Calcified cephalohematoma as an unusual cause of EEG anomalies: case report

Vera Vigo, MD,¹ Domenica Immacolata Battaglia, MD,² Paolo Frassanito, MD,¹ Gianpiero Tamburrini, MD,¹ Massimo Caldarelli, MD,¹ and Luca Massimi, MD, PhD¹

Departments of ¹Pediatric Neurosurgery and ²Pediatric Neurology, A. Gemelli Hospital, Rome, Italy

Cephalohematoma, one of the most common neonatal head injuries, generally undergoes spontaneous resorption. When calcified, it may cause cranial vault distortion and depression of the inner skull layer, although it remains asymptomatic. Surgery, indeed, is usually performed for cosmetic purposes. For these reasons, the long-term effects of calcified cephalohematoma (CC) are widely unknown.

The authors report the case of an 11-year-old girl with a persistent calcified CC causing skull deformity and delayed electroencephalography (EEG) anomalies. These anomalies were detected during routine control EEG and were not clinically evident. The young girl underwent surgical removal of the CC for cosmetic purpose. The EEG abnormalities disappeared after surgery, thus reinforcing the hypothesis of a correlation with the brain “compression” resulting from the CC. To the best of the authors’ knowledge this is the first time that CC-associated EEG anomalies have been described: even though these anomalies cannot be considered an indication for surgery, they merit late follow-up in case of skull deformity.

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KEY WORDS calcified cephalohematoma; cranial injury; newborn; electroencephalogram; trauma

CEPHALOHEMATOMA represents the most frequent cranial injury in the newborn, occurring in 0.2–2.5% live births.³⁰ It arises from the breaking of small pericranium veins during delivery (prolonged labor, and the use of forceps and a vacuum extractor) resulting in a collection of blood below the periosteum of the skull. In about 3%–5% of cases, such a blood collection fails to resorb, leading to a calcified cephalohematoma (CC).²⁹ Because of the flexible shape of the neonatal skull, when the hematoma expands and becomes sufficiently thick, it can encroach upon the cranial vault space.²⁹ Generally, the CC does not cause neurological sequelae.²⁰ Moreover, clinical sequelae, in contrast to esthetic effects, are rarely reported.²⁹ However, sagittal synostosis secondary to cephalohematoma has been described.^{11,15,16}

We report the case of an 11-year-old girl with a CC that caused late-onset cerebral electrical anomalies. To the best of our knowledge, this complication was not been described to date in the literature.

Case Report

This 11-year-old girl was born after a forceps-assisted vaginal delivery resulting in a bilateral parietal cephalohematomas. The left cephalohematoma gradually resorbed within the 1st month of life, whereas the right one became calcified; no operation was advised at that time. The young girl grew up without any relevant physical and psychomotor problems. However, she had some learning disabilities (writing and reading) associated with a mild attention deficit diagnosed based on neuropsychological testing. For this reason, she received serial electroencephalography (EEG) studies over time that did not show abnormalities. The last EEG study obtained during the follow-up (when the girl was 10 years old), however, showed posterior asymmetrical alpha activity in the background, with low amplitude on the right side and sporadic right frontoparietal shapes (Fig. 1). Therefore, brain MRI was performed that revealed a right parietal cephalohematoma (thickness

ABBREVIATIONS CC = calcified cephalohematoma; EEG = electroencephalography; TBI = traumatic brain injury.

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25 mm), with alteration of the inner layer of the skull and moderate brain parenchyma compression (Fig. 2). A new EEG study, performed 3 months later, showed the persistence of the same abnormalities of the previous study.

The patient was then admitted to our institution for neurosurgical treatment of the cephalohematoma, mainly for cosmetic purposes. At admission, she was in good clinical condition; physical examination did not indicate the presence of any neurological deficits. She complained of mild headache. We did observe a gross bony protuberance overlying the parietal area on the right side. After induction of general anesthesia (sevoflurane and remifentanyl), a circular craniotomy was made and the CC was removed. Bone remodeling of both the inner and the outer surface of the bone flap to thin it out was performed using high-speed drilling.

The patient's postoperative course was uneventful. The pathological report confirmed the diagnosis of CC (remodeled bone with calcified blood remnants). MRI showed the bone remodeling and the subsequent brain decompression (Fig. 3). EEG studies demonstrated the disappearance of the electric abnormalities in the right hemisphere 6 months after surgery (Fig. 4) and at the latest follow-up (12 months). No significant changes in the patient's learning disabilities and attention deficit were observed on neuropsychological tests performed 6 months after surgery.

Discussion

Cephalohematoma usually starts as a fluctuant mass

covering the parietal bone during the first 24–72 hours after the birth; it then becomes firm and tense and is completely resorbed from within 2–4 weeks to 3–4 months. In rare cases (3%–5%), it fails to resorb and induces a progressive sub-pericranial osteogenesis that results in a CC.^{21,29} What underlies the failure for the mass to resorb is not yet known.¹⁹ The calcification process begins when a ridge forms along the periphery of the hematoma, causing a distortion of the calvaria. Calcified cephalohematoma can be classified as Type 1 or 2, based on the contour of the inner lamella. In Type 1 CC, the inner bone is not involved and there is no encroachment into the cranial cavity. In contrast, Type 2 CC exhibits a depression of the inner bone that pushes into the cranial cavity. In spite of the encroachment occurring in Type 2, there are no reports on a CC causing focal neurological deficits or raised intracranial pressure,^{16,29} probably because its growth is slow enough to allow the nervous structures of the newborn to accommodate the mass.²⁹ Even if asymptomatic, the cephalohematoma can hide a skull fracture along with underlying epidural hematoma or intracranial hemorrhage, thus justifying the recommendation of transfontanelar ultrasound by some authors.^{14,22}

In a recent study, Swanson et al. reported on children presenting with nonsyndromic craniosynostosis who had a higher risk of birth trauma (both for mother and fetuses), such as subgaleal and subperiosteal perinatal bleeding.²⁴ In the past, some authors have argued that CC can cause scaphocephaly in some instances,^{5,16} leading to the premature closure of the sagittal suture, although at histological

FIG. 1. Preoperative EEG showing asymmetry of the background activity and sporadic spikes on the right side (arrow). Figure is available in color online only.

FIG. 2. Preoperative T2-weighted axial and coronal MR images showing the right CC with mass effect on the frontoparietal brain.

examination has not detected suture formation underlying the CC. Other authors have shown that the midline location of cephalohematoma occurs only in the case of sagittal craniosynostosis.^{11,17}

The long-term effects on the brain of CC encroaching into the cranial vault are unknown. A unique aspect of the present case is the occurrence of a possible late-onset neurological complication of the cephalohematoma. To the best of our knowledge, our report is the first to describe the long-term effects of CC, and the case shows that the long-lasting compression of the cephalohematoma on the dural layer and the cortex may cause abnormality of the brain activity on EEG. The surgical decompression was able to resolve such activity, although the follow-up dura-

FIG. 3. Postoperative T2-weighted axial and coronal imaging MR images, acquired 1 month after surgery, demonstrating the removal of the cephalohematoma and the mass effect resolution.

tion is still too short to consider this result as definitive. In our opinion, this is not enough to raise the question of treating all thick CCs, but it should suggest that untreated patients should be monitored with repeated EEG over the time. This kind of follow-up represents only a little change in the clinical practice but could be enough to detect early electric abnormalities resulting from brain irritation. Actually, EEG is usually easily manageable in children. On the other hand, more “invasive” investigations, like PET scanning or magnetoencephalography, can be used to refine the diagnostic assessment (especially in doubtful cases) and/or to look for possible changes after surgery.

FIG. 4. Postoperative EEG (6 months later the preoperative one) demonstrating the absence of the previous right anomalies (arrow). Figure is available in color online only.

A well-known consequence of traumatic brain injury (TBI) is posttraumatic seizure. Depending on their time of onset after TBI, seizures can be classified as immediate (less 24 hours from trauma), delayed early (within 1 week), and late (more than 1 week).¹ Many studies have been carried out to demonstrate the correlation between head injury and late posttraumatic epilepsy⁹ to settle on the timing of follow-up and treatment. After head injury, late posttraumatic seizures, both partial and generalized, can develop, usually from 1 week until 5 years.^{2,25} Information on this issue is generally available in cases of depressed skull fractures or growing fractures; in fact, some authors have reported that early seizures and depressed skull fractures are the main risk factors for late posttraumatic epilepsy.^{1,25} Cases of delayed epilepsy (after more than 5 years from TBI) have been reported as a possible consequence of a growing skull fracture.^{7,10,23} The chronic irritation of the dura and the underlying brain may be the cause of late EEG anomalies (as in our case) or epileptic events. Basically, the relevance of these anomalies is hard to define since they are usually subclinical and do not require a specific drug treatments, but, at the same time, they are associated with the risk worsening over time and evolving into epileptic activity. Other authors, however, have not pointed out a significant correlation between depressed skull fractures and late posttraumatic seizures,⁹ nor between linear skull fracture and EEG abnormalities.¹² This shows that a bone injury can be a cause of EEG abnormality, but it is probably associated with dural damage. As far as the other disabilities in our patient are concerned, although some of them could be correlated with the compression (namely, the attention deficit), no changes have occurred after surgery to date, thus suggesting no strict relationship with the cephalohematoma.

A second, infrequent finding presented by our case is the late appearance of a CC on MRI and at surgery. Usually, fluid cephalohematoma does not require neuroimaging investigations other than ultrasound, although it can hide intracranial hemorrhage¹⁴ or skull lesions with fluid-fluid levels (such as Langerhans cell histiocytosis or aneurysmal bone cysts),¹⁸ or it can mimic an encephalocele.²⁸ On the other hand, neuroimaging may be needed in CC to exclude a deformation of the inner cranial surface, for assessment of the type of lesion, and for operative planning (selection of appropriate reconstructive technique). Neuroimaging may be useful also to rule out alternative diagnoses, such as bone tumor or scalloping of the skull due to brain tumors/lesions. On MRI, cephalohematoma exhibits a bright signal on T1-weighted images and a predominantly high signal with mild heterogeneity on T2-weighted images, which depends on the presence of methemoglobin and indicates that the lesion is secondary to hemorrhage rather than to an intraosseous tumor.^{6,26,27} CT is an alternative option for assessment of CCs,²⁹ which appear as a uniformly homogeneous, hypodense, nonenhancing core encased by bone. Thanks to 3D reconstruction, CT can be very useful for preoperative planning in the selection of appropriate reconstructive technique.

The treatment of CC is still debated because of the lesion's benign clinical course. In neonates, only observation is suggested, because the cephalohematoma can be

gradually absorbed as the skull develops.^{6,30} Some authors have proposed a passive cranial molding-helmet therapy as a successful nonsurgical treatment for CCs causing cranial asymmetry.²⁰ An accepted indication for surgery is the correction of skull asymmetry for cosmetic reasons.³ Other indications may be the prevention of brain growth restriction, and diagnostic confirmation and treatment of associated craniosynostosis.^{11,13,15,16} Surgery is performed according to the features of the CC. Type 1 CC is treated by drilling of the outer lamella, whereas Type 2 is treated by craniotomy and cranioplasty.²⁹ The intraoperative appearance of CC depends on the degree of calcification of the hematoma.^{4,8,29} In our case, the intraoperative appearance was characterized by an abnormal thickening of the skull; for this reason, a remodeling of the outer and inner lamella of the skull was performed, without using any prosthetic material.

Conclusions

Although quite common as a neonatal head injury, cephalohematoma rarely calcifies and even more rarely requires a surgical correction. No long-term effects of this disease are reported except for the cosmetic impact. Based on the present case, subclinical EEG anomalies may be considered as a late consequence of CC causing skull deformation and brain compression and deserving a specific follow-up in selected cases.

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Disclosures

The authors report no conflict of interest concerning the materials or methods used in this study or the findings specified in this paper.

Author Contributions

Conception and design: Frassanito, Tamburrini, Caldarelli, Massimi. Acquisition of data: Battaglia, Frassanito, Massimi. Analysis and interpretation of data: Battaglia. Drafting the article: Massimi. Critically revising the article: Vigo, Tamburrini, Caldarelli, Massimi. Reviewed submitted version of manuscript: Vigo, Tamburrini, Caldarelli, Massimi.

Correspondence

Vera Vigo, Institute of Neurosurgery — A. Gemelli Hospital, Largo A. Gemelli, 8, Rome 00168, Italy. email: vera.vigo@gmail.com.