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Direct Intraoperative Sclerotherapy of an Aneurysmal Bone Cyst of the Sphenoid

Carl Chartrand-Lefebvre, Josée Dubois, Daniel Roy, Claude Mercier, and Jean Raymond

Summary: A young boy presented with a symptomatic aneurysmal bone cyst of the left great wing of the sphenoid bone. Arterial embolization had failed to produce thrombosis, and the lesion could not be resected surgically. Direct intraoperative sclerotherapy resulted in immediate thrombosis of 80% of the volume of the vascular malformation with no progression of symptoms. Two years later, the symptoms had completely resolved, and CT scans showed total ossification of the lesion.

Index terms: Interventional neuroradiology; Sphenoid bone

Percutaneous sclerotherapy by direct puncture is a well-established procedure for some superficial and accessible bone lesions (1, 2). However, the intracranial location of some lesions precludes the use of this approach. We describe an aneurysmal bone cyst of the great wing of the sphenoid bone treated by direct intraoperative sclerotherapy.

Case Report

A 4-year-old boy had a 9-month history of slowly progressive, painless left eyelid ptosis with transient strabismus of the left eye. Neuroophthalmologic examination revealed a mild proptosis. Those findings and an afferent pupillary defect and paralysis of the inward gaze of the left eye indicated involvement of the second and third left cranial nerves. All other neurologic findings were normal, and the boy was otherwise in good health.

Computed tomography (CT) showed a 2×3.5 -cm bone lesion arising from the left-sided great wing of the sphenoid bone. There were peripheral calcifications, causing mild enlargement of the superior orbital fissure (Fig 1A). The lesion was isointense on T1-weighted magnetic resonance (MR) images and hyperintense on T2-weighted images; however, it was hypointense centrally on both images. Parts of the lesion were enhanced after administration of contrast material.

A left-sided external carotid arteriogram revealed opacification of the lesion by posterior branches of the internal maxillary artery. Staining of the lesion was delayed and prolonged in the venous phase (Fig 1B).

Preoperative arterial embolization was undertaken with polyvinyl alcohol and gelatin sponge particles (Gelfoam, Upjohn Co, Kalamazoo, Mich) after superselective catheterization of the left internal maxillary artery. A left-sided frontotemporal craniotomy was done the next day. Surgical exploration revealed a highly vascularized, pulsating lesion that was varicose and purple in appearance. The adjacent bone and dura were highly vascularized. Because of a very high risk of hemorrhage, no resection or biopsy was attempted. Baseline angiography 3 weeks after the arterial embolization showed no change in the lesion.

One month later, a second left-sided frontotemporal craniectomy was performed to expose the lesion for direct puncture with a 20-gauge Jelco (Critikon, Tampa, Fla) intravenous catheter. Water-soluble contrast material was injected to confirm the absence of any dangerous venous drainage. The volume of the lesion was evaluated at 4 mL before opacification of the draining veins (Fig 1C). Subsequently, without moving the needle, a mixture of 2 mL of histoacryl *N*-butyl cyanoacrylate (Histoacryl; B. Braun, Melsungen, Germany) and 2 mL of iodized oil corresponding to the volume of the lesion were injected into the lesion under fluoroscopic guidance. The slow and diffuse bleeding from the lesion stopped, and no further bleeding occurred when the catheter was withdrawn. Because surgical access to the totality of the lesion was difficult, no resection was undertaken.

Postoperative arteriograms done immediately and at 3 and 16 months after intraoperative direct sclerotherapy showed an 80% decrease in opacification of the volume of the lesion. Postoperative CT scans showed no progression of the lesion. There were no complications. Sixteen months after sclerotherapy, the patient had only residual proptosis without eyelid ptosis or any other neurologic deficits. Two years later, a complete resolution of proptosis was noted, and a CT scan at that time showed complete ossification of the lesion (Fig 1D). Angiography revealed a

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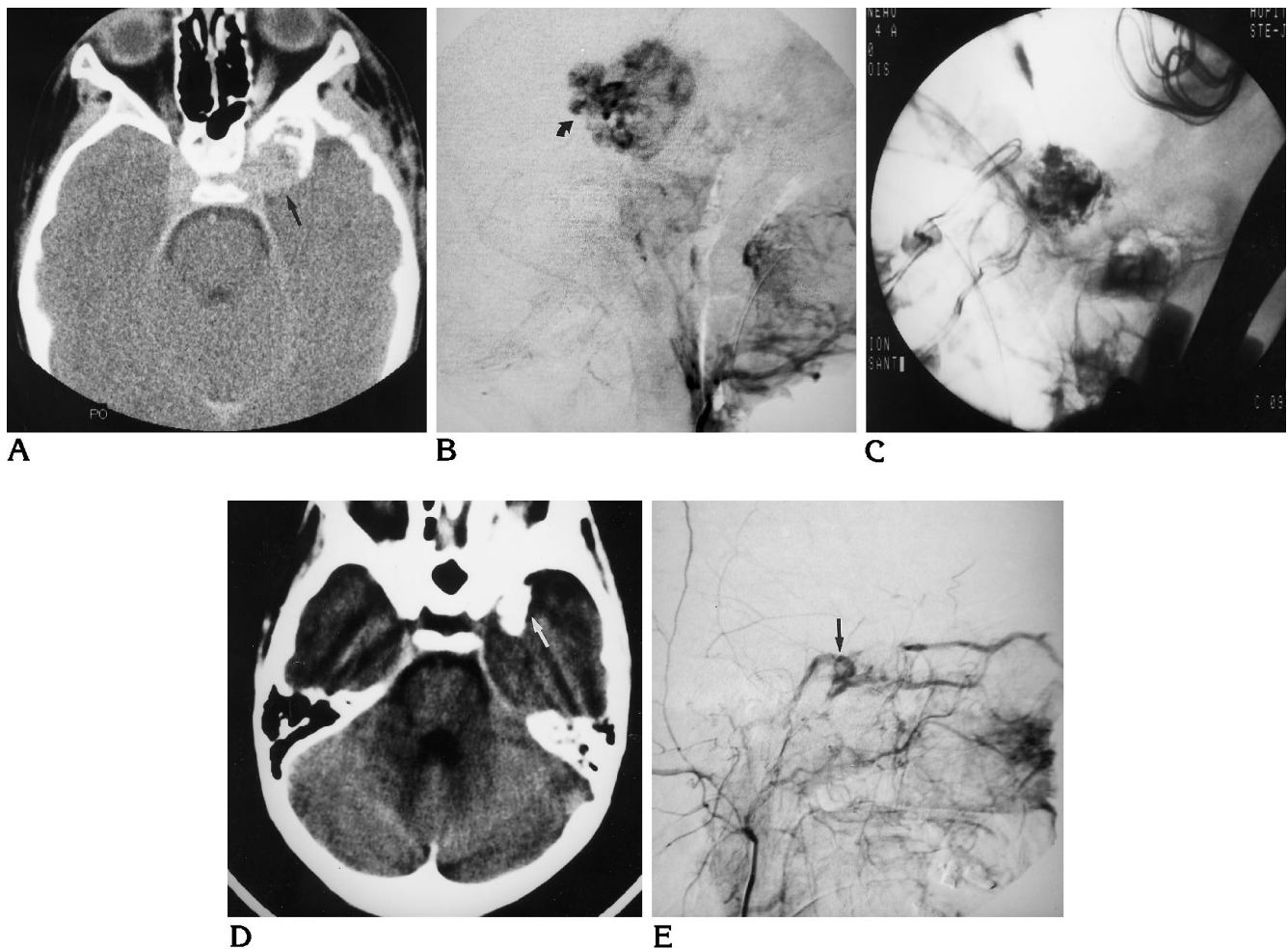


Fig 1. Four-year-old boy with slowly progressive, painless left eyelid ptosis with transient strabismus of the left eye.

A, Contrast-enhanced CT scan shows an expansile sphenoid lesion (*arrow*).

B, Selective catheterization of the external carotid artery shows significant contrast staining of the lesion in the venous phase (*arrow*).

C, Percutaneous phlebography was done with a 20-gauge catheter, which was inserted in the lesion with an injection of water-soluble contrast material.

D, CT scan obtained 2 years after treatment shows complete ossification of the lesion (*arrow*).

E, Angiogram obtained at same time as D shows complete regression of the lesion with only tributary veins of the cavernous sinus (*arrow*).

total regression of the lesion with only prominent cavernous sinus veins (Fig 1E).

Discussion

Because of the very high risk of intracranial bleeding of this type of vascular lesion, neither resection nor biopsy were undertaken, and therefore a histologic diagnosis was not available. The presumed clinical and radiologic diagnosis was an aneurysmal bone cyst. This lesion usually occurs in patients under 20 years of age (3). An aneurysmal bone cyst can be found, although rarely, in the cranial base (4-7). It

usually shows a heterogeneous aspect on CT scans, with a hypodense center, a hyperdense periphery, and partial enhancement. On arteriograms, it appears as a vascularized lesion with a persistent staining in the venous phase, as in our patient.

Direct intratumoral sclerotherapy can achieve better devascularization of the lesion than can arterial embolization, because with angiography, not all the arterial feeders can be embolized (8, 9). We found two cases of direct intraoperative sclerotherapy of intracranial lesions in the literature, a frontal hemangioperi-

cytoma and an intrasellar cavernous hemangioma for which the needle was inserted through a surgical opening (9).

Those two cases and our case demonstrate the feasibility of this technique for intracranial lesions not accessible percutaneously. It permits immediate visualization of the lesion and of its site of puncture; it allows easy detection of any bleeding and permits better control; and it may also diminish or even stop bleeding of the lesion, as in our case, before resection. However, phlebitis is a potential complication. For this reason, the injection must be followed under fluoroscopic guidance in order to avert reflux of the embolic agent into the surrounding veins, especially in a strategic area near the cavernous sinus, as in our case. Direct intraoperative sclerotherapy requires an excellent collaboration between the interventional radiology team and the surgeons.

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