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B L Koch, C Myer, 3rd and J C Egelhoff

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Congenital Epulis

Bernadette L. Koch, Charles Myer III, and John C. Egelhoff

Summary: Congenital epulis of the newborn is a rare gingival tumor that occurs along the alveolar ridge. We report the prenatal sonographic and postnatal MR imaging findings in an infant with maxillary and mandibular congenital epulides.

Index terms: Temporomandibular joint, abnormalities and anomalies; Fetus, ultrasound

The term *congenital epulis of the newborn* refers to a rare gingival tumor that most commonly occurs along the alveolar ridge of the maxilla in newborn girls, usually without associated abnormalities of the teeth or additional congenital malformations. A case was described by Neumann in 1871 (1). Since then, multiple cases have been reported, primarily in the pathologic, dental, and otolaryngologic literature (2–4). Two recent articles in the otolaryngologic literature report the in utero sonographic findings of larger lesions. In one case, the patient had hydramnios; in the other case, the authors also describe the postnatal computed tomographic (CT) findings (3, 4).

We describe the prenatal sonographic and postnatal MR imaging findings in an infant with two congenital epulides causing hydramnios and associated maxillary hypoplasia.

Case Report

A 39-year-old woman had prenatal sonography to evaluate hydramnios. Sonograms revealed a 2.9-cm round smooth mass protruding from the mouth or nose of the fetus (Fig 1A). The mass appeared to obstruct the mouth partially with resultant hydramnios. No obvious intracranial extension was identified. Because the patency of the infant's airway at delivery was uncertain, cesarean section was performed at 39 weeks. Before delivery, airway management and postnatal care were thoroughly discussed among the obstetrician, pediatric otolaryngologist, pediatrician, pediatric anesthesiologist, and pediatric plastic

surgeon. The infant was delivered without difficulty, with Apgar score of 8/8/9 at 1, 5, and 10 minutes. The infant required no mechanical respiratory support, as the mass did not obstruct the airway. After delivery, two separate pedunculated gingival masses were noted. One mass arose from the anterior maxillary alveolar ridge and measured 3 cm in greatest dimension; the second mass arose from the anterior mandibular alveolar ridge and measured 2 cm (Fig 1B). The masses were well defined, smooth, and erythematous. The size and position of the masses caused superior deviation of the upper lip. Both nares were patent with passage of an 8F catheter. The nose was flat and the nasal spine was absent, most likely as a result of deformation from the gingival masses. Clinically, the osseous maxillary and mandibular ridges were thought to be normal.

Postnatal MR imaging at 1 day of age revealed lobulated masses that did not extend into the soft palate, floor of the mouth, nose, or cranium. The lower mass indented the tongue slightly but appeared separate from it. The masses were heterogeneous in signal intensity on T1- (Fig 1C), intermediate-, and T2- (Fig 1D) weighted MR images. The unerupted upper teeth appeared normal. The anterior to posterior dimension of the maxilla was decreased, which was thought to be a mild form of midface hypoplasia.

The lesions were excised surgically without difficulty and the patient was tolerating feedings prior to discharge. Histologic examination revealed homogeneous cells with granular eosinophilic cytoplasm and basophilic centrally located nuclei, findings consistent with a congenital epulis.

Discussion

Congenital epulis of the newborn is a rare gingival tumor that occurs on the gum pads of infants. The lesion is most common in females, with a female-to-male ratio of 8:1, and is more common in the maxilla than the mandible (3:1) (2). Although many are single, they may be multiple. The histogenesis is uncertain, and proposed cells of origin include odontogenic epithelium, undifferentiated mesenchymal cells,

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From the Departments of Radiology (B.L.K., J.C.E.), Otolaryngology (C.M.), and Pediatrics (B.L.K., J.C.E.), Children's Hospital Medical Center, and the University of Cincinnati (Ohio) College of Medicine.

Address reprint requests to Bernadette L. Koch, MD, Department of Radiology, Children's Hospital Medical Center, 3333 Burnet Ave, Cincinnati, OH 45229.

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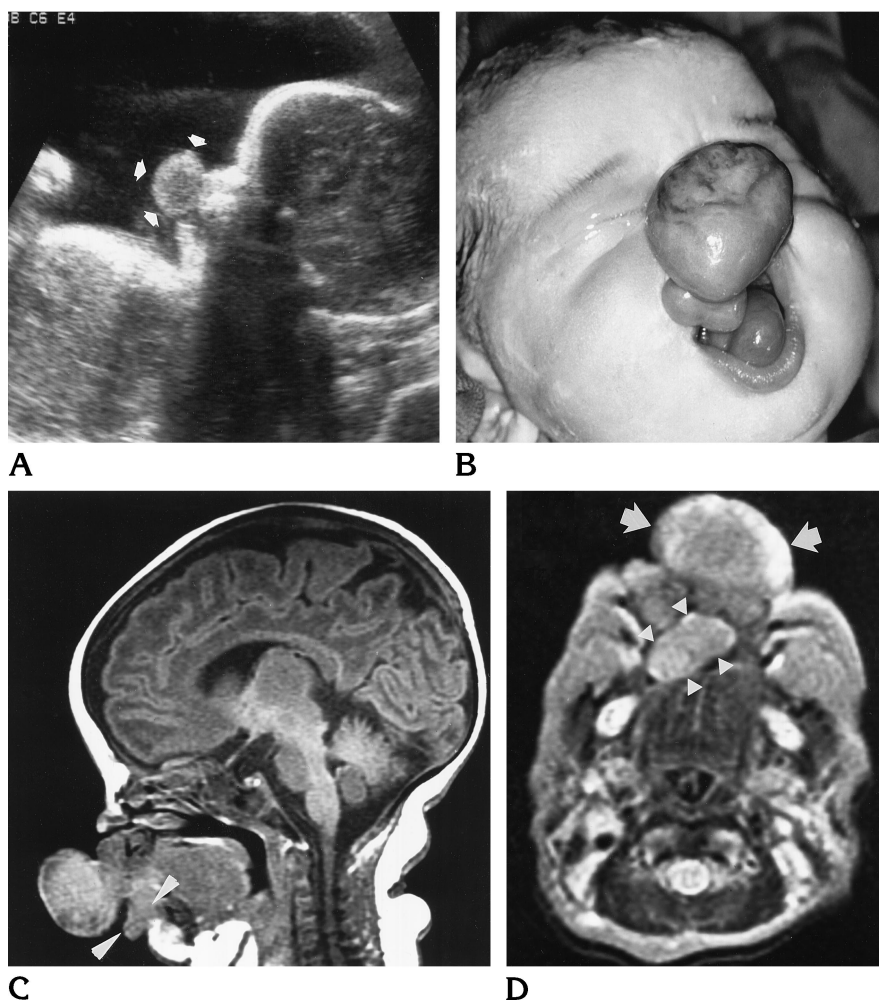
Fig 1. Infant girl with congenital epulis.

A, In utero sonogram shows a well-defined, smooth, round mass (*arrows*), measuring 2.9 cm in greatest dimension, protruding beneath the nose. A small amount of fluid is seen extending into the mouth inferior to the mass. There was associated hydramnios.

B, Postnatal photograph shows two separate paramidline masses, the largest originating from the gum pad of the maxillary ridge. A smaller lesion arose from the mandibular ridge.

C, Sagittal T1-weighted (500/12/4 [repetition time/echo time/excitations]) MR image shows the smooth, well-defined mass arising from the maxillary ridge without extension into the nasal airway, mouth, or cranium. A smaller mass is seen protruding from the mandibular ridge (*arrowheads*).

D, Axial T2-weighted (2500/100/1) MR image shows the maxillary mass as slightly heterogeneous, isointense with muscle centrally, with a peripheral hyperintense rim anteriorly (*arrows*). The smaller mandibular mass is only slightly hyperintense relative to muscle (*arrowheads*).



pericytes, fibroblasts, smooth muscle cells, nerve related cells, and histiocytes (2, 5–9). The reason for the female predominance is peculiar. An endogenous hormonal stimulus has been proposed (10) but this theory is not proved, since detectable estrogen and progesterone receptors within the lesions are lacking (6, 8, 11).

Pathologically, the lesions are composed of large cells with eosinophilic granular cytoplasm within vascular fibrous connective tissue (11–13). Small lesions may regress (14) and larger lesions must be resected, as they often interfere with airway patency and cause feeding difficulties. Complete surgical excision is curative.

The terms *congenital epulis* and *congenital gingival granular cell tumor* have been used synonymously in the literature. Although the congenital epulis is composed of granular cells and is similar to the true granular cell tumor (granular cell myoblastoma), the histology and epidemiology of these two lesions differ. Gran-

ular cell tumors are less vascular, often have a component of pseudoepitheliomatous hyperplasia, and contain more conspicuous nerve bundles than do congenital epulides. Congenital epulides only occur in the gum pads of infants, whereas granular cell tumors usually occur in adults (between 20 and 60 years of age) (11) and may involve multiple organs. Thirty to fifty percent of granular cell tumors occur in the tongue and 30% occur in the skin. Other sites include the supraglottic and infraglottic airways, bronchus, mastoid, orbit, breast, muscle, and lip (12). Less than 1% of granular cell tumors are malignant (13), and some authors argue that those are actually misdiagnosed sarcomas, histiocytomas, and rhabdomyosarcomas (12).

In the literature, congenital epulis is reported to be an isolated finding without associated congenital abnormalities, with the exception of an occasional report of a hypoplastic or absent underlying tooth (15). The patient reported here

also had a mildly deformed upper lip and mild maxillary hypoplasia, probably caused by the mass, which prevented normal growth of the maxilla. The nose was flat and the nasal spine was absent, consistent with "Binder syndrome" (16, 17), most likely resulting from deformation by the gingival masses. Other causes of Binder syndrome include fetal warfarin syndrome, fetal alcohol syndrome, and chondrodysplasia punctata. There was no history of warfarin or significant alcohol ingestion by the mother, and radiographs of the lower extremities were without multiple punctate patellar calcifications, as are seen in chondrodysplasia punctata.

The prenatal hydramnios was caused by partial obstruction of the oral cavity and resultant ineffective swallowing. This is not unexpected in a patient with a mass of this size and has been described previously (4).

In conclusion, congenital epulis is a rare lesion occurring in the gum pads of neonates. It is similar histologically to the more common granular cell tumor that typically occurs in adults; however, unlike granular cell tumors, it does not recur and does not seem to have a particular malignant potential. These lesions have been diagnosed by prenatal sonography in the past, but the prenatal findings are not specific and differential diagnosis would include congenital malformations, such as cephalocele and dermoid, as well as benign and malignant neoplasms, such as lymphatic malformation and rhabdomyosarcoma. The postnatal MR imaging findings of a mass isolated to the gingiva, without involvement of the unerupted teeth, are also nonspecific, and would include other soft-tissue masses, such as fibroma or a vasoformative lesion. The added unsuspected feature that our patient had was midface hypoplasia. We hypothesize that the pathogenesis of this is related to the presence of the large maxillary ridge

mass, deformation of the underlying maxilla, and resultant inhibition of growth.

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