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Case Report

Is skull fracture necessary for developing an intradiploic pseudomeningocele as a complication of head injury in adulthood?

A. Menkü, R. K. Koç, B. Tucer, and **H. Akdemir**

Department of Neurosurgery, Erciyes University, School of Medicine, Kayseri, Turkey

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Summary

We report the case of an adult with a posttraumatic intradiploic pseudomeningocele which caused an expanded osteolytic skull lesion. Local pain and swelling, the only symptoms of the lesion, regressed after surgery. Intradiploic pseudomeningocele must be distinguished from intradiploic leptomeningeal cyst, which is of traumatic origin or arachnoid cyst and epidermoid cyst, which are of congenital origin. We also discuss the development of intradiploic pseudomeningocele after head trauma without skull fracture in adulthood and suggest a possible mechanism.

Keywords: Arachnoid cyst; head trauma; leptomeningeal cyst; osteolytic skull lesion; pseudomeningocele.

Introduction

Intradiploic pseudomeningocele is a rare osteolytic skull lesion [7, 10, 13]. It is especially associated with head trauma in childhood [7, 10, 13]. A possible mechanism in children has been suggested in the literature [10]. However, the mechanism of development of posttraumatic intradiploic pseudomeningocele in adulthood is still debated. Magnetic resonance (MR) imaging is a useful diagnostic method for differential diagnosis from other osteolytic skull lesions [2, 9, 11]. However, the most useful diagnostic method is surgical exploration and histopathological examination. Epidermoid cyst or intradiploic arachnoid cyst should be considered in the differential diagnosis. We report an intradiploic pseudomeningocele with a history of trauma which caused a circumscribed osteolytic skull lesion in an adult.

Case reports

A 30-year-old woman was admitted with complaints of constant headache and a swelling in right parietal area of her skull, both of which had been present for 6 months. The swelling was approximately 2 cm in diameter when it was first noticed, but had enlarged in the past 6 months. There was a history of trauma. Five years before, she had an accident. Plain X-rays of the skull were taken but no fractures were seen. However, now physical examination revealed a firm, painful, fixed swelling of 4 cm in diameter involving the right parietal bone. The patients' neurological examination and laboratory values were normal. Skull radiography showed a large lucent area with a well-defined sclerotic margin in the right parietal bone (Fig. 1a, b). Computed tomography revealed a defect of the inner table, widened diploic space, and thinned outer table (Fig. 2). MRI showed a cyst containing cerebrospinal fluid. The cyst appeared as hypo-intense on T1 and hyperintense on T2-weighted images (Fig. 3a, b). Dural defect also was seen on MR images. Axial diploic section of MR showed a round hyperintense area (Fig. 4).

At operation a scalp flap was lifted and a thinned shell-like outer table of the skull was revealed (Fig. 5). When opened, clear cerebrospinal fluid drained from a large intradiploic space, which was lined by a thin membrane. There was intra- and extra-cranial bulging of the calvarium. The posterior wall of the cavity and the dura mater behind it contained a small round defect (Fig. 6). After resection of the cyst wall and closure of the dural defect, the involved cranial defect region was reconstructed with the outer table of the parietal bone (Fig. 7). Postoperative course was uneventful. At 6 months postoperatively, the patient was well and free of any symptoms or recurrence. Histological examination showed the lining membrane in the cavity to consist of non-specific fibrous tissue.

Discussion

A review of the literature indicates that the primary traumatic event almost always occurs in childhood and the cyst presented many years later [1, 4, 9]. However,

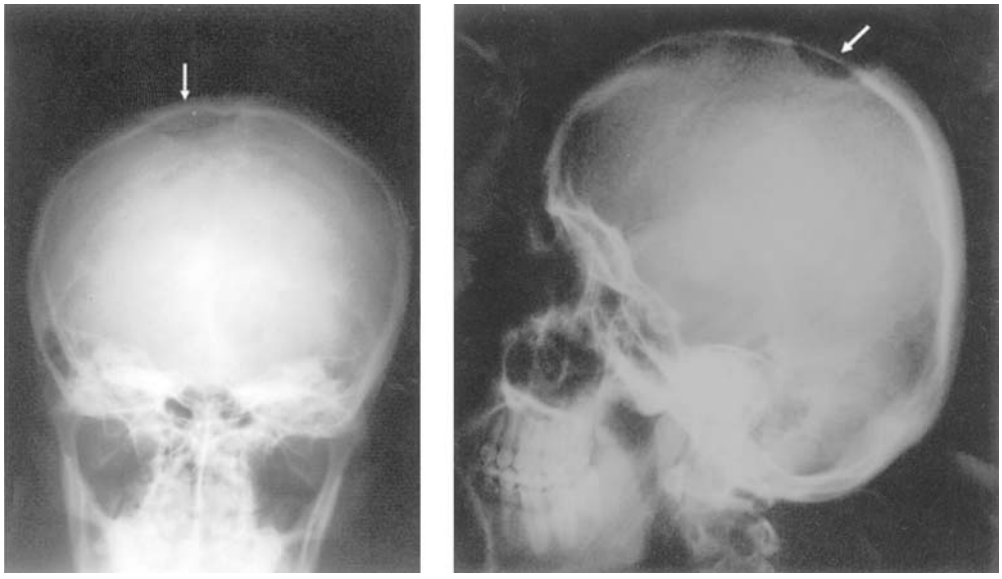

Fig. 1. Anterior-posterior (a) and lateral (b) skull radiographs showing a large lucent area with a well-defined sclerotic margin in the right parietal bone (arrow)

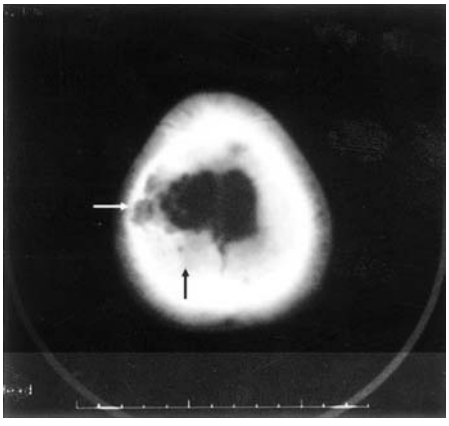

Fig. 2. Computed tomography revealing a defect of the inner table and widened diploic space (black and white arrow)

Sartawi *et al.* [12] reported the first case of so-called arachnoid cyst associated with a single osteolytic lesion of the occipital bone of the skull and a history of trauma 2 years previously. Cook *et al.* [5] described a case of so-called leptomeningeal cyst developing in an adult as a sequel to a skull injury 12 months earlier. Lunardi *et al.* [9] also described another leptomeningeal cyst developing after trauma in adulthood. Different mechanisms for their development have been suggested in the literature. According to D'Almedia and King [6], on the other hand, if the skull fracture affects only the inner table and dura mater, the so-called leptomeningeal cyst develops within the diploe, resulting in elevation of the outer table and flattening of the inner table with pulsation of CSF collection. Weinand *et al.* [14] described 2 patients in whom the cranial arachnoid cysts developed as diverticuli of the arachnoid membrane through a small defect in the dura mater. The arachnoid membrane is intact and these lesions have been considered as congenital. However, the wall of the cyst in our case consists out of aspecific fibrotic tissue.

The mechanism of development of an intradiploic pseudomeningocele is controversial. Mahapatra *et al.* [10] suggested that both arachnoid and dura mater are damaged at the time of the trauma. This leads to the formation of a collection of cerebrospinal fluid. The pulsation is transmitted through a localized collection. This separates the two tables of the fractured bone resulting in the formation of an intradiploic so-called 'pseudomeningocele'. This mechanism is similar to ours in some aspects. However, our case is an adult and there was no dural damage or skull fracture.

We think that this is the first case, which developed as a pseudomeningocele in an adult without a skull fracture. The wall of the cyst in our case contained only a specific fibrous tissue. Therefore, it is better to speak about a 'pseudomeningocele' instead of a 'leptomeningeal cyst'. According to the literature, minor or moderate head trauma may cause an intradiploic cyst especially in the parietal zone in adults many years after head trauma [9]. For these reasons, we think that intradiploic cyst, in an adult develops via the foveolae granulare. Foveolae

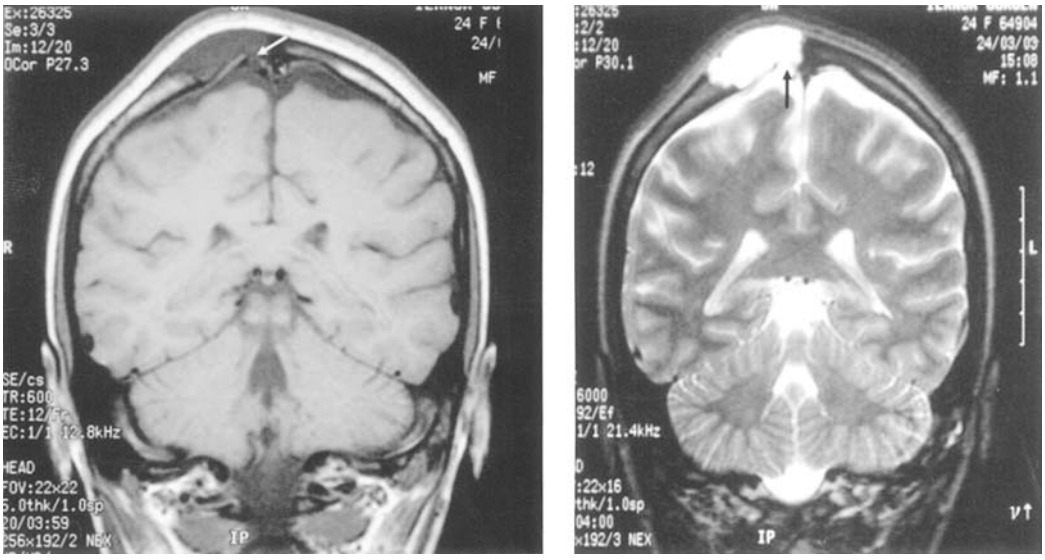

Fig. 3. Hypo-intense on T1 (a) and hyperintense on T2 (b) weighted coronal MR images showing the cystic lesion and dural defect (white and black arrow) in right parasagittal region

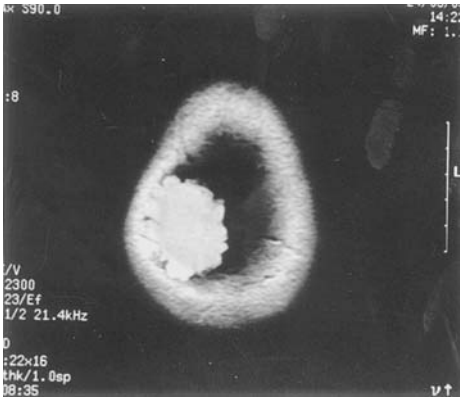

Fig. 4. Axial diploic MR image showing a round hyperintense area of 4 cm in diameter

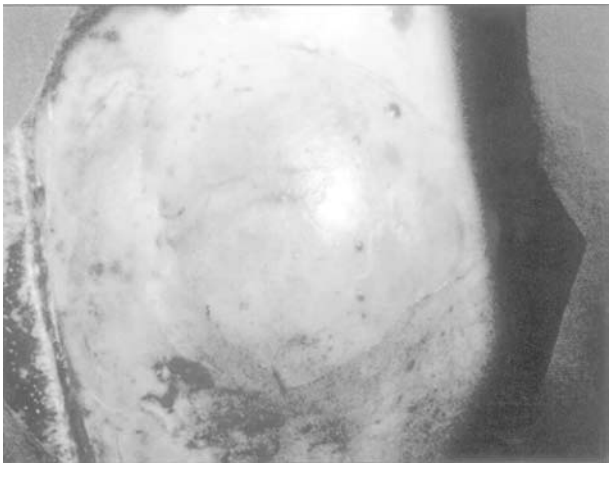

Fig. 5. Intra-operative photograph revealing thinned and eroded outer table of the skull

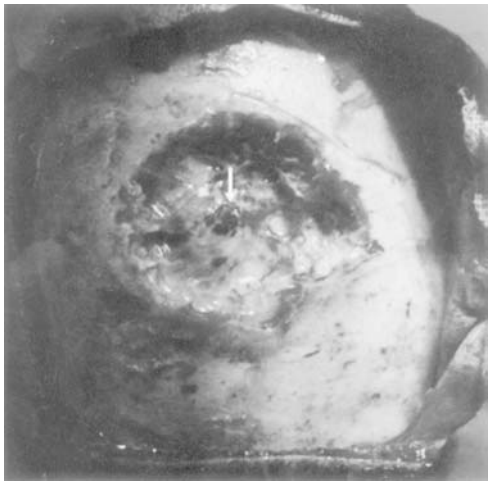

Fig. 6. Intra-operative photograph showing a small round dural defect (white arrow)

granulare are an anatomical and physiological defect which include Pacchioni an granulations of the inner table of the fronto-parietal bone and near the superior sagital sinus. The arachnoid membrane is damaged with head trauma but the skull is not fractured. Foveolae granulare expand along the diploe with pulsation of CSF collection. Thus, the process results with elevation of the outer table and flattening of the inner table (Fig. 8).

MRI is the most useful non-invasive diagnostic method for differential diagnosis from other osteolytic skull lesions. In our case, MRI revealed intradiploic cysts containing cerebrospinal fluid and showed a small

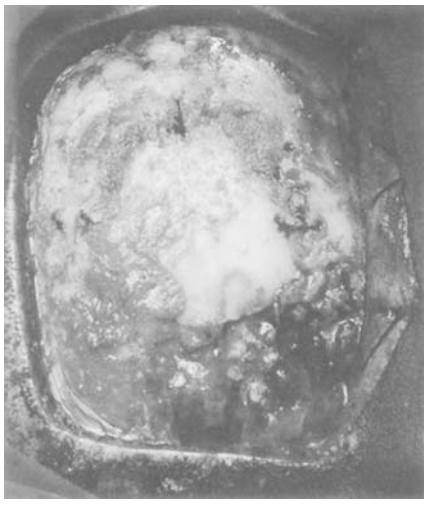

Fig. 7. Intra-operative photograph showing that the cranial defect was reconstructed with the outer table of the parietal bone

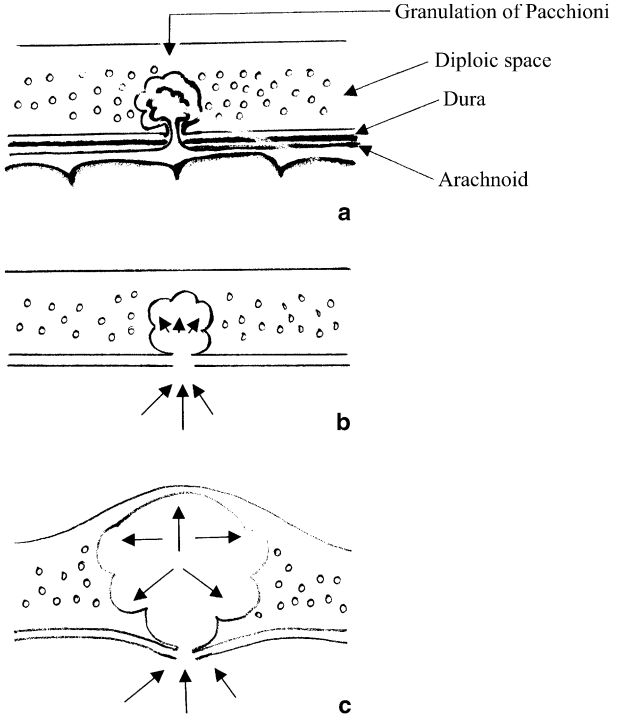

Fig. 8. Illustrations demonstrate development of an intradiploic pseudomeningocele. Normal anatomy of foveolae granulare including Pacchioni an granulations and diploe are shown are in the illustration (a); the arachnoid membrane is damaged by head trauma but the skull is not fractured (b), foveolae granulare expand along the diploe with pulsation of CSF collection and intradiploic pseudomeningocele develops (c)

dural defect. Operation also found the cyst filled with CSF and a small round dural defect.

Presenting symptoms of intradiploic cysts are headache and swelling such as in our case. However, neurological disorders were rarely seen with intradiploic cysts [1].

From the history, clinical findings and extensive radiological investigations, an epidermoid or an intradiploic arachnoid cyst was considered in the differential diagnosis. Intradiploic epidermoid cyst could be excluded because the lesion was of low intensity on diffusion-weighted MR images [15]. Intradiploic arachnoid cyst seems to be congenital in origin but commonly found in the elderly [3]. However, a forgotten injury might play a significant role [2, 11]. Arachnoid cyst is a benign lesion, so exploratory surgery should be avoided unless the cyst is symptomatic.

In conclusion, a physician should be reminded of the possibility of trivial head trauma causing the development of an intradiploic cyst not only in childhood but also in adulthood. In such cases, MR especially diffusion-weighted MR imaging is essential in the differential diagnosis and surgery should be performed if it is symptomatic.

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Comments

The authors present a rare case of intradiploic leptomeningeal cyst (ILMC) as a complication after a skull fracture in an adult. Usually the ILMC occur after skull fractures in children because of the more favourable conditions in this age group leading to such a complication. However, in adults the occurrence of this complication is unusual. The mechanisms for developing an ILMC described in the literature are two:

1. Damage to dura mater and arachnoid during the trauma allows a CSF collecton, which causes the cyst by pulsations.
2. Skull fractures affecting only the inner table and dura mater allows the creation of an ILMC due to the CSF pulsation pressure. This mechanism occurs with a high incidence in adults.

Modern neuro-imaging armamentarium makes a complete radiological diagnosis after cranio-cerebral trauma possible. The diagnosis of the inner table defect and the modifications of the diploic space made the surgical treatment necessary in order to stop the development of ILMC into the diploic structures.

The authors present the mechanisms and the surgical treatment of one of these rare cases in adulthood on the basis of a complex documentation. They explain these rare cases of ILMC on the basis of selected references focused on the main subject.

I consider it interesting to publish this article in ''Acta Neurochirurgica'' in order to keep the possibility of this complication after skull trauma in mind.

A. V. Ciurea

The authors report on an intradiploic pseudomeningocele occurring as a complication of head trauma. The occurrence of a noticed head trauma without a skull fracture clearly distinguishes the present case from the others described in the literature. This point has been correctly highlighted by the authors, and clearly makes this case unique as the first described pseudomeningocele in an adult without skull fracture. An additional value of this paper may be seen in focusing on the necessity of identifying a precise definition of the condition. Indeed, numerous, often confusing, descriptions of post-traumatic complications leading to disruption of the brain meningeal coverings have been reported in the past, and often incorrectly identified under the condition of leptomeningeal cysts. This nosographic inaccuracies have prevented appropriate comparison of experiences, especially in terms of appropriate management and evalution of outcomes.

Domenico d'Avella and *Antonino Germanò*
Messina, Italy

Correspondence: Ahmet Menkü, MD., Department of Neurosurgery, Erciyes University, School of Medicine, 38039, Kayseri, Turkey. e-mail: menkua@erciyes.edu.tr