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Case Report

Sialolipoma of salivary glands: Two case reports and review of the literature

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ABSTRACT

Sialolipoma is a rare neoplasm of salivary glands, described as a distinct entity by Nagao *et al*. in 2001. Thirty-six cases of sialolipoma in minor and major salivary glands have been reported thus far in addition to the two new cases of sialolipoma arising in the major salivary glands in this study. Thirty-six cases of sialolipoma published in English language reports were analyzed considering gender, age, location, size, duration of symptoms, treatment mode, follow-up, and histologic findings. Congenital sialolipomas were considered in this study. The first case occurred in a 45-year-old female and presented as a localized swelling in right parotid area. The second case occurred in an 18-year-old female as a swelling in the left parotid region. On histopathological examination, these lesions were diagnosed as sialolipoma.

Key Words: Histopathological feature, sialolipoma, salivary glands

INTRODUCTION

Sialolipoma is a rare tumor of salivary glands, described in 2001 by Nagao *et al*.[1] It is a benign lipomatous neoplastic growth of the salivary glands with mature adipose tissue. Sialolipoma occurs in the minor and major salivary glands with an unpredictable and potentially aggressive behavior.[2-5] Treatment of sialolipoma includes total parotidectomy for deep lobe involvement and superficial parotidectomy in tumor of outer lobes, with preservation of the facial nerve as in benign neoplasms of salivary glands.[3,6] The aim of this article is to report two new cases of sialolipoma and discuss the clinico-pathologic features, differential diagnosis, and histopathological findings of the lesions. A thorough review of the available English literature revealed only 17 cases of sialolipoma arising in the minor salivary glands and 19 cases in major salivary gland with no recurrences.

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CASE REPORTS

Case 1

A 45-year-old female presented with a painless mass in the right parotid gland. The patient had neither any systemic diseases nor any trauma on the region. The overlying skin was normal in color and appearance. A course of antibiotic therapy had been prescribed to rule out infection causes without any beneficial effect. An ultrasound imaging showed a relatively solid hypoecho nodule in the right parotid gland at the superficial lobe [Figure 1]. Fine needle aspiration (FNA) was performed. Under general anesthesia, superficial parotidectomy was done, superficial lobe with tumor was freely separated from adjacent tissue. The gross specimen contained a soft, brown-yellowish lobulated mass with definitive borders measuring 7.5 × 5 × 2.5 [Figure 2].

Microscopically, the specimen showed mature adipose tissue with some salivary gland structure. The ratio of mature adipose tissue to salivary gland tissue was 2:5 [Figure 3]. A one-year follow-up period showed no evidence of recurrence [Figure 4].

Case 2

An 18-year-old female was admitted to the Ghaem hospital, Mashhad, Iran, with a one-year history of painless swelling in the left parotid region. The mass did not grow significantly in the past year. Clinical examination showed a soft movable mass approximately 6 cm × 5 cm in size. Ultrasound imaging revealed a hypoecho nodule [Figure 5]. The gross specimen was 5 × 4 × 3 in size. The lesion was removed by superficial parotidectomy under general anesthesia. Histopathologic examination revealed a well-defined lobular structure, containing mature fatty tissue, within salivary gland element. The ratio of mature adipose tissue to salivary gland tissue was

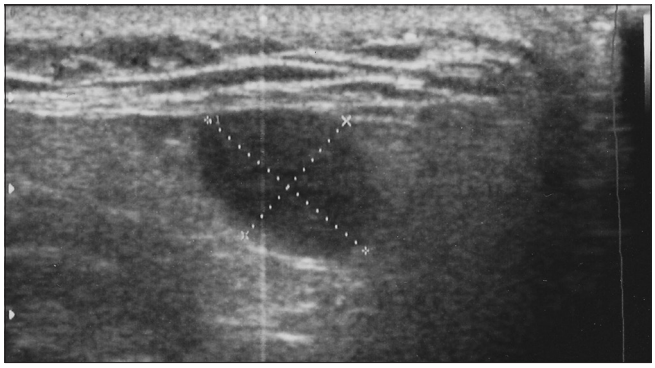

Figure 1: Ultrasonography of the lesion. A solid hypoecho nodule in right superficial parotid lobe

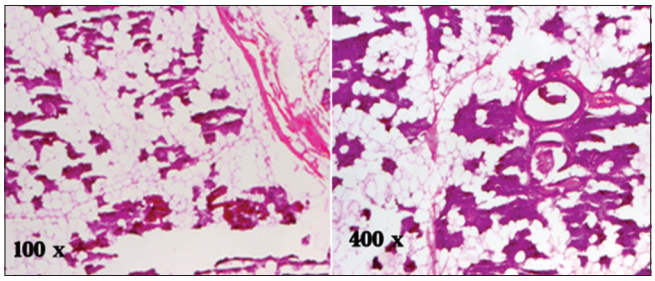

Figure 3: Microscopically, the lesion composes of mature adipose tissue with atrophic salivary glands (H and E staining)

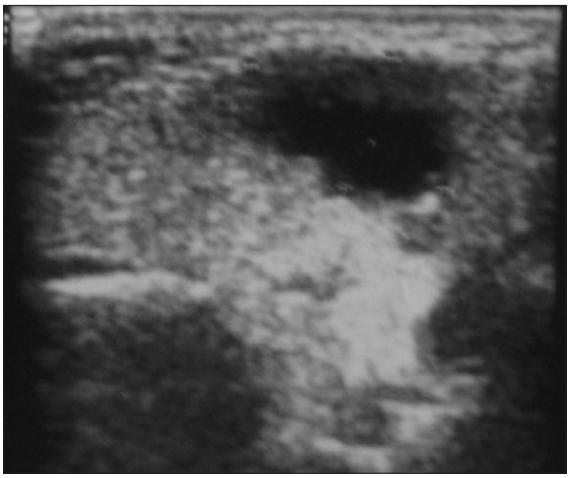

Figure 5: Ultrasonography of the left parotid region showing a hypoechoic mass

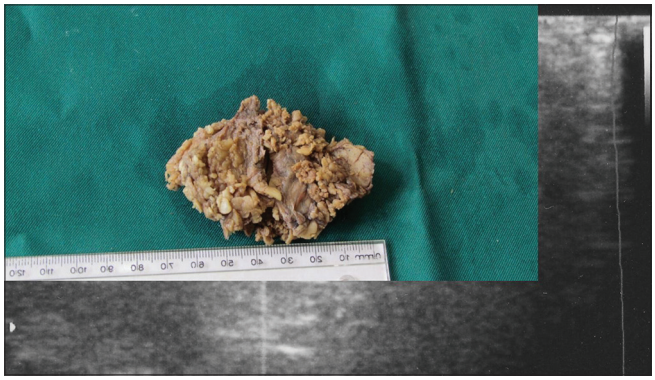

Figure 2: Gross examination showing a soft yellowish lobulated mass

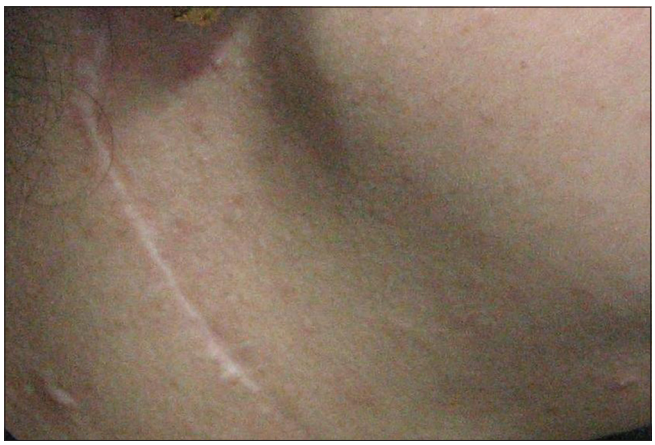

Figure 4: Clinical photograph: A one year follow-up period showing no evidence of recurrence

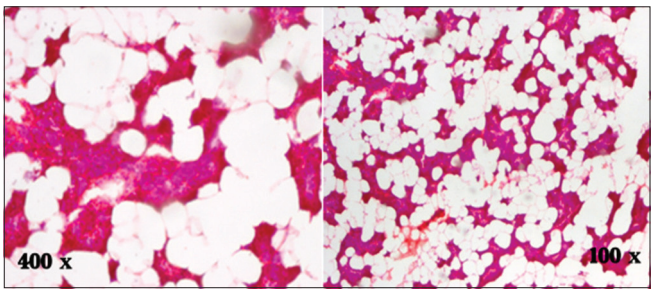

Figure 6: Histopathologic examination showing adipose tissue with salivary gland structure (H and E staining)

1:3 [Figure 6]. Eight-month follow-up after surgery showed no sign of recurrence.

DISCUSSION

Sialolipoma is a rare, recently recognized entity with only 36 cases published so far. It is a well-circumscribed lesion that has a thin fibrous capsule. Microscopically, this lesion characterizes with mature adipose tissue and non-neoplastic salivary gland tissue.[1]

Atrophy of the glandular structures, ductal dilation with scattered foci of fibrosis, sebaceous and squamous metaplasia, oncocytic changes, myxoid islands, lymphocyte infiltration, inside the lipomatous proliferation are also present.[1-3,7,8] The histopathologic differential diagnosis of sialolipoma includes lipoma and pleomorphic adenoma.[7,9,10] Lipoma usually contains mature encapsulated fatty cells, but sialolipoma has salivary gland elements between adipose tissue. Fine needle aspiration (FNA) is not an appropriate biopsy technique for diagnosis of sialolipoma, because both fatty and salivary gland tissues are not dysplastic. MRI and ultrasound are useful in the diagnosis of this lesion. Nonaka *et al.* reported the mass in tongue that had heterogen intensity in T1 and T2 sequences.[11] Sakai reported a hard plate mass with a hypointensity in T1 and isointensity in T2 images. Our cases had pre-operative ultrasound, which revealed a solid hypoecho nodule.[5]

Thirty-six cases of sialolipoma published in English language reports were analyzed considering gender, age, location, size, duration of symptoms, treatment mode, follow-up, and histologic findings

Table 1: Summary of clinicopathological features of minor salivary gland

Case	Author	Age	Sex	Location	Duration	Size	Treatment	Follow-up	Histological findings
1	Nagao *et al.*[1]	66	M	Soft palate	72 mo	2.2 cm	Surgical excision	11 mo	Duct dilatation, atrophy, fibrosis, myxoi degeneration
2	Nagao *et al.*[1]	75	M	Hard palate	3 y	1 cm	Surgical excision	NA	Duct dilatation, atrophy, fibrosis, squamous metaplasia
3	Lin *et al.*[7]	67	F	Floor of mouth	1 y	3 cm	Surgical excision	2 y	Duct dilatation, fibrosis, enlarged vessel
4	Sakai *et al.*[5]	60	F	Hard palate	10 y	1.8 cm	Surgical excision	No evidence of disease	NS
5	Fregnani *et al.*[15]	NA	NA	Tongue	NA	NA	Surgical excision	No evidence of disease	Duct dilatation, atrophy
6	Fregnani *et al.*[15]	NA	NA	Buccal sulcus	NA	NA	Surgical excision	No evidence of disease	Duct dilatation, atrophy
7	Ramer *et al.*[14]	84	F	Buccal mucosa	NA	1 cm	Surgical excision	11 mo	Duct dilatation, atrophy, enlarged vessel, lymphocyte infiltration
8	Ramer *et al.*[14]	43	F	Soft palate	NA	2 cm	Surgical excision	NA	Duct dilatation, atrophy, lymphocyte infiltration
9	Ponniah *et al.*[4]	60	M	Floor of mouth	NA	2 cm	NA	2 y	Duct dilatation, fibrosis, oncocytic metaplasia
10	de Freitas *et al.*[16]	38	M	Lower lip	NA	1 cm	Surgical excision	NA	NS
11	Okada *et al.*[6]	66	F	Hard palate	10 y	1.2 cm	Surgical excision	NA	Duct dilatation, atrophy, fibrosis, lymphocyte infiltration, enlarged vessel, oncocytic metaplasia, myxoid degeneration
12	de Moraes *et al.*[12]	72	F	Hard palate	15 days	1.7 cm	Surgical excision	NED at 8 months	Duct ectasia, Atrophy, fibrosis. hyperplasia ductal
13	Nonaka *et al.*[11]	27	F	Tongue	5 y	1 cm	Surgical excision	1.5 mo	Duct dilatation, atrophy, fibrosis, lymphocyte infiltration, squamous Metaplasia of duct cells
14	Nonaka *et al.*[11]	73	F	Floor of mouth	NA	4 cm	Surgical excision	NA	Duct dilatation, atrophy, fibrosis, lymphocyte infiltration, squamous and oncocytic Metaplasia
15	Nonaka *et al.*[11]	65	F	Buccal mucosa	2 y	2 cm	Surgical excision	NA	Duct dilatation, atrophy, oncocytic Metaplasia
16	Nonaka *et al.*[11]	68	F	Retromolar pad	NA	0.9 cm	Surgical excision	14 mo	Duct dilatation, atrophy, lymphocyte infiltration
17	Akrish et al.[13]	67	F	Palate	NA	4 cm	Surgical excision	1 y	Duct dilatation, atrophy, lymphocyte infiltration, ectasia

Lt: Left; Rt: Right; mo: Months; NED: No evidence of disease; NA: Not available; recent case; NS: Not stated

[Tables 1 and 2]. Minor salivary gland tumors were in hard plate (5 cases),[1,5,6,12,13] floor of the mouth (3 cases),[4,7,11] buccal mucosa (2 cases),[11,14] soft plate (2 cases),[1,14] buccal sulcus (1 cases),[15] tongue (2 cases),[11,15] lower lip (1 cases),[16] and retromolar pad (1 case).[11] In this group 11 patients were female. Hard palate is the first common site of occurrence [Table 1].

Nineteen cases of sialolipoma were reported in major salivary glands without our cases [Table 2]. Fourteen cases have been reported in the parotid gland and 5 cases in the sub-mandibular gland.[1-3,8,12,17-25]

There was no report of multifocal sialolipoma or bilateral occurrence. There were only two cases of deep lobe involvement, and other cases were seen in superficial lobe. No recurrence of the tumor was reported after excision of the tumor. Based on table II, most patients were in their fourth decade of life and above with a slight male predilection.

Symptoms were present for a long time, from 2 months to 11 years [Table 2]. Tumor size was larger in major salivary glands, although large lesions, up to 4 cm in the largest diameter, were reported in minor salivary glands.

Table 2: Summary of clinico-pathological features of major salivary gland

Case	Author	Age	Sex	Location	Duration	Size	Treatment	Follow-up	Histological findings
1	Nagao *et al*.[1]	20	M	Parotid (Rt)	4 mo	3.5 cm	Superficial parotidectomy	91 mo	Atrophy, sebaceous metaplasia
2	Nagao *et al*.[1]	45	F	Parotid (Lt)	10 y	6 cm	Superficial parotidectomy	85 mo	Atrophy
3	Nagao *et al*.[1]	67	M	Parotid (Rt)	2 mo	1.7 cm	Superficial parotidectomy	37 mo	Atrophy, lymphocyte infiltration
4	Nagao *et al*.[1]	66	F	Parotid (Lt)	5 mo	6 cm	Superficial parotidectomy	35 mo	Atrophy, oncocytic metaplasia, small lymph node, peripheral nerve
5	Nagao *et al*.[1]	42	M	Parotid (Lt)	10 y	6 y	Superficial parotidectomy	20 mo	Atrophy
6	Hornigold *et al*.[2]	7 week	F	Parotid (Lt)	2.5 mo	3 cm	Superficial parotidectomy	2 y	Duct ecatsia, atrophy, lymphocyte infiltration, fibrosis
7	Michaelidis *et al*.[3]	44	M	Parotid (Rt, deep lobe)	18 mo	3.5 cm	Total parotidectomy	2 y	Peripheral nerve
8	Walts and Perzik[21]	48	M	Parotid (Lt)	NA	3.5 cm	Superficial parotidectomy	No evidence of disease	NS
9	Walts and Perzik[21]	65	M	Parotid (Lt)	2 mo	2.6 cm	Superficial parotidectomy	No evidence of disease	NS
10	Baker *et al*.[25]	44	M	Parotid (Rt)	2 mo	1 cm		30 mo	NS
11	Kadivar *et al*.[17]	3	F	Parotid (Lt)	8 mo	3 cm	Surgical excision	NA	Duct dilatation, sebaceous and squamous metaplasia, fibrosis
12	Bansal *et al*.[18]	11	M	Parotid (Lt)	11 y	7 cm	Surgical excision	1 y	Duct dilatation, lymphocyte infiltration
13	Dogan *et al*.[20]	33	M	Parotid (Lt)	1 y	2 cm	Superficial parotidectomy	17 mo	Adipose tissue and salivary gland components
14	Parente *et al*.[24]	77	F	Sub-mandibular	Months	3 cm	Superficial parotidectomy	22 mo	Enlarged vessel, oncocytic metaplasia, lymphocyte, infiltration
15	Pusiol *et al*.[8]	73	M	Sub-mandibular		9 cm	Surgical excision	Months	sebaceous and oncocytic metaplasia, hyperplasia of small and large ducts
16	Jang *et al*.[19]	62	F	Sub-mandibular	3 y	5 cm	Superficial parotidectomy	NA	Atrophy, lymphocyte infiltration, oncocytic metaplasia
17	Akrish *et al*.[13]	52	M	Sub-mandibular	NA	3.5	Surgical excision	1 y	Ectasia, oncocytic metaplasia, lymphocyte infiltration
18	Sato *et al*.[22]	3	M	Sub-mandibular	2 mo	4	Surgical excision	3 y	NS
19	Kidambi *et al*.[23]	6 weeks	M	Parotid (Lt)	NA	6.5 cm	Total parotidectomy	3mo	NS
20	Present case	45	F	Parotid (Rt)	9 mo	7.5 cm	Superficial parotidectomy	1 y	Atrophy
21	Present case	18	F	Parotid (Lt)	1 year	5 cm	Superficial parotidectomy	8 mo	Atrophy, lymphocyte infiltration

Lt: Left; Rt: Right; mo: Months; NED: No evidence of disease; NA: Not available, recent case; NS: Not stated

Recommended treatment of parotid sialolipoma, is surgery with preservation of facial nerve.[3,6]

Only two cases were treated with total parotidectomy, while other cases were treated by superficial parotidectomy with preservation of facial nerve.[3,23]

CONCLUSION

Fine needle aspiration (FNA) is not an appropriate biopsy technique for diagnosis of sialolipoma, since the lesion contains mature adipose tissue and non-neoplastic salivary gland tissue.

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