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Sialoblastoma (Embryoma): MR Findings of a Rare Pediatric Salivary Gland Tumor

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Summary: We report the findings in a 21-month-old girl who had a noninfiltrating mass in the left cheek, just anterior to the masseter muscle, which, at surgery, proved to be a sialoblastoma. Sialoblastoma has a histologic appearance reminiscent of a primitive state of salivary gland development; that is, it shows an arrested state of salivary maturation. MR imaging in this case showed that the lesion was isointense with muscle on T1-weighted images, had a high-intermediate signal intensity similar to that of fat on T2-weighted images, and enhanced sparsely and nonhomogeneously.

Index terms: Salivary glands, neoplasms; Children, neoplasms

Tumors that originate in the ductal or secretory epithelial cells of the salivary gland are exceedingly rare in children younger than 2 years of age (1). In 1966, Vawter and Tefft (2) reported two cases in neonates and used the term *embryoma* to describe these tumors that they believed were unique to neonates. In their cases, the location, histology, and histochemistry of the tumors established their parotid origin. Since then, a variety of names, such as *congenital basal cell adenoma*, *basal cell adenoma*, *basaloid adenoma*, and *congenital hybrid basal cell adenoma-adenoid cystic carcinoma*, have been used to describe histologically similar or identical tumors that, retrospectively, are virtually indistinguishable from one another (1). In 1988, Taylor (3) suggested the term *sialoblastoma* to describe these lesions, since it conveyed both the dysontogenetic character and the site of the tumor in a single name; since then, this has become the preferred term. The purpose of this article is to present a case of sialoblastoma that occurred in a 21-month-old girl, and to describe the magnetic resonance (MR) imaging appearance of this rare lesion.

Case Report

The patient was one of premature twins born at 28 weeks' gestation to a 37-year-old mother. At birth, she was noted to have an extensive congenital nevus on the left side of the face involving the scalp, external ear, and neck. In the perinatal period, a noncontrast computed tomographic (CT) scan of the head was obtained at another institution to ascertain whether there was any related intracranial disease. This study was reported to show a left temporoparietal hypodense collection consistent with a small subdural hematoma. No other intracranial abnormality was seen. The patient was followed up clinically, and at age 19 months a contrast-enhanced CT examination of the head was performed. This study showed only the superficial scalp lesion. No salivary mass was identified in the region of the left cheek.

At age 21 months, the patient underwent the first stage of a planned multistage excision of the nevus. Just before this surgery, a nodular, firm, mobile 1- to 2-cm mass was noted in the left cheek. Facial nerve function was intact. A transoral biopsy was performed, and the pathologic diagnosis was that of a sialoblastoma. The patient was then referred to the otolaryngology service. MR imaging, performed to locate the mass better, showed a noninfiltrating nodular tumor with small satellite lesions adjacent to it. These images showed a similar mass to that described on a subsequent MR study.

When the patient was 26 months old, the lesion was surgically removed through a parotid-type incision. At surgery, the distal branches of the facial nerve were identified and were closely adherent to three nodular masses that were just anterior to the parotid gland overlying the masseter muscle. These tumors were removed with preservation of the facial nerve. Pathologic examination confirmed the presence of a sialoblastoma (Fig 1A and B).

Over the ensuing months, we noted the progressive return of an indurated mass just anterior to the parotid gland. Although the facial nerve remained intact, by the age of 26 months, the mass had grown to a size of 4 to 5 cm. A repeat MR study, performed on a 1.5-T scanner,

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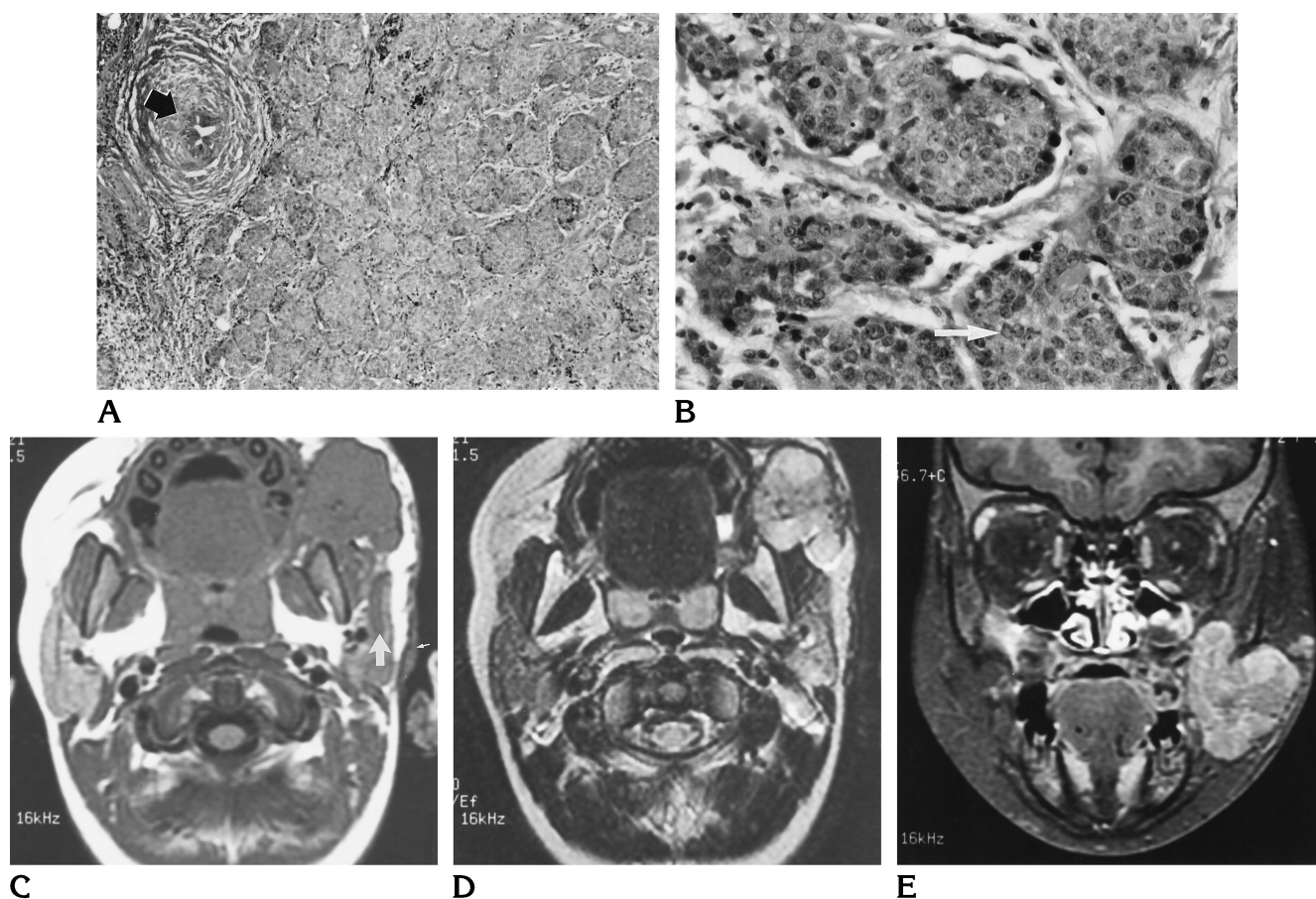


Fig 1. A 26-month-old girl with sialoblastoma.

A, Photomicrograph shows that the tumor forms nests of epithelial cells with an organoid pattern. A residual parotid duct (*arrow*) is identified (hematoxylin-eosin, low-power magnification).

B, Photomicrograph at a higher power than in A shows the organoid pattern formed by moderately pleomorphic cells with angulated nuclei (*arrow*) and prominent nucleoli. Occasional mitotic figures are seen (hematoxylin-eosin, high-power magnification).

Axial T1-weighted (C), T2-weighted (D), and coronal T1-weighted postcontrast fat-suppressed (E) MR images show a lobulated noninfiltrating mass in the left cheek, lying just along the anterior border of the masseter muscle and adjacent to the maxillary alveolus. The mass has a slightly nonhomogeneous low-intermediate signal intensity in C and a slightly higher intermediate signal intensity in D, suggesting a highly cellular mass. On the coronal image, one of the small satellite lesions is seen along the upper border of the main mass. The tumor enhances moderately and nonhomogeneously. In C, there is also dilatation of the main parotid duct (*large arrow*) and thickening of the overlying skin (*small arrow*), which represents the patient's nevus.

showed a lobulated, noninfiltrating mass, approximately 4 cm in greatest dimension, in the left buccal region (Fig 1C-E). The lesion was located along the anterior margin of the masseter muscle, in the area in which accessory parotid gland tissue often occurs. Although the tumor abutted the maxillary alveolus, the ramus of the mandible, the masseter muscle, and the overlying subcutaneous fat, there was no evidence of invasion of these structures. Several smaller (approximately 1-cm diameter) satellite lesions were present in the adjacent subcutaneous tissues. The lesions were isointense with muscle on T1-weighted images (660/15/2 [repetition time/echo time/excitations]); that is, they had a minimally nonhomogeneous low-to-intermediate signal intensity. On T2-weighted im-

ages (4000/99/8), they had a high-intermediate signal intensity, similar to that of fat. The larger mass was nonhomogeneous and appeared to be divided into lobules. After administration of contrast material, the lesion showed sparse, nonhomogeneous enhancement. The mass obstructed the proximal Stensen's duct and the main intraglandular duct. The impression was that of a benign or low-grade cellular tumor. Surgery was again performed at age 31 months and the lesion was grossly removed with a wide excision. No attempt was made to preserve the facial nerve, and the oral mucosa at the previous biopsy site was removed en bloc with the tumor. There was resultant loss of the buccal division of the facial nerve. The patient has done well in the 8 months since surgery.

Discussion

Fewer than 5% of all salivary gland tumors occur in children, and for the epithelial neoplasms, most are discovered in the second decade of life (4). Thus, in the first decade, epithelial neoplasms are often considered curiosities (4); we found 20 reported cases of such epithelial salivary gland neoplasms (1-9). Of these 20 lesions, 19 had a known site of origin; 15 of these arose in the parotid gland and four developed in the submandibular gland. Five (25%) of the 20 were malignant (three patients died of poorly differentiated carcinomas and two patients survived after surgery for basaloid carcinomas with nerve invasion). Of the 15 benign cases, one was a hamartoma and one or two may have been diagnosed as adenomas. The remaining 12 or 13 cases appear to be classifiable as sialoblastomas (7).

Histopathologically, Batsakis et al (4, 5) categorized the perinatal salivary gland tumors into four groups. The first group includes the histologically benign tumors that have adult counterparts (such as pleomorphic and monomorphic adenomas). This group of neoplasms is reported to be the least common of the perinatal salivary gland tumors. The second group comprises the hamartomatous tumors. In the third group are the lesions that "histologically are evocative of the embryonic epithelial anlage of the major salivary glands at various stages of its branching morphogenesis and cytodifferentiation" (4). These tumors have a histologic appearance similar to that of the epithelial salivary gland anlage; that is, they appear as "organoblasts" that are in a primitive state of organ development and in arrested maturation (4, 5). These anlage-type tumors are the most common of the perinatal lesions and are referred to primarily as either sialoblastomas or embryomas. The last group contains the malignant salivary gland tumors.

The term *embryoma* evokes a tumor that appears to arise in the course of embryogenesis. However, it does not distinguish this tumor from embryomas that arise at other anatomic sites and the term resembles *dysembryoma*, a word used synonymously with the unrelated *teratoma* (6). By comparison, Taylor (3) argued that congenital salivary gland tumors originate from the cells of a primitive blastema and these lesions are analogous to blastomas that arise in other organs and that are capable of differenti-

ation along more than one cell line. His hypothesis would account for the mixture of cell types observed and reported in the literature and the overall similarities within the group. He suggested the terms *benign* and *malignant sialoblastoma*, depending on the histology, cytology, and clinical features.

The sialoblastomas are often mitotically active, primitive cell masses with formative ducts and pseudoductular spaces without acinar differentiation. Their appearance recalls the pre-acinar stages of morphogenesis. There is a loose investing mesenchyma with an embryonic appearance, which completes the embryonic nature of the tumor. The cells (organoblasts), although committed to the development of salivary tissue, remain in a primitive state of organ development and are in maturational arrest. Surgery is the treatment of choice without irradiation or chemotherapy. A review of the literature on sialoblastomas showed that a few cases recurred locally and only one case had regional lymph node involvement. No distant metastasis has been reported (1, 4). It is not clear from the limited literature why these lesions tend to have local recurrences; however, while they are considered locally aggressive, they are not thought to be malignant.

In at least one instance, the histologic, immunohistochemical, and ultrastructural features of a sialoblastoma were studied. In this case, the tumor was characterized by solid nests of epithelial cells intermingled with proliferating ductal structures that were lined by a double layer of cells. Immunoperoxidase staining showed the presence of cytokeratins in the ductal cells as well as the presence of vimentin, actin, and S-100 protein in the outermost layer of the ducts. The solid nests of cells were focally reactive to S-100 and vimentin. Ultrastructural studies showed myoepithelial cells with replication of basement membrane material (1).

Compared with the sialoblastomas, the hamartomas are more advanced in their morphogenesis and cytogenesis, and the embryonic quality of the differentiating cells is no longer evident. There are also acini present.

Although the majority of the reported cases of sialoblastoma have occurred in the neonatal period, the case presented here was first identified at age 21 months. Whether there was subclinical tumor before this time cannot be evaluated. Thus, this case may represent an unusual presentation of a rare tumor.

References

1. Hsueh C, Gonzalez-Crussi F. Sialoblastoma: a case report and review of the literature on congenital epithelial tumors of salivary gland origin. *Pediatr Pathol* 1992;12:205-214
2. Vawter GF, Tefft M. Congenital tumors of the parotid gland. *Arch Pathol* 1966;82:242-245
3. Taylor GP. Congenital epithelial tumor of the parotid-sialoblastoma. *Pediatr Pathol* 1988;8:447-452
4. Batsakis JG, Frankenthaler R. Embryoma (sialoblastoma) of salivary glands. *Ann Otol Rhinol Laryngol* 1992;101:958-960
5. Batsakis JG, Mackay B, Ryka AF, Seifert RW. Perinatal salivary gland tumours (embryoma). *J Laryngol Otol* 1988;102:1007-1011
6. Harris MD, McKeever P, Robertson JM. Congenital tumours of the salivary gland: a case report and review. *Histopathology* 1990;17:155-157
7. Luna MA, Batsakis JG, El-Naggar AK. Salivary gland tumors in children. *Ann Otol Rhinol Laryngol* 1991;100:869-871
8. Roth A, Micheau C. Embryoma (or embryonal tumor) of the parotid gland: report of two cases. *Pediatric Pathol* 1986;5:9-15
9. Ito M, Nakagawa A, Nakayama A, Uno Y, Takahashi I, Asai J. Undifferentiated carcinoma of the parotid gland in a 10-month-old child. *Acta Pathol Japan* 1990;40:149-152