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F A Eames and J B Waldman

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CT of Posttraumatic Intradiploic Pseudomeningocele of the Skull Base: A Case Report

Frederick A. Eames¹ and John B. Waldman²

A leptomeningeal cyst is a posttraumatic arachnoid cyst or pseudomeningocele associated with an expansile calvarial defect at the original site of fracture [1]. These occur infrequently in adulthood, and involvement of the skull base is extremely rare. We represent the case of a large posttraumatic leptomeningeal cyst of the sphenoid bone that appeared approximately 30 years after injury.

Case Report

A 36-year-old woman presented with a 1-year history of headaches and increasing retroorbital pain on the right.

Contrast-enhanced CT in axial and coronal planes showed a large low-density lesion in the skull base on the right (Figs. 1A–1C). This appeared as an expansile intradiploic cavity within the greater wing and body of the sphenoid, clivus, and right side of the sphenoidal sinus, with erosion of the anterior clinoid process and extension into the pterygoid process and the lateral orbital wall. A discontinuity in the medial part of the inner table of the floor of the middle fossa could be identified. The preoperative diagnosis was probable mucocoele of the sphenoidal sinus.

A transnasal sphenoidotomy was performed, resulting in a copious flow of CSF. Packing material was placed in the upper nasal cavity and no further intervention was attempted. Repeat cranial CT showed pneumocephalus, and the patient's headaches became more severe. Transnasal packing of the sphenoid cavity failed to stop the CSF leak. Postoperatively, a meningitis developed, which was treated with appropriate antibiotics, but the CSF rhinorrhea continued. With close questioning, additional history of severe head trauma at age 5 (fall out of a moving vehicle) was obtained. Nine weeks after her first admission, the patient underwent a right temporal craniotomy. In the medial part of the floor of the right middle fossa, the dura was dehiscence over an area considerably larger than the bone defect apparent on CT. There was no herniation of brain through the defect. A free graft of pericranium was harvested and sutured over the dural defect to create a water-tight closure. An exploration of the lateral part of the cavity, in the orbital wall, was also performed; no tumor was found, but a biopsy of the lining of the cavity wall was done. Postoperatively, CSF rhinorrhea ceased, but it recurred several weeks later, at which time the patient was referred to another institution. The skull base defect was closed from an extracranial approach, using the posterior half of the temporalis muscle. One year

after initial presentation, intermittent headaches persisted, but CSF rhinorrhea has not recurred. Follow-up CT showed nearly complete obliteration of the intradiploic cavity with fat/muscle density. Microscopic examination of the lining of the cavity wall showed a nonspecific fibrous-appearing matrix, with no evidence of atypia or mitotic activity (Fig. 1D). Immunohistochemical stains (S-100, NSE) for neural elements were negative. Vimentin stain was positive, indicating cells of meningotheelial origin.

Discussion

The clinical and radiologic features of leptomeningeal cysts are well known [1–3]. They are uncommon complications of head injury, developing in approximately 0.6% of all skull fractures [4], and occur almost exclusively in infants and young children. The fracture occurs in children under the age of 3 years in 90% of cases [3]. Both a dural tear [1] and arachnoid opening [5] at the time of injury appear to be required for formation of a leptomeningeal cyst. Generally, arachnoid adhesions prevent free communication of the cyst with the subarachnoid space (Fig. 2A) [1, 6]. Histologically, the cyst wall sometimes resembles arachnoid, but more often is described as nonspecific fibrous tissue or a fibrocollagenous neomembrane [3].

Adult presentation of leptomeningeal cyst is rare. Of nine cases identified in the literature [4, 7–14], five were consequent to head injury in adulthood (1–12 year interval before clinical presentation); four cases were consequent to childhood injury (16–50 year interval). In three patients (two with adult trauma, one with childhood trauma) there was involvement of the sphenoid and/or temporal bones in the skull base; two patients had trigeminal neuropathy, the other presented with CSF rhinorrhea. In each case there was extensive bone erosion, but there was no intradiploic cavity. In none of these was there evidence of free communication between cyst and subarachnoid space. On the other hand, there are three reports of adults with posttraumatic expansile intradiploic cavities in the occipital bone freely communicating with the subarachnoid space [8, 10, 12]; these could be properly termed intradiploic pseudomeningoceles (Fig. 2C). Very simi-

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¹ Department of Radiology, Neuroradiology Section, Albany Medical Center Hospital, 43 New Scotland Ave., Albany, NY 12208. Address reprint requests to F. A. Eames.

² Department of Surgery, Division of Neurosurgery, Albany Medical Center Hospital, Albany, NY 12208.

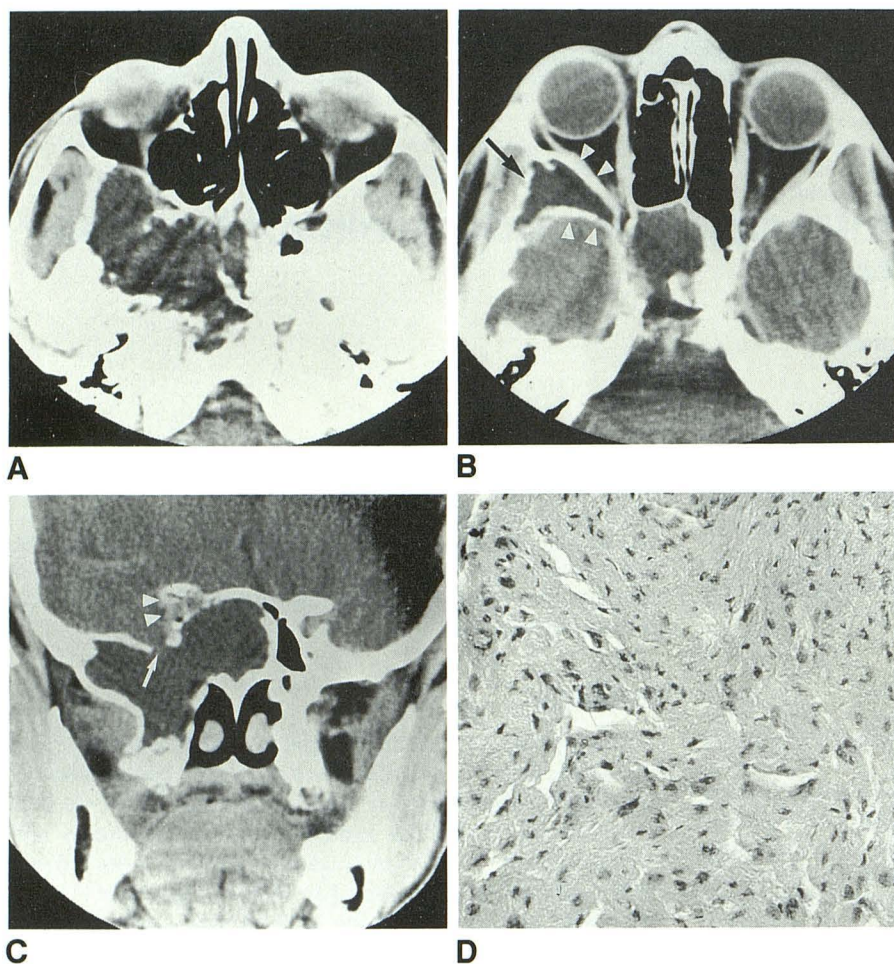


Fig. 1.—36-year-old woman with 1-year history of headaches and increasing retroorbital pain on right side.

A and B, Preoperative axial contrast-enhanced CT scans show expansile low-attenuation lesion in clivus, floor of middle fossa, and lateral wall of orbit (arrow), where a biopsy of cyst was done. Note expanded inner and outer tables of greater sphenoid wing (arrowheads).

C, Coronal contrast-enhanced CT scan shows extent of cavity in body of sphenoid and pterygoid process. Note focal defect in floor of middle fossa (arrow) and erosion of anterior clinoid process (arrowheads).

D, Histologic section from tissue lining cavity of lateral orbital wall. Numerous spindle cells are arranged in a haphazard pattern. There is no atypia or mitotic activity. Immunohistochemical stains suggested that cells were of meningeothelial origin. (H and E, original magnification $\times 40$)

lar to these are four cases of intradiploic communicating cysts, reported by D'Almeida and King [15] and Weinand et al. [16]. None of these four patients had a clear history of significant head injury. D'Almeida and King suspected, nevertheless, a traumatic origin, and termed these "idiopathic intradiploic CSF fistulae"; there was no arachnoid in the diploic space. Weinand et al. favored a developmental origin in their cases and preferred the term "intradiploic arachnoid cyst," noting that pathologic examination in these cases identified true arachnoid membrane lining the cyst wall.

Congenital basal encephaloceles are rare anomalies, and transsphenoidal or intrasphenoidal encephaloceles are the least frequent type (1 in 700,000 live births [17]). Transsellar sphenoidal encephaloceles have been reported [18] but we have found no reports of transsphenoidal encephalocele with such marked expansion of the diploic space. Furthermore, there was no intracranial CNS abnormality in our case, nor was any neural tissue detected in the cyst, although the presence of neural tissue would not rule out a traumatic pathogenesis [19].

Also related are cases of spontaneous (nontraumatic) CSF rhinorrhea attributed to enlarged "pitholes" in the anterior medial part of the middle fossa floor [20, 21]. In these, there were fistulae between the subarachnoid space and a large lateral extension (pterygoid recess) of the sphenoid sinus.

The reported position of these "pitholes" matches closely the bone defect in our own case, but in our case marked expansion of the diploic space has occurred without communication with the true cavity of the sphenoid sinus.

The present case appears to be an extremely unusual presentation of an intradiploic pseudomeningocele of the sphenoid bone. A large mucocoele or primary expansile bone lesion, such as an aneurysmal bone cyst, cystic fibrous dysplasia, or nonossifying fibroma might have a similar appearance, but the dural defect can be explained only by a developmental or traumatic etiology. In our case, we cannot establish the cause or origin with certainty, but we favor a traumatic one. There was a definite history of head injury in childhood. The apparently free CSF communication is atypical for leptomeningeal cyst but, as noted above, communicating posttraumatic intradiploic cysts have been previously reported. The histology of the cyst wall in our case is consistent with previous reports of posttraumatic cysts.

Clearly, CT-cisternography would have been useful in the preoperative evaluation of our patient. However, CSF rhinorrhea had not occurred, and initially there was no reason to suspect a dural defect.

In conclusion, we believe that this case is most properly considered an intradiploic pseudomeningocele of the skull base, probably posttraumatic, presenting some 30 years after

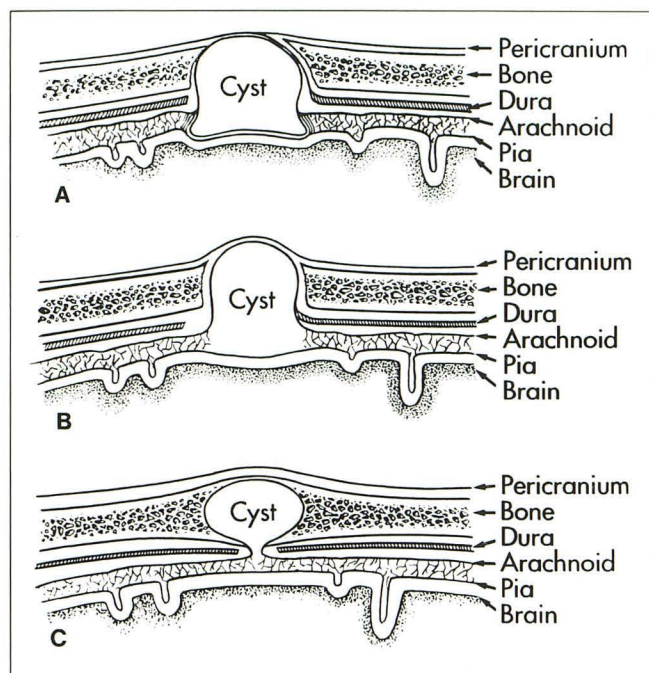


Fig. 2.—Comparative diagrams of: A, leptomeningeal cyst; B, pseudomeningocele; and C, intradiploic pseudomeningocele. The typical leptomeningeal cyst is considered a noncommunicating pseudomeningocele; note the marginal arachnoid adhesions in A. In each case, the location and continuity of the dural margin are variable.

injury. It represents one of several types of atypical leptomeningeal cysts that have been reported in adults.

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