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Somatosensory evoked potentials are abnormal with plagiocephaly

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Background: Deformational plagiocephaly is usually managed conservatively, as it tends to improve over time and with the use of conservative measures. However, before the year 2017 we operated on patients with severe plagiocephaly and neurological symptoms at the Helsinki Cleft Palate and Craniofacial Center.

Methods: Of the 20 infants with severe deformational plagiocephaly and neurological symptoms referred to us between 2014 and 2016, 10 underwent cranioplasty open reshaping of the posterior cranial vault. The parents of the last 10 patients were given information on the natural history of the condition and the patients were followed up with an outpatient protocol. The aim of this study was to gain information on the brain electrophysiology and recovery of patients after total cranial vault reconstruction by measuring the electroencephalogram (EEG) somatosensory evoked potentials (SEP; median nerve).

Results: Of the 10 participants in the operation arm, six had abnormal SEP at least on the affected cerebral hemisphere and all SEPs were recorded as normal when controlled postoperatively. In the follow-up arm, eight out of 10 participants had abnormal SEP at the age of approximately 24 months, and all had normalized SEPs at control visits.

Conclusion: Our data suggest that cranioplasty open reshaping of the posterior cranial vault did not affect abnormal SEP-EEG recordings. We have abandoned the operations in deformational plagiocephaly patients due to findings suggesting that expanding cranioplasty is not beneficial for brain function in this patient group.

Abbreviations: CT, computer tomography; EPs, evoked potentials; ERPs, event-related potentials; SEP-EEG, somatosensory evoked potentials electroencephalogram

Keywords: Craniotomy / Electroencephalogram / Plagiocephaly, nonsynostotic

INTRODUCTION

Deformational plagiocephaly, also known as posterior positional plagiocephaly or flat head syndrome, is attributed to deformational forces during craniofacial growth and maturation. After the launch of the “back to sleep” campaign in 1992 to reduce sudden infant death syndrome, the reported incidence of plagiocephaly increased from 2% to 20%–48% [1]. Deformational plagiocephaly develops after birth, with initially unremarkable head shape. Flattening of the skull becomes progressively worse if no alterations in sleeping position are applied [2–4].

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Recording of event-related/evoked potentials (ERPs and EPs) is a traditional noninvasive method used to measure neural responses evoked by external stimuli such as sensory, visual, or auditory events [5]. Because recording of ERPs and EPs have a high capacity for temporal resolution and does not require a behavioral response from the subject, this technique is particularly well suited for neuropsychological study of early deviations from typical development in small children. Neurophysiological study methods, particularly somatosensory evoked potentials (SEPs), are useful in determining the integrity and functionality of the somatosensory pathway and cortex in order to predict clinical outcomes after perinatal asphyxia [6]. Recent studies show that electrophysiological brain recordings are a promising method for monitoring brain development in children with cranial disorders [7].

The aim of this cohort study was to report our experience of brain electrophysiology and recovery of deformational plagiocephaly patients after total cranial vault reconstruction by measuring the electroencephalogram (EEG) and median nerve SEPs.

METHODS

The study protocol was approved by the Institutional Committee of Helsinki University Hospital. The tenets of the Declaration of Helsinki were followed.

From 2012 to 2014, altogether 20 children with clinically severe deformational plagiocephaly and neurological delay were referred to the Cleft Palate and Craniofacial Centre, Helsinki University Hospital and University of Helsinki, Finland. The diagnosis of deformational plagiocephaly was established by clinical examination and computer tomography (CT) scan to rule out synostosis. All 20 infants with neurological symptoms, including delayed motor skills and speech, were sent for further investigations with EEG and SEPs.

Of these 20 infants, 10 with severe deformational plagiocephaly and neurological symptoms underwent cranioplasty open reshaping of the posterior cranial vault between 2014 and 2016. The decision to operate was always made after a detailed discussion with the parents about operative risks and prognosis. The parents of the last 10 patients were given information on the natural history of condition and the patients were followed up with an outpatient protocol.

In this retrospective study, we reviewed the patient records and SEP-EEG recordings of these 20 patients and evaluated the severity of plagiocephaly from CT scans (Argenta clinical classification of deformational plagiocephaly: type 1-5, as described by Branch et al. [8]). The Argenta classification is a clinical

5-point scale for unilateral deformational plagiocephaly and a 3-point scale for central deformational plagiocephaly.

EEG somatosensory evoked potentials

EEG SEPs were evaluated in these patients first at the age of 14–42 months, and if abnormal, were controlled 12–18 months later. SEPs were recorded in a polyclinic EEG using our in-house developed routine where EEG and SEPs are recorded simultaneously, as described in detail previously [5]. The latencies and interpeak latencies of the patients were compared with normal values by Doria-Lamba et al. [9]; latencies were categorized as abnormal if the value was outside the normal range defined as mean+2.5 standard deviation. SEPs were individually categorized into normal and abnormal, with the responses classified separately for the left hemisphere (right-sided median nerve stimulation) and the right hemisphere (left-sided median nerve stimulation). The responses were categorized as abnormal if the latency of the first cortical response (N20) was delayed or if the central conduction time (measuring the delay between N13 and N20 responses) was abnormally prolonged.

Statistical analysis

Fisher exact test was used for statistical analysis to compare the two groups. Values of $p < 0.05$ were considered to be significant.

Table 1. Characteristics of children with and without operative treatment for deformational plagiocephaly

Characteristics	Follow-up, No.	Operative, No.
All patients	10	10
Sex		
Girls	3	2
Boys	7	8
Age at SEP-EEG testing (mo), mean	25.9	25.0
Child delivered < 38 wk		
No	8	6
Yes	2	4
Order of birth: twin		
No	8	7
Yes	2	3
Severity of plagiocephaly, mean ^{a)}	2.2	3.5
Type I	1	0
Type II	5	1
Type III	3	3
Type IV	1	6
Type V	0	0

SEP-EEG, somatosensory evoked potentials electroencephalogram.

^{a)}Argenta classification.

RESULTS

Demographic data of 20 deformational plagiocephaly patients are provided in Table 1. Standard awake and sleep EEG findings were normal in most patients. In two patients, the basic EEG recording revealed a slight slowing of the posterior dominant rhythm (Table 2, patient nos. 3 and 8). Of these patients, 10 were surgically treated by cranioplasty, and all of the parents agreed to the operative plan after a discussion with the surgeon. Of the 10 participants in the operation arm, six had abnormal SEP at least on the affected cerebral hemisphere, and all SEPs were recorded as normal when controlled postoperatively. No postoperative complications were recorded. In the follow-up arm, 8/10 participants had abnormal SEP at the age of approximately 24 months, and all had normalized SEPs at control visits (Tables 2). The difference between the numbers of participants with abnormal SEPs in these two groups was not significant ($p < 0.63$). The mean Argenta stratification type for operative patients was 3.5 and in the follow-up arm 2.2 (Table 1).

DISCUSSION

The management of deformational plagiocephaly is usually

conservative, as it tends to improve over time and with the use of conservative measures [3,4,10-14]. Typical methods are based on positioning the child so that he/she is lured to turn the head to the non-flat side and laying the baby on the stomach for brief periods while awake. Also, physiotherapy and home exercises are often recommended in cases with congenital or acquired torticollis [15]. Irrespective of any intervention, the prevalence of deformational plagiocephaly demonstrates a significant drop over time in most patients. By 2 years of life, the prevalence decreases to about 3% [15]. Thus, in most centers surgical correction of the skull is not a method of choice in the management of deformational plagiocephaly [16].

The developmental implications of deformational plagiocephaly are poorly understood. There is an unclear association between deformational plagiocephaly and developmental delay. Miller and Clarren [17] reported that children with persistent plagiocephaly use more school-associated special help programs than their healthy siblings. Panchal et al. [18] showed mild delays in cognitive and psychomotor development associated with either deformational plagiocephaly or craniosynostosis. Collett et al. [19] followed individuals with deformational plagiocephaly up to 36 months, finding lower Bayley Scales of Infant Development scores in cognition, language, parent-reported adaptive behavior, and motor development. Posterior cranioplasty and an open reshaping of the posterior cranial vault have been applied in severe cases with neurological symptoms after obtaining informed consent from parents until the year 2017 in our center.

Here, we reviewed our experience with the operative treatment of 10 deformational plagiocephaly patients, with a control group of 10 conservatively treated deformational plagiocephaly patients and a special focus on SEP-EEG measurements.

Six of the 10 patients in the surgical group had abnormal SEPs (6/6 on the affected cerebral hemisphere), and all SEPs were recorded as normal when controlled postoperatively. In the control group, eight out of 10 patients had abnormal SEPs at the age of about 18 months, and all had normalized later.

SEP normalized in both operated and non-operated groups. Our data suggest that cranioplasty open reshaping of the posterior cranial vault did not affect abnormal SEP-EEG recordings. Based on our results, surgery has no impact on neurocognitive function in deformational plagiocephaly. However, these surgeries carry many risks and must be justified when performed.

The optimal management of deformational plagiocephaly remains a topic of considerable ongoing debate. The lack of randomized control trials on the operative treatment of deformational plagiocephaly is widely acknowledged [20,21]. The current conservative treatments address aesthetics and skull shape

Table 2. All patients and findings

	Patient No.	Argenta type	Affected side	SEP I	SEP II
Follow-up	1	2	Right	Abnormal bilateral	Normal
	2	3	Right	Abnormal right	Normal
	3	4	Left	Abnormal bilateral	Normal
	4	2	Right	Normal	NA
	5	2	Right	Abnormal right	Normal
	6	2	Right	Abnormal right	Normal
	7	2	Right	Abnormal bilateral	Normal
	8	1	Bilateral	Abnormal bilateral	NA
	9	3	Left	Normal	NA
	10	3	Right	Abnormal bilateral	Normal
Operative	11	4	Right	Abnormal right	Normal
	12	3	Right	Abnormal right	Normal
	13	4	Right	Abnormal right	Normal
	14	3	Right	Normal	NA
	15	4	Right	Abnormal right	Normal
	16	4	Left	Normal	NA
	17	4	Left	Normal	NA
	18	2	Right	Normal	NA
	19	4	Right	Abnormal bilateral	NA
	20	3	Right	Abnormal bilateral	NA

SEP, somatosensory evoked potential; NA, not applicable.

more than neurological function. Further, controversy exists regarding the neurological implications of positional plagiocephaly. Collett et al. [22] noted that infants with deformational plagiocephaly show differences in brain shape, consistent with the skull deformity characteristic of this condition. Their findings suggest that infant brains with deformational plagiocephaly take the shape of an asymmetric and compressed skull. A critical issue is whether these effects have functional consequences. A large study of 187 patients by the Collett group was published in 2019 showing that school-aged children with moderate to severe deformational plagiocephaly had lower scores on cognitive and academic measures, but causality remains hard to prove [23].

Our basis for operative treatment was severe deformational plagiocephaly with developmental concerns and/or neurological symptoms and intracranial impressions in CT scan. Further, we based our operative decision-making on the assumption that the physical deformity in deformational plagiocephaly patients might correlate with some degree of neurological deficit, as Yang et al. [24] showed for metopic synostosis patients. The causative reasons and mechanisms leading to this are unknown. The core question “does cranial vault surgery contribute to normalized brain development and improve cognitive performance later in life” remains to be answered.

There are several limitations to our study that warrant consideration. This is a retrospective cohort study with a relatively small patient group. We acknowledge selection bias; decision to operate was not based purely on severity of deformational plagiocephaly or neurological symptoms. We do not know whether abnormal SEPs reflect a true risk for neurological function or whether they merely reflect abnormal shaping of the skull *per se*. The SEP technique is based on positioning the EEG electrodes on predefined places on the skull, assuming that the brain area underneath is constant across subjects. The latencies and amplitudes of the responses differ depending on the exact location at which they have been measured; thus, if the part of the brain beneath the electrode in patients with deformational plagiocephaly differs from that of the normal population, the SEPs may be affected.

To conclude, the main finding is that abnormal SEP-EEGs became normal in both operatively and conservatively treated patient groups over time. We have abandoned the operations in deformational plagiocephaly patients due to findings suggesting that expanding cranioplasty is not beneficial for brain function in this patient group.

NOTES

Conflict of interest

No potential conflict of interest relevant to this article was reported.

Ethical approval

The study was approved by the Institutional Committee of Helsinki University Hospital (approval No. 2284) and performed in accordance with the principles of the Declaration of Helsinki. The informed consent was waived because this study design is a retrospective chart review.

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