Original Articles

Auditory ERPs Reveal Brain Dysfunction in Infants With Plagiocephaly

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It is suspected that the developmental delay in school-aged children diagnosed as infants suffering from plagiocephaly is caused by the modification of the skull form. To detect possible cognitive impairment in these children, we examined auditory ERPs to tones in infant patients. The infants with plagiocephaly exhibited smaller amplitudes of the P150 and the N250 responses to tones than healthy controls. Differences between the patients and control subjects indicate that already at this early age the presence of the plagiocephalic skull signals compromise of brain functioning. The present data suggest that most of the plagiocephalic infants have an elevated risk of auditory processing disorders. In the current study we demonstrated, for the first time, that the central sound processing, as reflected by ERPs, is affected in children with plagiocephaly.

Key Words: Plagiocephaly, event-related potentials (ERPs), brain dysfunction, infants, central auditory processing, P150, N250

Plagiocephaly is a descriptive term defined by Virchow in 1851 that connotes an asymmetrically oblique or twisted head. Plagiocephaly is etiologically and pathogenically heterogeneous. Well-known types include synostotic anterior and posterior plagiocephaly (unilateral coronal and lambdoid synostosis, respectively), and the deformational or positional forms of plagiocephaly where the sutures are patent.[1] In synostotic plagiocephaly, unilateral coronal craniosynostosis results in more severe cranial distortion than that found with unilateral lambdoid synostosis, since more growth occurs at the coronal than at the lambdoid suture. In general, the severity of plagiocephaly depends on the developmental phase at which synostosis occurs, being more severe the earlier it occurs. Non-synostotic (deformational) form of plagiocephaly, either anterior or posterior, can be caused by a number of factors in utero. Factors such as hypotonia, fetal positioning, and prematurity can produce asymmetric flattening of the occiput that becomes favored by infants sleeping on their backs, exaggerating the plagiocephaly.[2]

The clinical and imaging features of true lambdoid synostosis versus those of deformational plagiocephaly are inadequately described in the literature and poorly understood.[3] The difficulty in diagnosing posterior plagiocephaly is partly the result of confusion in the medical literature concerning the true characteristics of the lambdoid synostosis. Posterior plagiocephaly in the absence of the usual characteristics of suture fusion in operative and histopathological examination has been called the "functional" lambdoid synostosis.[4] The findings in patients with functional lambdoid synostosis were considered typical of lambdoid synostosis, although synostosis was not confirmed. According to Huang and Posnick[3,5], the features of positional plagiocephaly are clearly distinguishable from those of lambdoid synostosis on physical examination. True lambdoid synostosis is actually quite rare, constituting only 3 to 5% of all cases of craniosynostosis.

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Whether or not to perform surgical correction on a child with anterior plagiocephaly seldom poses a problem especially in cases where true coronary synostosis is present. The resulting deformity is usually clearly visible and readily diagnosed, demanding surgical correction on aesthetic basis alone. On the contrary, the diagnosis and treatment of posterior plagiocephaly is one of the most controversial aspects of craniofacial surgery. It is generally accepted that most cases of posterior plagiocephaly occurring due to positional molding can usually be managed nonsurgically by conservative positional measures or by using a molding helmet.[6,7] Some authors[8] mention that physiotherapy in an early stage, directed toward plagiocephaly and associated disorders, results in a complete disappearance of symptoms in a high percentage of the cases.

Due to the lack of aesthetic need, this type of deformity may become neglected especially in cases where the unilateral posterior flatness is relatively mild. The decision of surgical correction is usually reserved for cases with true lambdoid synostosis or, in the absence of obvious synostosis, for patients with the most severe form of posterior flatness.

A few recent studies have pointed out a new perspective on plagiocephaly. These studies addressed an increased rate of developmental delay in school-aged children presented as infants with deformational plagiocephaly without obvious signs of delay at the time of the initial evaluation.[9,10] Miller et al.[9] mentioned that 25 out of 63 (40%) children with persistent deformational plagiocephaly had received special help in primary school including special education assistance, physical therapy, occupational therapy, and speech therapy. Only 7 of 91 siblings (8%), serving as controls, required similar assistance.[9] Another study[10] showed that all 12 children diagnosed as plagiocephalic had auditory processing and language disorders. Furthermore, some of these children had attention deficit disorder or attention deficit with hyperactivity disorder. These results are at odds with those suggesting that no functional neurological sequelae can occur subsequent to the skull molding and that, consequently, early surgical intervention for patients with plagiocephaly is not necessary or desirable.[3,11]

The use of electrophysiological measures, such as event-related potentials (ERPs), has gained increased popularity in predicting the developmental outcome of infants. Event-related potentials are elicited, among other things, by external stimuli and appear as small voltage changes in the ongoing electrical brain activity (EEG). They provide a direct measure of neural information processing and can be non-invasively and safely recorded from infants starting from birth. The ERPs are extracted by averaging the EEG across multiple presentations of the stimuli. The ERP curve is composed of a number of peaks and troughs labeled according to their polarity and latency. In primary-school-aged children, the ERP consists of the P100-N250-N450 peaks.[12] The maturation of the ERP components during the first year of life is characterized by the shortening of the peak latencies and the increase of the amplitudes[13] commonly related to advanced myelination, increase in synaptic density, efficacy, and synchronization. Our recent longitudinal study[14] has shown that the precursors of all peaks observed at the age of 1 year were identifiable already at birth and, by the age of 1 year, attained the morphology (P150-N250-N450) that is seen during the next 10 years of life.

Several recent studies indicate the possibility of predicting the developmental outcome on the basis of the ERPs recorded at birth. Molfese[15] and Molfese[16] reported that on the basis of the latencies and amplitudes of the neonatal N250 and N450 peaks, the verbal performance at 5 and 8 years of age can be efficiently predicted. Furthermore, Deregnier et al.[17] found a significant correlation of the slow negative wave of the newborn ERP with the 1-year Mental Developmental Index. This wave was elicited in response to a stranger's voice as compared with the mother's voice and was attenuated in the infants of diabetic mothers who are at risk for fetal metabolic abnormalities that potentially damage the recognition memory pathways. The difference in ERPs between the healthy infants and those with oral clefts, another craniofacial dysmorphology, was demonstrated by Çeponiene et al.[18,19] The amplitude of the infantile P150/P350 was significantly larger in healthy infants than in those with oral clefts at the newborn age. At the age of 6 months, the responses of the infants with cleft lip and palate showed an abnormal ERP waveform lacking the negative peaks (N250–N450).

In the present article, we will investigate the feasibility of the ERPs to measure aspects of brain functioning in the plagiocephalic patients in determining whether the infants with plagiocephaly differ from normally developing infants.

Patients and Methods

Fifteen infant plagiocephalic patients were included in the study. Ten infants had deformational posterior plagiocephaly with open sutures, two had posterior plagiocephaly with lambdoid synostosis, and three had anterior plagiocephaly