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Ethmoid Mucocele in an Infant with a Benign Fibrous Lesion

Nicholas J. Ferris and Robert D. Tien

Summary: We present a case of ethmoid mucocele occurring in a two-month-old child, and associated (presumably causally) with a benign fibrous lesion of the ethmoid bone. These two conditions may be more frequently associated than previously recognized; MR greatly facilitates their evaluation.

Index terms: Mucocele; Paranasal sinuses, mucocele; Pediatric neuroradiology

Magnetic resonance (MR) has recently been shown to be of great value in the assessment of sinonasal mass lesions, especially in the differentiation of their soft tissue and fluid components, and the demarcation of their margins. The use of MR in this context has led to the suggestion that an association between fibrous lesions of the paranasal sinuses and sinus mucoceles may be more frequent than previously recognized.

Case Report

A 2-month-old female infant was brought to the clinic because of divergent gaze and a "puffy" left eye that appeared smaller than the right eye. The infant had been delivered normally, and the only abnormality detected perinatally was a small Gartner's duct cyst of the vagina. Ophthalmologic examination at 11 weeks of age disclosed a preference for the right eye and limitation of ocular movements (especially abduction and adduction) on the left. The left globe was smaller than the right. Imaging studies were obtained to rule out an orbital mass on the left.

Computed tomography (CT) examination demonstrated an ovoid mass expanding the left posterior ethmoid complex and slightly displacing the globe. This was predominantly hypodense, with a few dense focal calcifications, and some soft-tissue density noted along its medial and anterior margins (Fig 1A and B). A similar soft-tissue density, with some calcifications, was seen to extend intracranially (Fig 1C). Atrophy of the extraocular muscles, and possibly of the optic nerve, was also noted.

On MR, the hypodense ovoid component was seen to be predominantly hyperintense on both T1- and T2-

weighted images and was therefore thought to represent proteinaceous fluid or possibly dilute methemoglobin. The mixed soft-tissue density and calcific areas seen medial and anterior to the ovoid lesion were hypointense on both T1- and T2-weighted images, apart from a small area of moderate T2 intensity anteriorly (Fig 1D and E). These areas enhanced moderately after intravenous gadolinium (Fig 1F). A small area of intracranial extension was again demonstrated (Fig 1G).

Rhinoscopic examination revealed a submucosal mass high in the nasal cavity on the left. Endoscopic operation was performed at the age of 6 months. The ethmoid bulla was found to be prominent; on incision of it, thick, tenacious, clear fluid was obtained. The cystic lesion then was widely opened and a total ethmoidectomy performed.

Histologic examination of biopsies from the lining of the cystic lesion disclosed respiratory epithelium. Examination of tissue fragments resected from the region of the ethmoid bulla showed clusters of irregular trabecular bone, separated by very dense fibrous tissue; the latter replaced the medullary spaces. No mitotic figures were identified. A few islands of cartilage were scattered through the fibrous component of the lesion. The interspersed bony trabeculae showed the "Chinese letter" pattern, and a few osteoblasts were noted on their surfaces. Before and after external pathologic consultation, these findings were interpreted as representing fragments from a benign fibrous lesion.

We concluded that the paraorbital mass represented a mucocele of the left ethmoid complex, secondary to the presence of a benign fibrous lesion, which had presumably obstructed the ostia of one or more ethmoid air cells.

Discussion

The CT and MR features of benign fibrous lesions of the facial bones have recently been reviewed (1). The term *benign fibrous lesion* is used in the pathologic literature to describe a spectrum of lesions ranging from fibrous dysplasia to ossifying fibroma and including the cementifying fibroma of the maxilla and mandible. There appear to be no CT or MR

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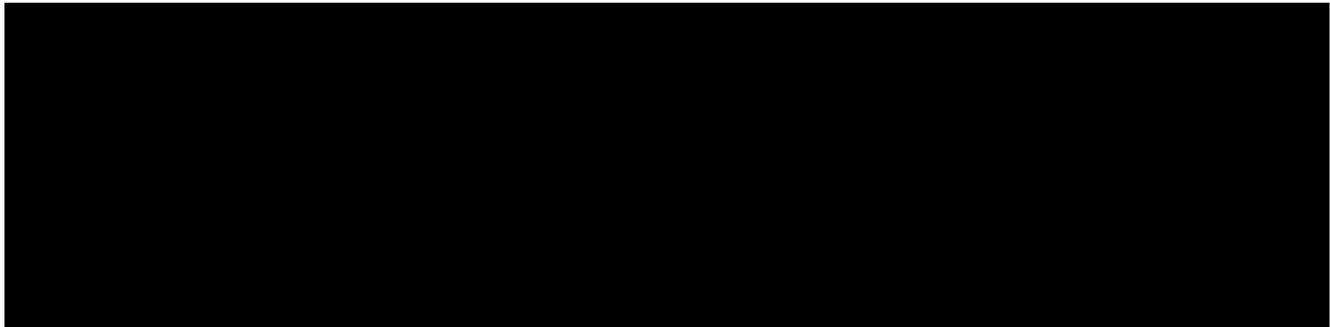
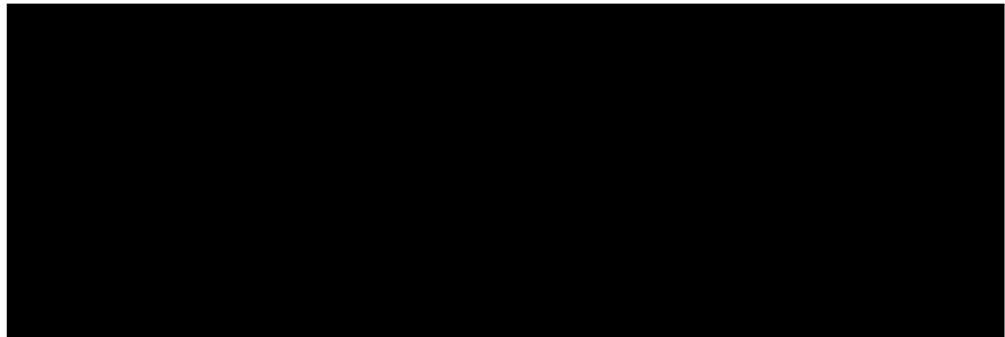


Fig 1. A, Axial CT image through the head, at the level of the inferior margins of the globes. An ovoid mass, of density intermediate between cerebrospinal fluid and soft tissue, is expanding the left ethmoid complex. The medial wall of the left orbit is thinned, and there is slight lateral displacement of the globe.



B, Axial CT image through the head at the level of the ocular lenses, after intravenous contrast, and using the bone algorithm. A low-density ovoid mass again is seen to expand the left ethmoid complex. A single focus of dense calcification is seen anteriorly within the low-density lesion. The medial wall of the left orbit again is seen to be bowed and thinned but does not appear to be transgressed by the lesion. At the anterior and medial margins of the low-density lesion, abnormal amounts of material of soft-tissue density are visible. Anteriorly this material is admixed with spicules of bone, some of which appear thickened. Note atrophy of medial and lateral rectus muscles on the left, along with displacement and possible atrophy of left optic nerve, and dysconjugate gaze.

C, Axial postcontrast CT image at a slightly more superior level demonstrates soft-tissue density material at the superior margin of the lesion partially replacing the bony medial wall of the orbit. Similar material, associated with abnormal spicules of calcification, is seen intracranially in the olfactory groove on the left.

D, Axial T1-weighted MR image (500/20/4 [repetition time/echo time/excitations]) at the level of the inferior margins of the globes. The low-density ovoid lesion seen at CT is seen to contain predominantly hyperintense material, with a poorly defined region of less marked hyperintensity anteriorly within it. Anterior and medial to the ovoid lesion, there is an abnormal amount of T1-hypointense material.

E, Axial T2-weighted MR image (2000/80/0.75) at the same level shows uniformly hyperintense contents in the ovoid lesion, with a thin low-intensity septum running transversely within it. The abnormal material medial to the ovoid lesion remains hypointense, but a small area of intermediate signal intensity is seen anteriorly.

F, Axial T1-weighted MR image (500/20/2 with fat saturation) at the same level, after intravenous gadolinium, shows moderate enhancement of the hypointense material anterior and medial to the ovoid lesion (*arrows*).

G, Parasagittal T1-weighted MR image (500/16/1) again demonstrates the mixed hyperintensity within the ovoid component of the lesion. This is seen to remain inferior to the planum sphenoidale. However, abnormal T1-hypointense material is seen anterior and superior to the ovoid lesion, and the hypointense material is seen to extend intracranially, corresponding to the lesion seen in C. After intravenous gadolinium, this material was seen to enhance moderately (not shown).

criteria that reliably distinguish between these entities (1); all are hypointense on T1-weighted images and hypointense to moderately intense on T2-weighted images. This is in contrast to the frequent finding of marked T2 hyperintensity in fibrous dysplasia elsewhere in the body (2). Craniofacial benign fibrous lesion shows moderate enhancement after intravenous gadolinium. A minority of such lesions contain areas of marked T2 hyperintensity, corresponding to cysts or mucoceles. On CT, these appear hypodense but are often difficult to dis-

tinguish from the soft-tissue component of the fibrous lesion. Som and Lidov (1) suggest that this may in the past have led to underrecognition of mucoceles associated with fibrous dysplasia.

In the present case, the T2 hyperintense component of the lesion was also hyperintense on T1-weighted images, presumably because of proteinaceous material within the fluid (which was thick and tenacious at surgery). The decrease in T1 hyperintensity seen in the anterior part of the lesion may have been caused by a

locally increased concentration of the proteinaceous material; complete signal loss on T1-weighted images has been reported when the secretions within a mucocele are hyperconcentrated (1).

Blood products within the cystic component of a fibroosseous lesion also could be hyperintense on both T1- and T2-weighted images; there is a report of such a finding in association with a psammomatoid ossifying fibroma (3). Psammomatoid ossifying fibroma is also within the spectrum of benign fibroosseous lesions; there is controversy about its potential for recurrence and local destruction (1, 3). Histologically, psammomatoid ossifying fibroma is characterized by the presence of multiple small, densely calcified psammomatoid ossicles (4). One report of three cases imaged with both CT and MR suggested that psammomatoid ossifying fibroma consistently appears multilocular, with septa of bone or soft tissue. The central components of these psammomatoid ossifying fibromas were consistently hypointense on T1-weighted images and hyperintense on T2-weighted images (3). However, ossifying fibromas, not otherwise specified, were indistinguishable radiologically from other forms of benign fibroosseous lesion in the larger study of Som and Lidov (1).

Aneurysmal bone cysts occurring in the paranasal sinuses have been reported to be predominantly hypointense on T1-weighted images and hyperintense on T2-weighted images, but characteristically contain fluid-fluid levels; the dependent layer is hypointense on T2-weighted images (5). Such layering would be very uncommon in conditions other than aneurysmal bone cyst.

Craniopharyngiomas and Rathke's cleft cysts may rarely occur in the region of the sphenoid or ethmoid; the contents of these lesions may be hyperintense on both T1- and T2-weighted MR images. Intrasellar craniopharyngiomas may show a T1- and T2-hypointense component, which enhances after intravenous gadolinium (6), and could thus mimic the imaging findings in the present case.

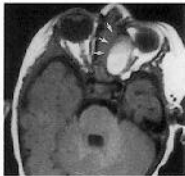
Mucoceles of the paranasal sinuses are rare in infants, presumably because it is unusual for masses to obstruct the ostia of the paranasal

sinuses in infancy. Almost all previously described cases of sinus mucocele occurring before the age of 10 years have been associated with cystic fibrosis (7). Craniofacial fibrous dysplasia also is unusual in infancy (8). Margo et al (4) reviewed a series of 21 cases of psammomatoid ossifying fibroma, including one in a 4-month-old child. The lesions in some of these cases contained cystic components, but the authors do not state whether this was the case in the 4-month-old (from the description given, these cystic components do not appear to have been mucoceles).

MR has important roles both in characterizing sinonasal masses and in defining their extent. The finding of an enhancing mass, hypointense on T1- and T2-weighted images, appears relatively specific for the spectrum of benign fibroosseous lesion. Areas of T2 hyperintensity suggest the presence of cysts or mucoceles. In the case we have presented, the development of a mucocele in an infant, in association with an adjacent benign fibroosseous lesion, strongly suggests that at least some of the cysts and mucoceles now being recognized in association with benign fibroosseous lesion are in fact secondary to the presence of the benign fibroosseous lesion.

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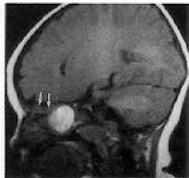
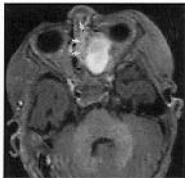


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