a population-based sample has shown that approximately 10% of children with newly diagnosed epilepsy have seizures that become pharmacoresistant [9]. Among those are the potential candidates for surgery.

In children, it has been long known that resective surgery for large structural lesions can be successful [10,11]. However, reported outcome after resection of discrete dysplastic lesions, especially if extra temporal, has been more guarded [12–16]. A multinational survey during the calendar year 2004 showed that a variety of pediatric ES procedures, both resective and palliative, are being performed worldwide although the study made no attempts to evaluate outcome or how properly and how fully such treatment may be utilized [17].

Precise data regarding the utilization of ES in the pediatric age are lacking although there are good reasons to assume that it is underutilized, not only because type and extent of the underlying pathology are often different but also for ethical considerations and concerns about issues related to brain development [18]. As a result, the surgical approach in children has been more cautious. Nonetheless, over the past decade, a significant increase of ES in children has been reported in the US [19]. In addition, recent series have reported encouraging outcomes at two years [20] and five years after surgery [21] with a success rate for extratemporal resections comparable with TLE surgery. This is probably the result of greater clinical experience [21] and of more effective diagnostic tools allowing individually tailored resections [22]. Even incomplete surgical resections of the underlying pathology have not precluded a seizure-free outcome [23]. Thus, surgery in refractory pediatric cases should no longer be regarded as "last resort" [24]. Furthermore, when resective surgery may not be feasible, other palliative surgical approaches can be considered such as callosotomy [25]. Nonetheless, the extent of the resection, the choice among alternative treatments, and the timing of the intervention remain areas of debate.

The burden of deciding on the best approach and how soon surgery should be considered is shared by the treating physicians and the parents of young patients with drug-refractory epilepsy. Increasing evidence that epilepsy surgery, when handled properly, can be as

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successful in children as it has been in adults may add urgency to the need of determining, as soon as possible, whether surgery may be an option and worthy of the risk.

Two recent surveys indicate that acceptance of ES among the adult population is not only contingent on the attitude and determination of the neurologists [26] but also on the preference of the patients [27]. Surprisingly, little is known about the inclinations, beliefs, and motivations of parents of children with epilepsy when facing the dilemma of ES. Searching the literature, we found only one report documenting a high degree of satisfaction among parents of children with tuberous sclerosis who had previously undergone surgical resections [28].

This work is an attempt to clarify how "ready" parents of children with epilepsy may be to consider and accept an unfamiliar, risky treatment such as ES and how the doctors prepare them for such a decision. The aim of this study was twofold: first, to assess parental attitudes toward ES, irrespective of the child's epilepsy syndrome, of the frequency and severity of seizures and regardless of whether ES may be indicated or not and to evaluate possible determining factors of such attitude; and second, to assess how pediatric neurologists approach the issue of ES because their attitude may influence parental choice and affect patients' care. Our hope is that a better understanding of parent–doctor interactions may lead to better management, better results, and higher satisfaction.

2. Methods

We distributed ad hoc questionnaires to parents of children with epilepsy scheduled for regular follow-up appointments at 6 pediatric epilepsy centers located in Lombardy, Northern Italy. Three centers were located in Milan: Istituto "C. Besta"; A.O. Fatebenefratelli e Oftalmico; and A.O. San Paolo, and three were located in the surrounding region: Istituto "C. Mondino", Pavia; A.O. Spedali Civili, Brescia; and A.O. "C. Poma", Mantova. The participants were consecutive subjects screened according to their ability to respond to the questionnaire and willingness to sign the informed consent. Inclusion criteria required all parents to be older than 21, to have a minimum of 5th grade education/reading skills, and to be free of any obvious or suspected cognitive impairment. A child should be 18 years of age or younger, should have a well-established diagnosis of epilepsy, and should be on at least one antiepileptic drug (AED). Our intent was to survey cross-sectionally an unselected population of parents. Therefore, the questionnaire was offered to all qualified comers, as they randomly presented to the clinic. To minimize opinion bias, we included patients whose seizures were responding to treatment as well as those whose seizures were not and excluded all patients who had had ES. However, we collected data to identify patients whose seizures were in remission and those whose seizures were pharmacoresistant.

The clinic's staff in charge of recruitment explained to all potential participants the rationale of the survey assuring that participation was entirely voluntary and personal information strictly confidential. After signing the consent, if both parents were present, each one was invited to fill out the questionnaire, independently from the other.

2.1. The development of group-specific questionnaires

No validated instrument to test attitudes toward brain surgery was available at the time of this survey. The "ad hoc" questionnaire used for investigating patient acceptance of ES was built on material collected in focus groups organized among individuals variously affected by the disease: adult patients, their close relatives, adolescent patients, and parents of pediatric patients, adolescents or younger. Under the guidance of a moderator, group participants discussed issues concerning self-esteem, coping mechanisms, and QOL, and described their reaction when they first heard about ES, their perception of the associated risks, and the importance of obtaining more information about this treatment. The questionnaire's "domains" reflected the main areas of interest and concern transpired in these encounters. The result was four distinct questionnaires with similar core structure and considerable overlap, but each adapted to the specific needs and expectations of the group.

2.2. The questionnaire for parents

This consisted of two blocks. The treating physicians filled the first block, specifying personal identifiers and clinical data of the child with epilepsy. The parents filled the second block, which consisted of two parts. The first included demographics: personal and psychosocial information of both parent and child as well as the number of specialists and centers consulted. The second part was the questionnaire proper. This consisted of 46 questions stratified in two sections. Questions 1 to 19 collected data on how the parent was coping with the disease. Questions 20 to 46 addressed issues pertaining to ES. Among those, 5 questions asked if responders were aware of specific facts about resective ES. They were: "*Do you know that medications may stop the seizures while surgery tries to remove the cause of epilepsy?*" (Q. 30); "*Do you know that surgery is only for those who have a well defined brain abnormality?*" (Q. 31); "*Did the neurologist tell you that a long series of tests is necessary before surgery?*" (Q. 32); "*Do you know that this type of operation has been done in specialized centers for many years and that is not experimental?*" (Q. 34); "*Do you know that epilepsy surgery is done according to strict rules and removes only the part of the brain that causes the seizures?*" (Q. 35). Each of these "informative" questions provided clues that could influence, in a positive or negative way, the attitude toward ES of the responders, regardless of whether they were already aware of ES or naïve to the subject. To simplify the task of the responders, considering that "resective" ES is most frequently performed and the risk-benefit ratio of this procedure is better known, we made no mention of "disconnection" surgery or other palliative treatments in this setting. The effect of providing additional information was assessed in question 46, asking whether, upon completing the questionnaire, the responder's attitude toward ES had changed and how. That represented the participant's "final attitude" (more favorable/more contrary/already favorable/still contrary).

Questions were grouped according to their content in various domains, listed in Table 3. The response to one key question within each domain was chosen to correlate domains with final attitude. The process is described in detail in the previous study [27] where adult patients were investigated using the questionnaire *for adults* that contained 15 domains.

The English translation of the original Italian questionnaires *for parents* and the responses to each question are available online as Supplementary material (Table S1).

To evaluate how child neurologists approach ES and how they manage potential surgical candidates, we extracted the data pertaining to child neurologists from a previous study that surveyed practicing neurologists treating patients with epilepsy of all ages [26]. As part of that investigation, we also asked a group of leaders in the field, selected among the stakeholders of the most advanced epilepsy programs in the region, to independently respond to the same questionnaire *for physicians* (see below). Based on their experience and prominent position, we assumed that these individuals were "opinion leaders" and, thus, their responses represented the "best" current practice, as opposed to the common practice expressed by other practicing neurologists.

The responses provided by the child neurologists were then compared with the responses of the adult neurologists and of the "opinion leaders".

2.3. The questionnaire for physicians

The construct of the instrument used for surveying physicians was based on the *theory of planned behavior*, an approach that has proved