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Original Contributions

Sialoblastoma: a clinicopathologic and immunohistochemical study of 7 cases☆

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Abstract Sialoblastoma is a rare congenital or perinatal salivary tumor that varies in histologic features and biologic potential. Seven cases from the files of the Armed Forces Institute of Pathology are presented. These tumors occurred in 4 males and 3 females with ages ranging from prenatal to 6 months at the time of discovery. Five lesions originated from the parotid gland; 2 lesions were from the submandibular gland. All lesions presented as nodular to multinodular swellings and ranged in size from 2.0 to 7.0 cm. The principal sign or symptom was rapid growth. Two histologic patterns with differing behavior predominated: (1) a favorable pattern had semiencapsulation of cytologically benign basaloid tumor cells with intervening stroma; and (2) an unfavorable histology of anaplastic basaloid tumor cells, minimal stroma, and broad pushing to infiltrative periphery. Four and three tumors had favorable and unfavorable growth patterns, respectively. One unfavorable lesion had vascular invasion, and another demonstrated perineural invasion. All 3 tumors with unfavorable histology recurred. Tumor cells in 3 cases were immunohistochemically reactive for keratin, S-100, smooth muscle actin, and calponin to varying degrees. All 3 tumors were reactive for p63. α-Fetoprotein was expressed in 2 unfavorable tumors. Ki67 was expressed at 3% in a favorable tumor and 40% and 80% in the 2 unfavorable lesions. Treatment involved surgical excision. One patient received adjuvant chemotherapy. Two sialoblastomas resulted in recurrences within a year and another developed a recurrence after 4 years. One sialoblastoma developed lung metastasis within 1 month of the original biopsy. Although a clinical correlation is suggested by a favorable/unfavorable histologic grading system the biologic behavior is nonetheless considered unpredictable.

Keywords: Sialoblastoma; Embryoma; Salivary; Parotid; Basaloid myoepithelial neoplasm; Immunohistochemistry; Congenital

1. Introduction

Sialoblastomas are rare salivary gland tumors that arise perinatally and are composed of varying amounts of basaloid epithelial and stromal components with disparate grades in histomorphology. From the report by Vawter and Tefft [1] in 1966 to the recent report by Ozdemir et al [2] of a sialoblastoma, 30 tumors matching the clinical and histopathologic features of sialoblastoma have been described. However, given reports of other similar congenital basaloid tumors, the number of sialoblastomas may be higher [3-5]. Synonyms have included embryoma, basaloid adenocarcinoma, and congenital basal cell adenoma. Cases of sialoblastoma have included histologically benign or malignant adult-equivalent salivary neoplasms and histologically malignant basaloid, undifferentiated tumors. In addition, the rarity of sialoblastomas has made comparative prognostic analysis challenging. A review of clinicopatho-

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Table 1
Immunohistochemical reagents

Immunohistochemistry	Dilution	Source	Company
Pancytokeratin cocktail	1:400	Mouse cytokeratins, mouse LP34	DAKO (Carpinteria, Calif), Chemicon (A1/A3; Temecula, Calif)
EMA	Predilute	Mouse	Ventana (Tuscon, Ariz)
CK7	1:400	Monoclonal mouse	DAKO
CK5/6	1:20	Monoclonal mouse	DAKO
Cam5.2	Predilute	Monoclonal mouse	Ventana
CK20	1:200	Monoclonal mouse	DAKO
K903	1:50	Monoclonal mouse	DAKO
AFP	1:320	Polyclonal rabbit	DAKO
Calponin	1:400	Monoclonal mouse	DAKO
SMA	1:1600	Mouse, clone 1A4	Sigma (St Louis, Mo)
GFAP	1:4000	Rabbit (poly)	DAKO
S-100 protein	1:600	Mouse	DAKO
bcl-2	1:500	Mouse/monoclonal clone 124	DAKO
Ki67	1:80	Mouse, clone MIB (012X101)	DAKO
p53	1:40	PAB1801, mouse	NovoCastra (Burlingame, Calif)
p63	1:100	Mouse, clone 4A4	Santa Cruz Biotech (Santa Cruz, Calif)

EMA indicates epithelial membrane antigen; SMA, smooth muscle actin; APF, α-fetoprotein.

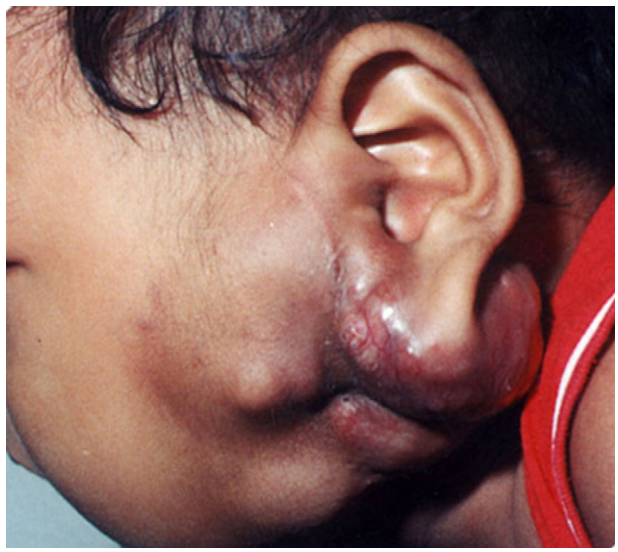

Fig. 1. Clinical rapid growth defined the multinodular deformation of this unfavorable sialoblastoma.

logic and immunohistochemical findings from a series of 7 cases retrieved from the files of the Armed Forces Institute of Pathology is analyzed to enhance our understanding of sialoblastoma. Along with a discussion of the pathogenesis

Table 2
Clinicopathologic features of 7 sialoblastomas

Case	Patient age	Sex	Tumor size/tumor duration	Location	Outside diagnosis	Metastases/ work-up/treatment	Follow-up
1	10 d	F	6 cm/PAB	R Parotid	Cellular mixed tumor	removed at autopsy - secondary finding	Died at 10 days unrelated to sialoblastoma
2	7 d	M	3 cm/PAB	L submandibular	Basal cell embryoma	Excision	A//W 43 years
3	18 mo (noticed at 5 mo; initial excision at 18 mo)	M	6 cm/ (after 13 mo duration)	Left parotid	Sebaceous carcinoma	Nov 68, excision Jan 69, metastasis to submandibular nodes Feb 69, 3 courses chemotherapy—vincristine, cytox, methotrexate, Apr 69, 5-FU, XRT, 5000 rad Nov 69 pulmonary metastasis	Last follow—1973 —A/W, then LFU
4	2 mo, 12 mo	M	2 cm, 5 cm	Parotid	Adenocarcinoma, NOS; adenocarcinoma, NOS	Excision, reexcision	No recurrence, status post 3 y, LFU
5 (reported by Hsueh and Gonzalez-Crussi [6])	PAB, 24 d, 17 mo	F	1 cm, 2 cm; 1.5 cm, recurr	R parotid	Salivary adenoma, ductal type (Gonzalez-Crussi), basal cell adenoma (Armed Forces Institute of Pathology) later described as sialoblastoma. Recurrent, as above	Excision, re-excision	NED status post reexcision, A/W 3 1/2 y
6	15 (tumor noted at 6 mo)	F	5 cm/9 mo after first noted)	R submandibular	Hybrid monomorphic adenoma, adenoid cystic carcinoma	Excision, no recurrence	A/W 14 y
7	PAB, 5 mo, 11 mo	M	5 cm 6; cm, recurr	L parotid	Carcinoma, undifferentiated; sialoblastoma	Excision, reexcision	LFU

PAB indicates present at birth; A/W, alive and well; LFU, lost to follow-up; 5-FU, 5-fluorouracil; XRT, x-ray therapy.