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Original article

Epilepsy surgery in children with accompanying impairments

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ABSTRACT

The aim of this study was to assess seizure outcome 2 years after epilepsy surgery in a consecutive series of paediatric patients, with special focus on children with learning disabilities and other neuroimpairments in addition to the epilepsy.

Outcome 2 years after surgery was assessed in 110 of 125 children operated upon for drug resistant epilepsy in Gothenburg 1987–2006.

More than half of the children had learning disabilities, 43% motor impairments and 30% a neuropsychiatric diagnosis. Fifty-six per cent of those with an IQ < 70 became seizure-free or had a >75% reduction in seizure frequency, and two thirds if the operation was a resection. The corresponding figure in those with more than 100 seizures per month was 15 out of 31, and another seven had a 50–75% reduction in seizure frequency.

The message is that learning disability, motor impairment and psychiatric morbidity should not be contraindications for paediatric epilepsy surgery. More than half of the children with learning disabilities had a worthwhile seizure outcome, with even better results after resective surgery. Children with drug resistant epilepsy and additional severe neurological impairments should have the benefit of referral to a tertiary centre for evaluation for epilepsy surgery.

1. Introduction

Children with medically refractory epilepsy constitute a heterogeneous group and the majority have other impairments in addition to epilepsy, such as learning disabilities, cerebral palsy and autism. For a long time, learning disabilities constituted a relative contraindication for epilepsy surgery, whereas now several reports support that children with learning disabilities and severe epilepsy may benefit from surgery and should be included.[1,2] Seizure outcome in children who undergo epilepsy surgery is considered good; the overall seizure-free outcome is reported to be about 70%.[3,4] Epilepsy surgery leading to the cessation of seizures might even prevent cognitive deterioration.[5] It is an option which

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should be considered when it becomes obvious that the third line drugs fail to be efficient or even earlier if there is a suspicion of a focal start in early onset epilepsy in children.[1,6]

The purpose of this study was to assess seizure outcome 2 years after epilepsy surgery in a consecutive series of paediatric patients, with special focus on children with learning disabilities or other impairments in addition to the epilepsy.

2. Material and methods

Children accepted for epilepsy surgery at Sahlgrenska University Hospital are assessed by a multidisciplinary team with an extensive evaluation before and 2 years after surgery. From 1995, data have been recorded prospectively, according to a national protocol, which includes information about social situation, epilepsy history, seizure type and frequency during the year preceding the presurgical investigations, antiepileptic drugs, neurological impairments, the results from the preoperative investigations, type of operation, histopathological diagnoses, complications during the presurgical evaluation or at surgery, and a 2-year follow-up of seizure outcome, antiepileptic medication and psychosocial data. A comprehensive neuropsychological assessment is part of the preoperative set-up and repeated at the 2-year follow-up. Since 2002, a neuropsychiatric assessment is included pre- and postoperatively.

A consecutive series of 125 children 18 years and younger underwent epilepsy surgery in Gothenburg during the 20-year period 1987–2006. A 2-year follow-up after the first operation was possible in 110 children, including one boy with a reoperation as a staging procedure ending with a hemispherectomy. One girl was lost to follow-up, one girl died from sudden unexpected death (SUDEP) within 2 years after surgery, and 13 children had a reoperation before 2 years had elapsed. Nine underwent a second operation after the 2-year follow-up. Three of the 22 individuals who had a second operation were adults by then and therefore not followed up within the paediatric programme. A third operation was performed in four children. The outcomes 2 years after the last reoperations are reported separately. A total of 153 procedures were performed in the 125 individuals.

The preoperative cognitive level was categorised as normal (IQ 70 and more), mild learning disability (IQ 50–69) and severe learning disability (IQ less than 50), as assessed by a neuropsychologist, using the Wechsler Scales or Griffiths Developmental Scales, or in eleven cases based on clinical assessment and school data. The occurrence of autism spectrum disorder (ASD), attention deficit/hyperactivity disorder (ADHD) and attention deficit disorder (ADD) was recorded (in all, 43 children had been seen by a neuropsychiatrist). The clinical neuropsychiatric assessment included an in-depth interview. Psychopathology was diagnosed according to DSM-IV criteria.[7] The assessment of children with ASD also included further multidisciplinary work-up using the Diagnostic Interview for Social and Communication Disorders (DISCO)[8] and the Autism Diagnostic Observation Schedule-Generic (ADOS-G).[9]

Motor function was assessed and categorised as cerebral palsy, other motor impairment or normal.

Seizure-free patients include those with aura only, those with a few initial seizures but subsequently seizure-free for 2 years, and those who have had atypical seizures related to withdrawal of antiepileptic treatment, i.e. Engel category Ia–d.[10] For those with seizures after surgery, seizure outcome was defined as the mean monthly seizure frequency during the year preceding the 2-year follow-up, i.e. the second postoperative year. The relative reduction in mean seizure frequency at the 2-year follow-up as compared with preoperatively was calculated and categorised as >75% reduction, 50–75% reduction, <50% reduction and increase in seizure frequency. Seizure freedom or more than a 75% reduction in seizure frequency was considered a worthwhile seizure outcome.

The histopathological diagnoses were categorised into four groups: focal lesions (ganglioglioma, dysembryoplastic neuroepithelial tumour (DNET), low-grade astrocytoma and cavernoma); malformations of cortical development (MCD), including major malformations and microdysgenesis, defined according to earlier published criteria[11]; gliosis, including mesial sclerosis; and other diagnoses, e.g. tuberous sclerosis and Sturge-Weber syndrome. Multiple pathology was recorded, and cases classified as focal lesions or gliosis could also have MCD.

The results are described by means of frequencies and percentages. The frequency distributions of seizure outcome are shown by bar charts.

According to the Swedish National Board of Health and Welfare, clinicians are obliged to secure the quality of care by performing and reporting results of clinical studies in everyday practice. Approval from an internal review board is not required for this type of research. All participants or caregivers gave their informed consent to participate in this follow-up. Participants and all data have been handled according to the Helsinki convention.

3. Results

There were 53 boys and 57 girls; the median age at surgery was 10 years 8 months (range 11 months–18 years 7 months). The median duration of epilepsy was 6 years (range 2 months–16 years 5 months). Forty-three children (39%) had a preoperative seizure frequency of more than 100 seizures per month. The preoperative IQ level was <50 in 37 (34%), 50–69 in 22 (20%), and 70 and more in 51 (46%). Thirty-two (29%) had a diagnosis of cerebral palsy and 15 (14%) other kinds of motor impairments. ASD had been diagnosed in 20 (18%) and ADHD/ADD in another 13 (12%). The surgical procedure was resection in 92 cases, callosotomy in 14, disconnection of hypothalamic hamartoma in two and multiple subpial transection in two. The surgical procedure was considered multilobal if more than one lobe was included in the resection. Table 1 shows the baseline data of the 110 children.

3.1. *Seizure outcome after resections (n = 92)*

Fifty girls and 42 boys underwent resection. Their median age at operation was 10 years 9 months (range 11 months–18 years 7 months). Seizure outcome after resection is shown in