Review Article

Sialoblastoma: A literature review from 1966-2011

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ABSTRACT

Sialoblastoma is a rare congenital tumour of the salivary glands arising mainly from the parotid gland. It is usually diagnosed at birth or shortly thereafter with a significant variability in histological appearance and clinical course. In extensive search of PubMed indexed journals, we got 46 cases of "sialobalstoma/embryoma/congenital basal adenoma", with one case was of German literature and three additional cases of adult sialobalstoma. This article has extensively reviewed the clinical, histopathological and immunohistochemical features, Magnetic resonance imaging (MRI) and Computerized Tomography (CT) findings, treatment and prognosis.

Key words: Congenital basal cell adenoma, embryoma, sialoblastoma

Introduction

Tumours that originate in the ductal or secretory epithelial cells of salivary gland are exceedingly rare in children < 2 years of age.[1] A group of tumours has been recognized those usually presents at birth or shortly thereafter, and are composed of basaloid and myoepithelial cells that recapitulate the developing salivary anlage. These tumours have been reported under a variety of names such as congenital basal cell adenoma, basal cell adenoma, basaloid adenocarcinoma, and congenital hybrid basal cell adenoma-adenoid cystic carcinoma, etc.[2] Vawter and Tefft[3] suggested the term embryoma in 1966 for this unique perinatal tumour. Alternatively in 1988, Taylor[4] suggested the term sialoblastoma, because it conveyed both the dysontogenetic character as well as the tumour site within a single name. In extensive PubMed search with keywords "sialoblastoma, embryoma, congenital basal cell adenoma". We have found out total of 46 cases with these key words. Most of reported cases are single case report. In addition three cases of adult sialoblastoma were identified. We have reviewed 40 cases tabulated by Saffari *et al*.[5] in 2011 [Table 1]. Additional 6 cases we have identified (2010-2011) in literature so all together we reviewed 46 cases of sialoblastoma/embryoma/congenital basal cell adenoma [Table 2]. 3 adult sialoblastoma[6] cases were also identified and reviewed [Table 2].

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Discussion

Epidemiology

Most tumours have been diagnosed at birth; cases from 34 week of fetus to four year of age have been identified in English language literature. [Tables 1 and 2] A Case of 7 year/F reported in German literature.[7] 29 cases (63.03%) were of less than 10 days of age. Male 22, female 23, almost equally affected. In one case sex has not been mentioned. 31 tumour were related to parotid,[3,7-29] 11 submandibular,[12,18,30-36] two cheek minor salivary gland,[5,37] one was in relation with eyelid minor salivary gland[38] and one presented as face and neck mass[39] [Tables 1 and 2]

Clinical features

Clinically most of babies presented as cheek and submandibular mass. The reported size of tumor ranges from a peanut to a maximum of 15 cm in diameter.[28,39] At one occasion it was associated with superficial haemorrhage and necrosis.[39] The clinical

Table 1: Congenital sialoblastoma/embryoma/basal cell adenoma[5]

Year*	References	Age/sex	Location	Follow-up	Remarks
2009	[23]	34 w M	R parotid	Died of septicemia	Associated with hepatoblastoma
2008	[31]	3 mo M	R submandibular	FT for 13 mo	Resembles lymph node
2008	[37]	37 w F	R minor salivary gland of the cheek	FT for 6 mo	Resembles hemangioma
2008	[32]	Term F R	submandibular	FT for 12 mo	Early surgical intervention
2008	[24]	4 y F	L parotid	FT for 6 mo after chemotherapy and surgery	Associated with a cutaneous hamartoma, lung metastasis
2007	[38]	18 mo	R minor salivary gland of the eyelid	FT 1 y after surgery/3 mo after chemo	Sarcomatoid differentiation of the myoepithelial component
2006	[18]	10 d F	R parotid	Died, unrelated to Sialoblastoma	Secondary finding
		7 d M	L submandibular	FT for 43 y	Basal cell adenoma
		18 mo M	L parotid	FT for 4 y after therapy	Metastasis to lymph Nodes
		2 mo M	Parotid	FT for 3 y	Recurrence/re-exision
		6 mo	R submandibular	FT for 14 y	Hybridmonomorphic/adenoid cystic carcinoma
		5 mo M	L parotid	NA	Recurrence/re-exision
2006	[33]	15 mo M	L submandibular	FT 1 y	Recurrence
2006	[8]	4 y F	L parotid	Died due to respiratory insufficiency	Local invasion
2005	[25]	37 w F	R parotid	FT for 6 mo	High AFP
2004	[9]	Term M	R parotid	FT for 8 mo	MRI findings
2003	[10]	21 mo F	R parotid	Multiple recurrence with lung met	Cytology findings of the metastatic lung nodule
2001	[49]	3 mo M	R Parotid	FT for 6 mo	Dysembrygenic alteration of adjacent parotid
2000	[34]	35 w M	R submandibular	FT for 2 y	Surgical technique
2000	[36]	Term M	R submandibular	FT for 12 mo	Surgical management
2000	[35]	Term M R	submandibular	FT for 6 mo	Associated with cutaneous hamartoma
2000	[11]	37 w	F R parotid	FT for 5.5 y	Associated with hepatoblastoma
1999	[15]	Term F	R parotid	FT for 1 y	Early conservative surgical approach
1999	[13]	21mo F	L parotid	FT for 1 y	Clinicopathology and immunohistochemistry study
1998	[7]	7 y F	Parotid article in Germany		Juvenile pleomorphic adenoma
1997	[14]	21mo F	L parotid	FT for 8 mo	Late presentation associated with congenital nevous
1997	[12]	Term M	R parotid	NA	Basal cell adenoma
		Term F	R submandibular	NA	Basal cell adenoma
		Term M	L parotid	NA	Basal cell adenoma
1992	[16]	38 w F	L parotid	FT for 3 y and 1/2 mo	Lymph node met
1990	[30]	Term F	R submandibular	FT for 15 mo	Suggesting the name of sialoblastoma
1988	[4]	41 w F	L parotid	Two further recurrence following 30 mo	Tumor recurrence
1994	[19]				
1989	[20]	Term F	R parotid	NA	Basal adenoma with recurrence
1988	[17]	37 w M	L parotid	FT for 3 w	Primary diagnosis: Embryoma
1986	[21]	Term M Term M	L parotid L parotid	FT for 10 y FT for 9 y	Ambiguity of the term embryoma
1980	[22]	Term M	R submandibular	FT for18 mo after surgery	Basal cell adenoma of the submandibular gland
1972	[50]	7 d M	R parotid	FT for 4 y	Primary diagnosis: Adenoma
1966	[3]	Term M	R parotid	FT for 12 y after recurrence	Primary diagnosis: Embryoma

FT: Free of tumour, NA: Not available

diagnosis in one of the case was found to be Hemangioma.[37] In two of the reported cases, associated hepatoblstoma with increased level of Alfa-Feto Protein (AFP),[11,23] was found and another, two cases have associated cutaneous hamartoma[24,35] and one with congenital nevus has also been reported.[14] Ozdemir *et al*.[25] reported a case of congenital sialoblastoma presenting with the premature centromere division (PCD) and a high level of AFP, which associations have not previously been reported. After resection of tumour AFP level decrease to normal. Their case was the first sialoblastoma associated with high levels of AFP. The principal site of synthesis of AFP is the fetal liver. In the human fetus, the level of AFP falls with increasing maturation.[40] Elevated values of AFP in the neonatal period have been detected in malignant germ cell neoplasms, massive hepatocellular carcinoma, and hereditary tyrosinemia type I.[41,42] Siddiqi *et al*.[11] reported a case of sialoblastoma and hepatoblastoma in a neonate. The liver of the case reported by Ozdemir *et al*.[25] was examined by ultrasound and magnetic resonance imaging and there was no finding of hepatocellular carcinoma.

MRI and CT findings

Imaging features of sialoblastoma have been documented in a limited number of cases.[11,14] The CT appearance is of a soft-tissue mass hypodense to the brain and isodense to muscle. Som *et al*.[14] detected low–intermediate signal intensity and slightly higher intermediate signal intensity

Table 2: Cases of paediatric and adult sialoblastoma 2010-2011, 2010* adult SB

Year	References	Age/sex	Site	Size (cm)	Treatment	Recurrence/Rx	Metastasis	Histological diagnosis	Follow up DF (Disease free)
2011	[5]	AT Birth, FM	R cheek	2 × 2 cm, within 5 days size increase up to 4.5 × 4.5 cm	Surgical excision	No recurrence	No	SB	7 Month DF
2011	[29]	3 Mo FM	L parotid	Not mentioned	3.5 year age chemo	After 1.5 year, orbital recurrence, total resection, adj radio	Age of 6.5, At 6.5 years of age, metastasis to the right lung. Metastasis to the left lung two years later, 6 months later, third pulmonary recurrence in the right upper lobe	SB	7 year DF
2010	[28]	18 Mo/FM	L parotid	Size increase from peanut to table tennis ball in 1 month (3 × 3 × 3)	Surgical excision	Aft 6 month, 125 I seed implant brachytherapy, complete response	No	SB	21 month DF
2010	[26]	12 Mo/M	R cheek	H/O swelling since birth, operated two month before, noBx Report, swelling gradually increasing	Surgical excision	Radical surgey, with chemo, recurrence time not mentioned	No	SB	12 month DF
2010	[27]	At Birth/F	L Parotid	FNA diagnosis was PA, spontaneous regression after FNA, 4 year later 3 cm mass	Surgical excision	1 year later recurred (6 × 7 × 7)	Rt and Lt lung met, chemo, complete response	SB	12 months DF
2010	[39]	3 days/M	L face and neck mass	15 × 10 × 8 (superficial haemorrhage necrosis)	Sx, margin + ve	Recurrence after 3 month, treated with chemotherapy	No		4 year
2010* 1	[6]	46 year/ FM	R Parotid	3 × 2 cm six month duration	Sx	15 year later lump in surgical scar, after 5 treated with Sx and radio	No		7 year DF
2		83/M	Palate	2.5 × 2.5 cm, exact duration did not mentioned		Incision biopsy	Tumor did not progress till last 8 month, surgery avoided due to patient factors		
3		55/M	Palate	No clinical data			No follow up detail		

*Year of publication, FT: Free of tumour, NA: Not available, Ref: Reference, M: Male, F: Female, R: Right, L: Left, w: Week, mo: Month, Sx: Surgical excision , DF: Disease free, SB: Sialoblastoma

on T1-weighted (T1-W) and T2-weighted (T2-W) images, respectively. In a case reported by Yekeler *et al*.[9] the greater part of the lesion excluding necrotic and haemorrhagic areas was mildly hyper intense on T2-W images, which was lower than that described by Som *et al*.[14] The finding of mild hyperintensity on T2-W images suggests a high nucleus/cytoplasmic ratio belonging to blastoma and can be predictive for the diagnosis of blastomas. In their case,[14] the cause of intralesional haemorrhage on MRI was unclear. It could have occurred spontaneously into the fragile tumour tissue or have resulted from minor trauma during vaginal delivery.

Histopathological features

The morphology of sialoblastoma is very characteristic with the presence of nests of basaloid cells with a palisading pattern at the periphery and maturation toward the centre. Sialoblastomas may have admixed histologic appearance, ranging from benign hamartomatous lesions with marked similarity to normal fetal salivary gland tissue to highly malignant tumour.[16] Batsakis and colleague[17] proposed histologic criteria for assessment of malignancy in a sialoblastoma, which included "invasion of nerves or vascular spaces and ancillary findings of necrosis and cytological atypia beyond that expected or presumed for an embryonic epithelium. Mitotic figures are variable, 3-4/High Power Field (HPF), and 6-7/10 HPF to 20/10 HPF has been reported in recurrent cases.[13] Areas of necrosis also increased in recurrent cases[13] necrosis with focal calcification in isolated cases[34] and necrosis with total tumour replacement of tumour stroma after chemotherapy.[28] Proliferative index from 3cell/10 HPF to 94 cell/10 HPF has been reported.[13]

Williums *et al*.,[18] studied clinicopathological and immunohistochemical features of seven cases from the files of the Armed Forces Institute of Pathology. According to their study there is a male to female ratio of 4:3 and age ranging from prenatal to six 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 behaviour predominated: (1) A favourable pattern had semi encapsulation of cytologically benign basaloid tumour cells with intervening stroma; and (2) an unfavourable histology of anaplastic basaloid tumour cells, minimal stroma, and broad pushing to infiltrative periphery. Four and three tumours had favourable and unfavourable growth patterns, respectively. One unfavourable lesion had vascular invasion, and another demonstrated perineural invasion. All three tumours with unfavourable histology recurred. Tumour cells in three cases were immunohistochemically reactive for keratin, S-100, smooth muscle actin, and calponin to varying degrees. All three tumours were reactive for p63. AFP was expressed in two unfavourable tumours. AFP positivity also reported by xx. Ki67 was expressed at 3% in a favourable tumour and 40 and 80% in the two unfavourable lesions. Yasi *et al*.[5] has reported Ki-67 (30%) in their case. As a prognostic marker, Ki67 immunostaining demonstrated expected findings of a lower proliferative index in the favourable tumour in one case as opposed to significantly higher indices in unfavourable tumours two cases. In the case study by Brandwein *et al*.[13] Ki67 developed as a significant finding in the course of the recurrences of tumour. The incorporation of Ki67 index with the favourable/unfavourable histologic indicator may be useful for the prognosis of these tumours. So immunohistochemical these tumour express S-100 and vimentin diffusely. Cytokeratin accentuated the ductal structure.[2]

Recurrence and metastasis

10 cases recurred after initial treatment.[3,4,16,26,28,29] Most of them recurred within 24 month. Four cases of lung metastasis with two cases have multiple lung metastasis and two cases of cervical lymph node metastasis have been reported.[10,27,29] In reported cases no case succumbed to death due to recurrence or metastasis [Tables 1 and 2].

Treatment

Various treatment modalities like surgical excision, chemotherapy, radiotherapy and combination for primaries, recurrent and metastatic cases have been mentioned. One case spontaneously regresses after FNAC till one year than increased in size, treated with superficial parotidectomy and recurred. After metastasis to lung treated with chemotherapy and got complete response.[27] One case treated with 125 I brachytherapy,[28] cured completely [Table 1 and 2].

Prognosis

Only three cases succumbed to death; due to septicaemia,[23] respiratory unsuffeciency[8] and unrelated to sialoblastoma,[18] one in each category. In reported literature maximum of 43 year of disease free survival has been reported.[18] In most of cases functional and aesthetic efficacy has been achieved.

Cytogenetics

Ozdemir *et al*.[25] reported a case of congenital sialoblastoma presenting with the PCD and a high level of AFP, which associations have not previously been reported.

Adult sialoblastoma

Essentially, sialoblastoma is a disease of infancy with the oldest case presenting at four and seven year of age in English and German literature respectively [Tables 1 and 2]. About one third of pediatric sialoblastoma cases will have a cribriform growth pattern. No adult cases have been reported with a specific diagnosis of sialoblastoma. If even focal cribriforming were present, such cases have undoubtedly been diagnosed as adenoid cystic carcinoma.[6] Such was the circumstance in the three adult tumours presented by Dardic *et al*.[6] Each case, however, has the primitive histopathology with discrete nests of basaloid tumour cells, associated bilayered ductal structures and the fibromyxoid stroma characteristic for sialoblastoma with its resemblance to fetal salivary gland or salivary gland with arrested development. Sialoblastoma, whether in a child or adult with or without a cribriform growth pattern, appears to have a more favourable prognosis than adenoid cystic carcinoma. Detailed data mentioned in Table 1 (2010 * 1, 2 and 3).

In adult sialoblastoma cases, cribriform histology needs to be tempered with the overall primitive organization of the basaloid tumor cells, often with associated single or branching ductal structures, enclosed by loose collagenous to myxoid stroma. The latter aspects produce a likeness to developing salivary gland in the fetus and, as a hallmark of sialoblastoma, are an essential diagnostic feature. It is notable that sialoblastomas are generally circumscribed and even partially encapsulated. A cribriform growth pattern does not necessarily imply adenoid cystic carcinoma with its inherently poor long-term prognosis.[6] The synthesis of excess glycosaminoglycans myoepithelial cells-responsible for discrete intercellular spaces or histologic variants associated with this process-is common to a number of salivary gland tumors, including pleomorphic adenoma, myoepithelioma, basal cell adenoma, epithelial-myoepithelial carcinoma, and polymorphous low-grade adenocarcinoma.[43-46] Ultrastructural studies of some cases of sialoblastoma reveal reduplication of basal lamina,[16,47] and in one adult example in this report, frank intercellular accumulations of glycosaminoglycans and basal lamina in association with basaloid tumor cells.[48]

and basal lamina by neoplastic basal/Sialoblastoma is, therefore, another salivary gland tumor with a potential for a cribriform element.

Conclusion

Sialoblastoma is rare salivary gland tumours, almost all cases have been reported below four year of age in English literature. Recently cases of adult sialoblastoma have been reported. And it has been suggested that cribriform pattern is evident in sialoblastoma. Surgical excision with negative margin is mainstay of treatment, yet in unresectable case brachetherapy has been used successfully. Instead of rapid growth potential prognosis is good; no death has been reported due to metastasis. Existence of adult sialobalstoma needed further clarification with large case series.

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