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Short Illustrated Review

Non-traumatic intradiploic arachnoid cyst

A. C. Iplikcioglu, C. Dinc, K. Bikmaz, S. Bek, C. A. Gokduman, and **M. Cosar**

Neurosurgery Clinic, Ministry of Health Okmeydani Education and Research Hospital, Istanbul, Turkey

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Summary

In this report, we present a case of non-traumatic intradiploic arachnoid cyst in a 65 year-old woman with a slow growing swelling in the right frontotemporal region without a history of head trauma, which was diagnosed intra-operatively. Extradural intracranial location of non-traumatic arachnoid cyst is a rare clinical entity with a few reported cases in the literature. Characteristic features of non-traumatic intradiploic arachnoid cysts are also described in this mini-review article.

Keywords: Arachnoid cyst; bone; diploic space; non-traumatic.

Introduction

Arachnoid cysts are benign developmental abnormalities of the arachnoid membrane which may occur anywhere in the cerebrospinal axis. They are rare lesions and account for only 1% of all intracranial space-occupying lesions [8]. Despite the intracranial arachnoid cysts that are located intradurally, spinal arachnoid cysts are rarely intradural [10, 15]. However, non-traumatic intradiploic arachnoid cyst is the only extradural example of intracranial arachnoid cysts. In this report, we present a case of non-traumatic intradiploic arachnoid cyst in association with a mini review of the literature.

Literature review

The Medline database on the internet (PubMed, http://www.ncbi.nlm.nih.gov/PubMed/) was searched for the following keywords in the English language between 1966–2004: Non-traumatic, intradiploic and arachnoid cyst.

7 articles were found [1, 2, 4, 6, 10, 15, 16] (Table 1). In addition, a standard neurosurgical textbook was used [8].

Analysis

A total of 13 cases were found in 7 articles in the literature [1, 2, 4, 6, 10, 15, 16]. 5 out of the 13 cases were reported in an article by Hasegawa [6].

5 cases were male and 8 female. The ages ranged between 30 and 74 with an average of 61.6. All patients except one were over the age of 50 and most of the lesions were located in the occipital bone. Half of these cases had single lesions while the others were multiple. In one case, multiple intradiploic arachnoid cysts were located in two different regions [15]. However, other multiple lesions were located in the same region. 3 out of the 13 cases had local pain [1, 15] and only one case had proptosis [10]. The others were asymptomatic.

In Hasegawa's [6] series all five patients were asymptomatic and the lesions were located in the occipital bone. In this report, 4 out of the 5 cases had multiple lesions and only one patient was not operated upon.

11 out of the 13 cases underwent explorative surgery and diagnosis was established based on the peroperative observations and histopathological examinations. In one report, the case was diagnosed by diffusion-weighted MR, and was not operated upon [16].

Illustrative case

A 65-year-old woman was admitted with a slow, hard, growing swelling in the right frontotemporal region. She had noticed the swelling 5 years ago, but she did not experience any head trauma. Neurological and physical examinations were normal except for a right frontotemporal hard swelling with a 6 mm diameter.

Table 1. *Summary of reported cases of intradiploic arachnoid cysts*

Author (year)	Age/sex	Multiplicity	Affected bones	Symptoms	Surgery
D'Almedia and King (1981)	61/M	single	rt parietal	no	yes
	53/M	single	rt frontal	no	yes
Weinand *et al.* (1989)	70/F	multiple	occipital, lt parietal	yes: local pain	yes
	68/F	multiple	occipital	yes: local pain	yes
Hasegawa *et al.* (1992)	54/F	multiple	occipital	no	yes
	70/F	single	occipital	no	yes
	58/F	multiple	occipital	no	yes
	71/M	multiple	occipital	no	yes
	74/F	multiple	occipital	no	no
Alfieri *et al.* (1996)	57/F	single	lt frontal	yes: local pain	yes
Asahi *et al.* (1998)	63/M	single	rt parietal	no	yes
Krupp *et al.* (1999)	30/M	multiple	rt frontal	yes: proptosis	yes
Yamaguchi *et al.* (2002)	72/F	single	occipital	no	no
Present case	65/F	single	rt frontotemporal	yes: swelling	yes

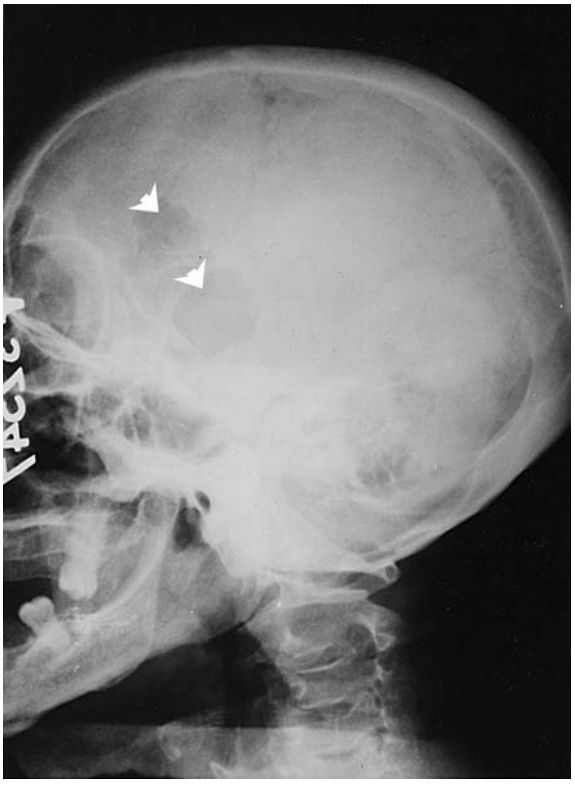

Fig. 1. Lateral plain skull film showing multiloculated osteolytic lesions (arrows) with slightly sclerotic margins

Plain skull X-rays revealed a multiloculated osteolytic lesion with slightly sclerotic margins (Fig. 1). On Computed Tomography (CT) scans an intradiploic hypodense lesion was seen with thinned internal and external tables (Fig. 2). On Magnetic Resonance Imaging (MRI) the lesion was iso-intense with Cerebro-Spinal-Fluid (CSF) on T1 weighted images and the adjacent brain tissue was normal (Fig. 3).

Fig. 2. CT scan showing an intradiploic hypodense lesion; with thinned internal and external tables

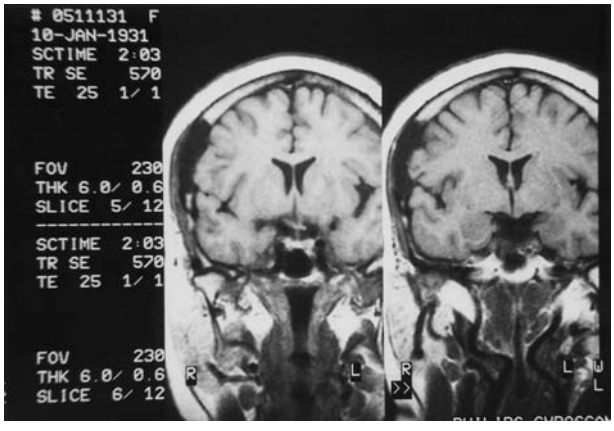

Fig. 3. MRI coronal sections: iso-intense right frontotemporal lesion with CSF on T1 weighted images, the adjacent brain tissue was normal

A right frontotemporal craniotomy was performed. The outer table was thin and appeared to be eroded and a clear fluid was encountered when a burr hole was

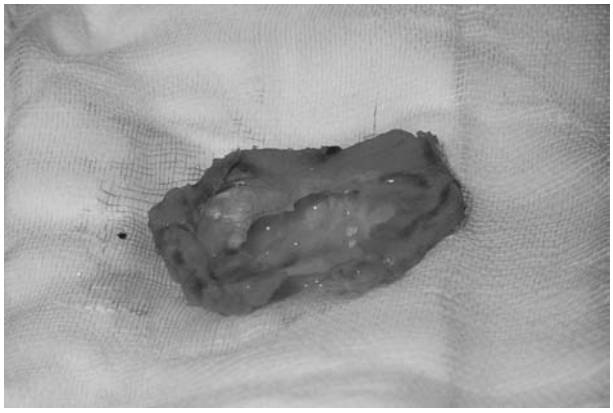

Fig. 4. Surgically removed specimen; intraosseous lesion was a thin walled cyst, filled with clear fluid and created multiloculation in the diploic space

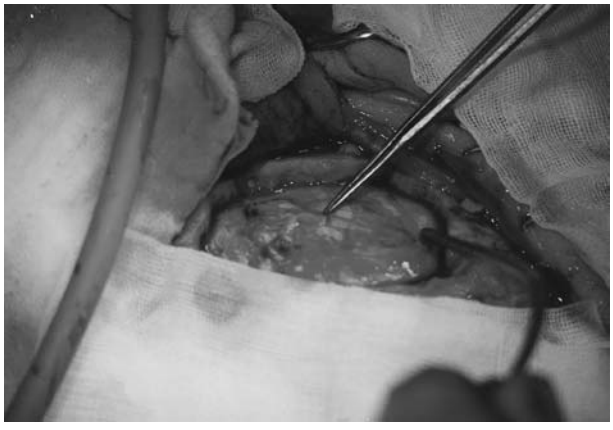

Fig. 5. Cyst had a pedicle (tip of the forceps) entering through a small round defect of dura of several millimeters in diameter

opened. The intraosseous lesion was a thin walled cyst filled with clear fluid which created multiloculation in the diploic space (Fig. 4). However, the cyst had a pedicle entering through a small round defect of dura of several millimeters in diameter (Fig. 5). After the resection of the cyst pedicle, the dural defect was closed with a small piece of temporal fascia. The bone defect was reconstructed with acrylic.

The postoperative course was uneventful. Histopathological examination of the cyst revealed an arachnoid cyst.

Discussion

Pathogenesis

Intracranial arachnoid cysts usually develop as a CSF pocket within the two layers of the arachnoid membrane which are not normally differentiated. However, non-traumatic intradiploic arachnoid cysts develop as a diverticulum of the arachnoid membrane through small defects in the dura mater. The continuous pulsations of the herniated arachnoid through the defect erodes the inner table and subsequently expands the intradiploic space which is less resistant than the inner and outer tables. The congenital extradural spinal cyst and growing skull fractures also have similar pathogenesis while developmental dural defects are present in the former and acquired defects in the dura are present in the latter [3, 7, 13]. Therefore, non-traumatic intradiploic arachnoid cyst resembles the cranial equivalent of a spinal extradural arachnoid cyst.

Weinand *et al.* [15] first described two adult patients with intradiploic arachnoid cysts which developed as diverticula of the arachnoid through small dural defects. Hasegawa *et al.* [6] reported 5 cases of occipital intradiploic arachnoid cyst without any symptoms and operated upon 4 of the patients. Recently, Krupp *et al.* [10] reported a case of large intradiploic arachnoid cyst involving craniofacial osseous structures. The cyst had an extension from the right anterior cranial fossa to the infratemporal fossa.

However, lesions described with various names such as traumatic arachnoid cyst without a fracture and intradiploic cerebrospinal fistula probably represent the same pathological process. In this case, the authors concluded that the lesions were caused by a forgotten trauma [4, 9, 14].

In the case reported by Handa and Karapurkar [5], a large intradiploic hematoma without a dural defect was found at the operation. This case represents chronic diploic hematoma rather than an intradiploic arachnoid cyst.

Clinical findings

Non-traumatic intradiploic arachnoid cysts are usually present in the elderly. Local pain is the most common symptom. Most of the patients were diagnosed incidentally who had radiological examinations that were obtained for other reasons. In Krupp's [10] case the patient presented with facial deformation.

Differential diagnosis from growing skull fracture

The rarity of non-traumatic intradiploic arachnoid cyst is due to two factors. First, it is not included in the classical textbooks of neurosurgery; second it is very

similar to the growing skull fracture, a more common and well known clinical entity.

Non-traumatic intradiploic arachnoid cysts have several characteristic features different from that of traumatic growing skull fractures. First, the dural defect is round and small in non-traumatic cases while it is linear in configuration and has a long axis parallel to the skull fracture in traumatic cases. Second, in the growing skull fracture the outer table is usually involved with scalloped and everted margins, although the outer table is intact in the non-traumatic intradiploic arachnoid cyst [7]. Finally, there is an absence of brain tissue damage adjacent to the non-traumatic arachnoid cyst. In growing skull fractures there is usually some degree of brain tissue damage (contusion, cystic mass, dilatation of the ipsilateral ventricle) under the bone lesions, although exceptionally traumatic cases without skull fracture have been reported [11, 12]. This damage is well seen on the MRI examinations.

Radiological imaging

On plain skull films the differential diagnosis of intradiploic arachnoid cyst includes myeloma, intradiploic meningioma, chronic intradiploic hematoma, metastasis, epidermoid cyst, dermoid cyst and eosinophilic granuloma. Chronic intradiploic hematomas can be differentiated based on different blood density and intensity on CT and MRI examinations, while tumors other than epidermoid cysts usually show contrast enhancement. However, on CT and MRI examinations it usually mimicks intradiploic epidermoid cysts. Recently, Yamaguchi *et al.* [16] used diffusion weighted MRI to differentiate intradiploic arachnoid cyst from epidermoid cyst. Epidermoid cysts appear as a high signal intensity on diffusion weighted MRIs, whereas arachnoid cysts have a low signal intensity. It is believed that CT examination with intrathecal contrast injection may be helpful in the diagnosis.

Treatment

The surgical treatment of non-traumatic intradiploic arachnoid cyst is simple and successful. The excision of the pedicle and the repair of the small dural defect and the bone defect are the proper steps of the surgical treatment. However, in most of the reported cases, an operation was performed for the histological diagnosis. If preoperative diagnosis of intradiploic arachnoid cyst can be established in asymptomatic patients with small lesions radiological follow up should be done due to the benign nature of the lesion.

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Comment

As a mini-review of this rare entity of non-traumatic intradiploic arachnoid cysts, I think it provides informative values to the readers.

Dong Gyu Kim
Seoul

Correspondence: Cem Dinc, Icerenkoy Emniyet Cik. Sk. Aydin Kardesler Apt. No. 4/16, 34752 Kadikoy, Istanbul, Turkey. e-mail: drcemdinc@yahoo.com