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A case of chronic ossified hematoma presented as a skull lesion: A literature review on two rare conditions, cephalhematoma, and intradiploic hematoma

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Abstract

Ossified cephalhematoma (OCH) is a rare condition, especially when it presents as a skull lesion in the older pediatric population, which can managed with craniectomy and en-bloc excision of the organized hematoma.

A case of chronic ossified hematoma presented as a skull lesion: A literature review on two rare conditions, cephalhematoma, and intradiploic hematoma

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Abstract:

Cephalhematoma is a frequent condition in newborn infants due to birth-related trauma, but ossified cephalhematoma (OCH) is a rare condition, especially when it presents as a skull lesion in the older pediatric population. Chronic intradiploic hematoma (CIH) is another rare condition caused by an organized hematoma between the inner and outer tables of the skull. Differentiating CIH from OCH could be difficult for young neurosurgeons. We present an 18 months old girl with an OCH presented as a skull lesion, which was managed with craniectomy and en-bloc excision of the organized hematoma. This manuscript discusses the differences between OCH and CIH in diagnosis and management.

Background:

Cephalhematoma is a frequent condition in newborn infants due to birth-related trauma(1), but ossified cephalhematoma (OCH) is a rare condition, especially when it presents as a skull lesion in the older pediatric population(2). OCH is located under the periosteum (pericranium), and the pericranium plays a crucial role in enveloping the organized hematoma by an ossified tissue(3). The previous history of a soft fluctuant mass just after the birth is the key to the diagnosis of OCH(4).

Chronic intradiploic hematoma (CIH) is another rare condition caused by an organized hematoma between the inner and outer tables of the skull. Differentiating CIH from OCH could be difficult for young neurosurgeons. CIH presents as a slow-growing skull mass. The exact pathogenesis is unclear, but repetitive bleeding in the intradiploic space initiated by trauma may be the leading cause(3).

We represent a case of OCH presented as a skull lesion managed with craniectomy and en-bloc excision of the organized hematoma. We will discuss the differences between OCH and CIH in diagnosis and management.

Case report:

An 18-month-old girl presented with a non-tender slow-growing skull lesion in the right parietal region. The lesion was round, and bony textured on touch(figure1a). The neurological examination was normal. There was no medical complaint about the lesion other than a cosmetic problem. The patient had been born by standard vaginal delivery (NVD) to a nulliparous mother. Based on the mother's report, the baby suffered bilateral parietal soft masses just after birth. Although the left-sided lesion had vanished over time, the other side bulging grew for 18 months.

On the pre-operative examination, it was approximately 7*7*5 cm. The routine lab test was normal without any coagulopathy. Brain computed tomography scan (CT-scan) with a bone view showed a hypo-dense expanded mass between the outer and inner tables of the skull in the parietal region (Figure 2a) without any enhancement after contrast administration (Figure2b). History and CT scan were characteristics of OCH, so we decided against performing an MRI. Under general anesthesia, the skin was incised semi-circularly around the lesion in the supine position by turning the head 60 degrees to the left. The pericranium was dissected from the skin flap intending to use for final cranial reconstruction at the end of surgery. We performed a craniectomy after placing a burr hole over the ossified overlying layer on top of the lesion and blunt dissection over the pseudo-membrane. (Figure 1b, c). Subsequently, the organized hematoma was dissected circumferentially from the cranial bone (Figure 1d). After en-block hematoma removal, the uneven lateral borders of the skull were smoothened using a high-speed drill to maintain the proper shape of the skull (Figure 1e). Finally, we reformed the surgical site utilizing a layer of bone chips (harvested from the outer layer of the hematoma) and an overlying pericranium for better future cranial bone shaping and remodeling (Figure 1f). The postoperative course was uneventful. Figures 2c and 2d show the normal contour of the skull just post-operation. Histopathologic examination confirmed an organized hematoma with hemosiderin deposition and calcification in line with OCH (Figure 3).

Discussion:

Birth-related traumas could cause various cranial hematoma in neonates(5). Cephalhematoma is among the most seen hematomas, but OCH is a rare condition. According to previous reports, approximately 20 percent

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of cephalhematomas undergo calcification, but most resolve after conservative follow-up (2). When found at older ages, OCH could be mistaken for other ossified skull lesions (1). OCH can resemble a depressed fracture that needs different management(6). The pericranium covers the hematoma and, in older patients, causes the ossified outlining layer to reach the external table, which could be not distinguished with histological examinations. CIH is another rare pathology that could be mistaken with OCH. The exact etiology is still unknown, but the use of anticoagulants, trauma in birth, and shunt surgeries have been previously mentioned in the literature(7). Usually, CIH resolves spontaneously, but if it is not, a connective tissue surrounds the hematoma and forms a (pseudo) capsule(8). On conventional radiography and CT scan, the typical presentation of a CIH is a well-circumscribed, expanded intra skull lesion with or without surrounding sclerosis with varying attenuations and internal enhancing components after contrast injection. (9) Table 1 shows the previously reported cases of CIH in patients with confirmed histopathological diagnosis and without coagulopathies. The critical points for differentiating these two conditions could be obtained by a previous history of a soft fluctuant mass just after birth(4). Imaging could be helpful in cases where previous diagnostic hints are not available. In OCH cases, the contour of the underlying skull remains normal, but in CIH cases, narrowing of the inner and outer tables of the skull could be seen(3). Preoperative imaging is vital to correct diagnosis and treatment; MRI and CT scans are complementary, but in some cases, as in our patient, with a history of birth trauma, the CT scan may be diagnostic, and MRI is not mandatory.

The surgical management of these two conditions could be different. In CIH patients, the destruction of the inner table could need a reconstruction which can be done with cranioplasty. Still, in OCH patients, due to the excellent contour of the underlying skull, there is no need for cranioplasty, and an en-block excision could be obtained with a simple craniectomy as we did in our case(1)(3). Therefore, we suggest craniectomy with en-bloc organized hematoma removal as the treatment of choice for OCH. High speed-drill could be used to smoothen the borders of the underlying bone to give the skull a proper shape, as we did in this case. On the other side, in CIH patients with the destruction of the inner table, craniotomy followed by a cranioplasty is a better option.

Conclusion:

We presented a rare case of OCH treated with craniectomy and en-bloc mass resection. We briefly reviewed CIH diagnosis and management as an important differential diagnosis. Previous knowledge about these lesions could be helpful to un-experienced neurosurgeons for proper diagnosis and management.

Table 1. Demonstrating published case reports with CIH. We have excluded the patients with intradiploic hematoma that were finally diagnosed as giant cell reparative granuloma, hemophilic pseudotumor.

Author/Year	Age/Gender	History of trauma	Location	Management
Yausa et al./ 1992(10)	20 years old/ Male	Minor trauma	Parietal	Craniotomy and cranioplas
Sato et al./ 1994(11)	20 years old/ Male	Minor trauma	Parietal	Craniotomy and cranioplas
Uemura et al./ 1997(12)	32 years old/ Male	Minor trauma	Frontal	Excision (no data available
Yucesoy et al./ 1999(13)	25 days old/ Male	Birth trauma	Parietal	External table craniectomy
Mobbs et al./ 2000(14)	Three years old/ Male	Major trauma	Parietal	Curettage following aspirat
Goel et al./ 2000(15)	58 years old/ Male	Minor trauma	Frontal	Craniectomy without crani
Batista et al./ 2011(16)	45 years old/ Male	Recurrent trauma	Parietal and occipital	Craniotomy and cranioplas
Tokmak et al./ 2015(7)	16 years old/ Male	Minor trauma	Frontal	Excision (no data available
	64 years old/ Male	Minor trauma	Frontal	Curettage (no data availab
Luo et al./ 2017 (17)	50 days old/ Male	Birth trauma	Parietal	Craniotomy and cranioplas
Park et al./ 2020(9)	54 years old/ Male	Major trauma	Parietal	Craniectomy (no data avai

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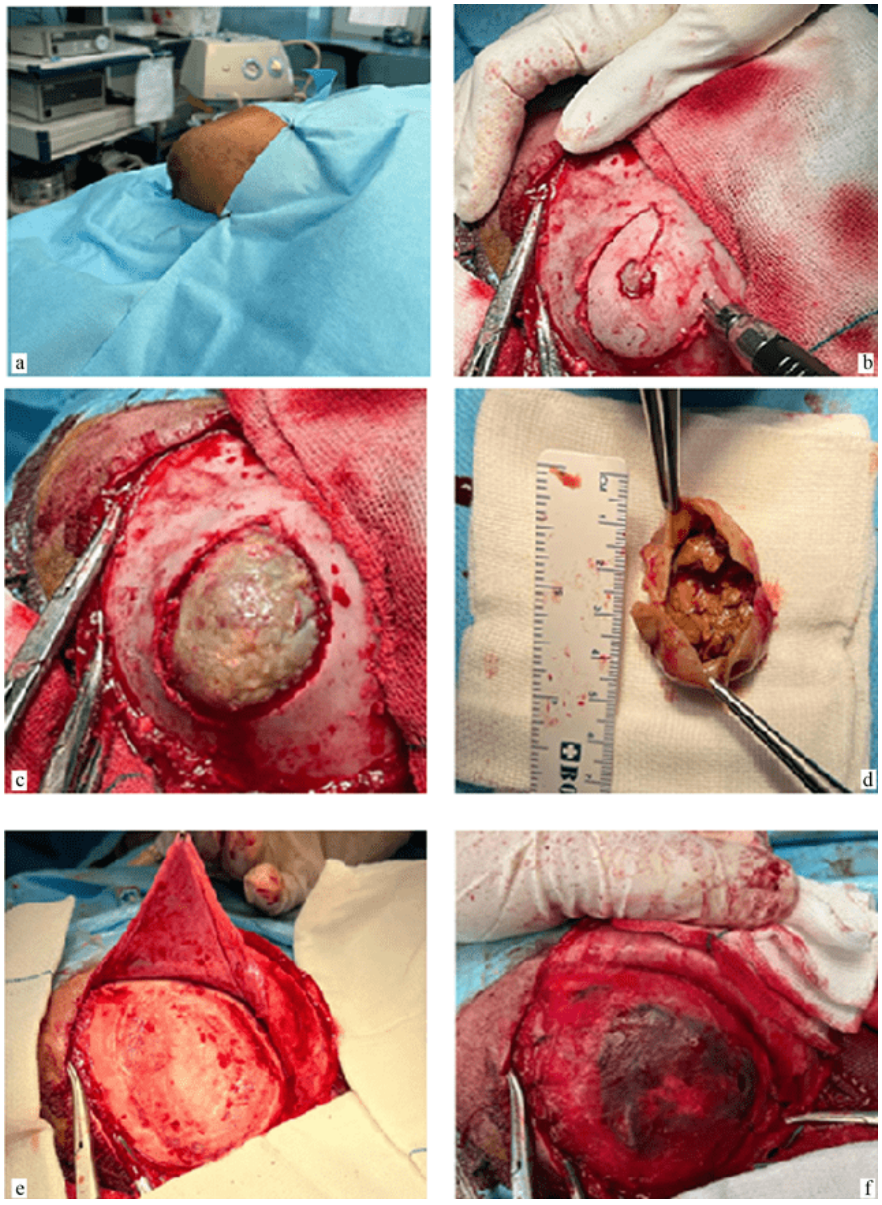

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