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Case Report

Oncocytic lipoadenoma of the parotid gland: a report of a new case and review of the literature

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Abstract: Oncocytic lipoadenoma is a rare salivary gland tumour composed of adipose tissue and oncocytic epithelial cells in varied proportions. This tumour is still not included in the current WHO classification of salivary gland neoplasms. We herein report a further case of oncocytic lipoadenoma originating in the parotid gland of a 55-year-old woman. The tumour presented as a slowly growing asymptomatic left-sided parotid gland mass. The resected tumour measured 2.7 cm in maximum diameter and was composed of oncocytoma-like epithelial component admixed with mature adipocytes that made up 10% of the whole mass. Foci of sebaceous differentiation were seen. This rare variant of lipomatous salivary gland tumours is in need of more recognition and should be distinguished from other fat-containing salivary gland lesions, particularly lipomatous pleomorphic adenoma and myoepithelioma.

Keywords: Salivary gland tumours, oncocytes, sialolipoma, lipoadenoma

Introduction

Sialolipoma is a relatively new and uncommon type of benign salivary gland tumours that found a place in the WHO classification of salivary gland tumours only in 2005 [1]. The term sialolipoma was suggested by Nagao [2] in 2001, who reported a series of 7 salivary gland tumours, well circumcised and comprised of mature adipose elements, as well as glandular tissue, which closely resembled the cellular and structural composition of normal salivary glands. Since the tumour's original description, around 35 cases of sialolipoma have been reported in the English and German literature [2-22] (**Table 1**). However, the variant composed chiefly of oncocytic cells is quite rare and still is not included in the current WHO classification of salivary gland tumours. This variant has been most commonly referred to as oncocytic lipoadenoma. Here we present a case of oncocytic lipoadenoma of the parotid gland and attempt to review the relevant literature.

Case report

A 55-year old female patient presented to our Outpatient Department with a slowly progressing asymptomatic swelling of the left parotid region. The patient has noticed the swelling two months before and reported that it has gradually increased in size. The swelling did not fluctuate in size with eating. On examination, an approximately 20mm x 20mm round lesion was palpated in the left parotid region. The lesion was not mobile on palpation. There were no signs of inflammation. The salivary secretion from both the parotid glands was normal. A magnetic resonance tomography (MRT) was performed as the next step of the diagnostic procedure. A round, well defined, heterogeneous lesion with intermediate signal intensity in both T1 and T2 series was described (**Figure 1A**, **B**). The lesion was considered to be a parotid gland adenoma and a lateral parotidectomy was planned. The procedure was completed without any complications, fully preserving the function of the facial nerve. One year after the surgical removal of the tumour, there were no signs of recurrence.

Pathological findings

The resected specimen contained an encapsulated tumour measuring 24 x 18 x 27 mm.

Table 1. Summary of the reported cases of sialolipoma/lipoadenoma in the literature

Case No	Author	Year	Age in years	Sex	Site	Size (mm)	Fatty Tissue %	Oncocytic Metaplasia
1	Nagao	2001	20	M	Parotid	35x30x22	90	No
2	Nagao	2001	45	F	Parotid	60x30x20	90	No
3	Nagao	2001	67	M	Parotid	17x17x17	90	No
4	Nagao	2001	66	F	Parotid	max 60	90	Yes
5	Nagao	2001	42	M	Parotid	max 60	90	No
6	Nagao	2001	66	M	Soft Palate	22x15x15	50	No
7	Nagao	2001	75	M	Hard Palate	max 10	50	No
8	Fregnani	2003			Tongue			No
9	Fregnani	2003			Buccal Mucosa			No
10	Hornigold	2003	0	F	Parotid	35x26x17		No
11	Lin	2004	67	F	Floor of Mouth	30x20		No
12	Michaelides	2006	44	M	Parotid	max 35		No
13	Sakai	2006	60	F	Hard Palate	18x12x10		No
14	Kadivar	2007	3	F	Parotid	max 30		No
15	Bansal	2007	11	M	Parotid	max 70		No
16	Ramer	2007	84	F	Buccal Mucosa	14x10x10		No
17	Ramer	2007	43	F	Soft Palate	20x20x20		No
18	Ponniah	2007	60	M	Floor of Mouth	max 20		No
19	Parente	2008	77	F	Submandibular	30x20x18	80	Yes
20	Pusiol	2009	73	M	Submandibular	max 20	80	Yes
21	Okada	2009	66	F	Hard Palate	20x16x10	40	Yes
22	Dogan	2009	33	M	Parotid	26x21x17		No
23	Jang	2009	62	F	Submandibular			Yes
24	Fritzsche	2009	43	M	Parotid	65x50x20		Yes
25	Sato	2010	32	M	Submandibular	40x30		No
26	de Moraes	2010	72	F	Hard Palate	17x13x6		No
27	Nonaka	2011	27	F	Tongue	max 10	50	No
28	Nonaka	2011	73	F	Floor of Mouth	40x10	60	No
29	Nonaka	2011	65	F	Buccal Mucosa	max 20	60	Yes
30	Nonaka	2011	68	F	Retromolar	max 9	60	Yes
31	Akrish	2011	52	M	Submandibular	35x20x15		Yes
32	Akrish	2011	67	F	Hard Palate	50x40x40		No
33	Kidambi	2012	0	M	Parotid	45x30x45	50	No
34	This report	2012	55	F	Parotid	27x24x18		Yes

Macroscopically the tumour was lobulated and whitish-brownish to yellow in appearance and was surrounded by a thin connective tissue capsule. Histological examination showed a predominance of large polygonal oncocytic cells arranged in large lobules separated by thin fibrous septa that occasionally contained residual normal serous acini (**Figure 2A**). The oncocytic component was associated with a variable fatty component that formed 10% of

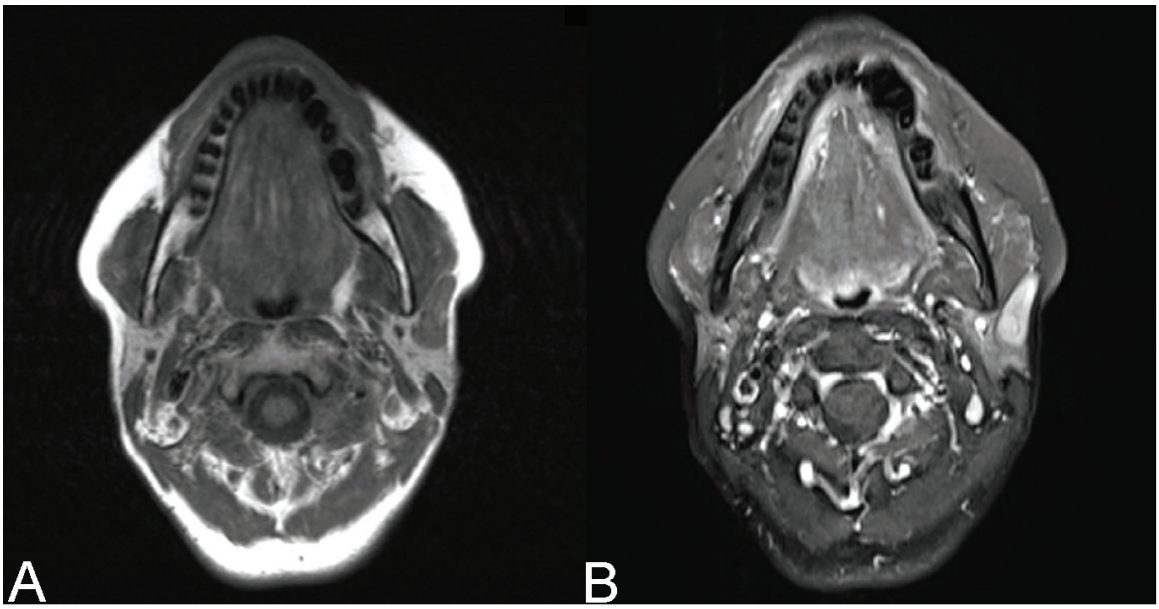

Figure 1. A, B : Magnetic Resonance Tomography imaging of the lesion: In both T1 and T2 a well defined, heterogeneous lesion with intermediate signal intensity could be depicted.

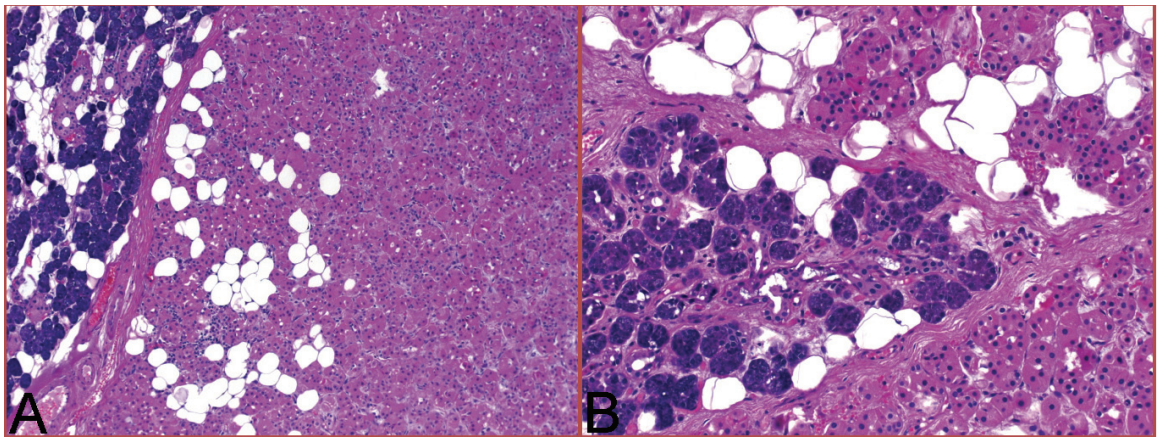

Figure 2. Histological evaluation showed an encapsulated mass surrounded by normal parotid parenchyma and composed predominantly of oncocytic epithelial cells. The fatty component varied greatly from minor (A) to almost equally distributed component (B).

the tumor mass and occasionally predominated in small areas of the tumour (**Figure 2B**). Scattered small foci of sebaceous differentiation were seen between oncocytic cells and were commonly associated with ductal dilatation, chronic inflammation and periductal fibrosis. The tumour cells were monomorphic without evidence of atypia or mitotic activity. There was no perineural invasion, lymphovascular invasion or extracapsular infiltration by the tumour.

Discussion

Lipoma in general is a relatively common tumour of subcutaneous soft tissue. However, its occurrence in the salivary glands is rare. It has been noted that only approximately 0.5% of all salivary gland tumours are lipomas [1, 23-27]. Lipoma is also quite rare in the oral cavity with a reported incidence of 1.1-4% of all benign oral cavity lesions [3, 20, 28]. Since Nagao [2] first described sialolipoma as a path-

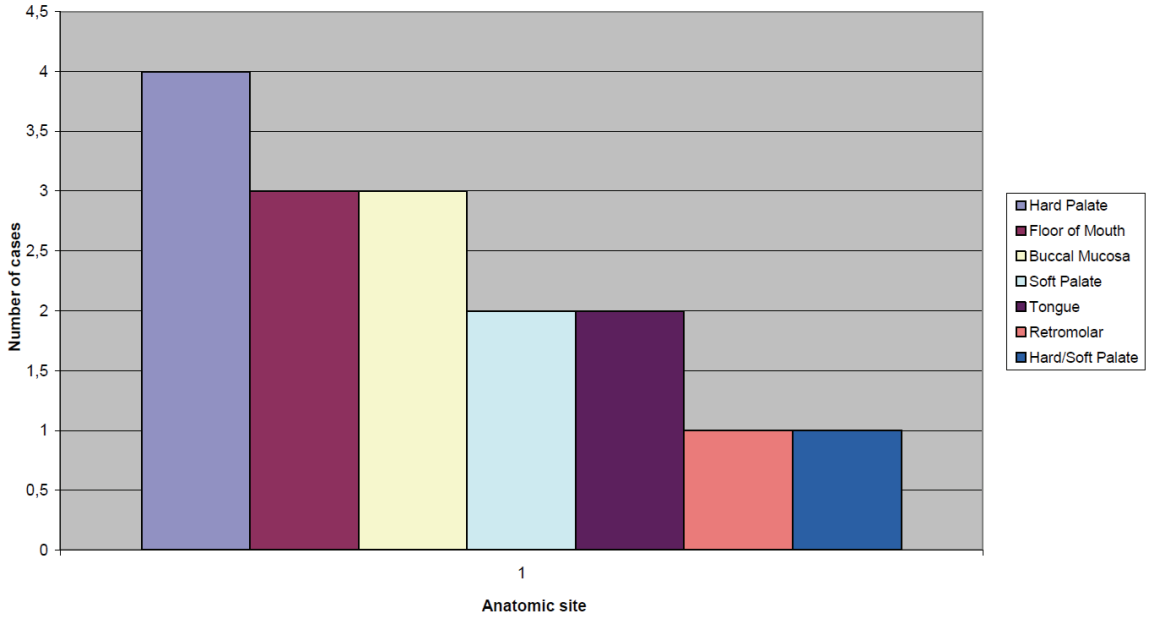

Figure 3. Distribution of minor salivary gland sialolipomas/lipoadenomas.

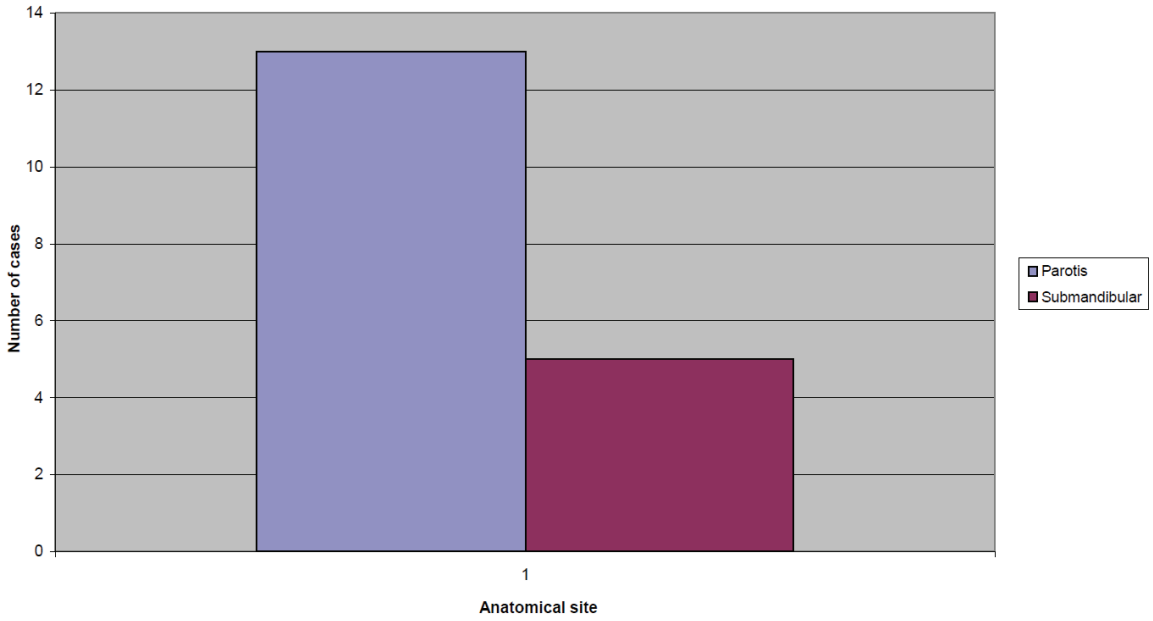

Figure 4. Distribution of major salivary gland sialolipomas/lipoadenomas.

ological entity in 2001, there have been so far 35 cases of sialolipoma (including the one we present) reported in the literature (summarized in **Table 1**). Although sialolipoma and oncocytic lipoadenoma still remain as rare tumours, increased awareness of these entities has resulted in increased number of reported cases during the last few years.

Sialolipomas present at wide age range with the youngest patient reported being 7 weeks old [4] and the oldest 84 years old [11]. Children were affected in 4 cases [4, 7, 9, 22]; all of them represented congenital tumours. The mean age of presentation is 52 years. This benign tumour shows a slight predilection for females, with a male to female ratio of approximately 1:1.4.

Both major and minor salivary glands can be affected but most lesions arise in the parotid gland. In this review we found an almost equal distribution of the tumour between major and minor salivary glands (16 cases localized in minor salivary glands, 18 in major salivary glands).

The parotid gland is the most common localisation of sialolipoma with 13 described cases (including the present case) [2, 4, 6, 7, 9, 13, 22]. The submandibular gland was involved in the other five cases affecting major salivary glands [12, 15, 17, 19, 21], whereas there has been so far no report of sublingual gland involvement. As far as minor salivary glands are concerned, the most common localisation of sialolipomas is the hard palate with 5 cases described [2, 8, 16, 18, 19], followed by the floor of the mouth [5, 10, 20] and the buccal mucosa [3, 11, 20] (3 cases each), the soft palate [2, 11], the tongue [3, 20] (2 reported cases each) and the retromolar region [20] (1 case) (**Figure 3**, **4**).

The majority of the patients presented with an asymptomatic mass or swelling in the respective site. The duration of symptoms varied greatly from 15 days to 11 years. Since many patients did not have any other complaints related to the tumour, there have been many cases where the tumour remained undiagnosed for many years, until it significantly increased in size.

Treatment has been surgical in all the reported cases. In the 13 cases where the tumour was localised in the lateral lobe of the parotid gland the treatment of choice was lateral parotidectomy in 9 of them, while local excision was performed in the other 4 cases. Therapy was local excision for all tumours at other sites. No case of recurrence has been reported so far.

The excised lesion was either spherical or oval in shape with sizes ranging from 0.9 to 7 cm (mean size: 3.2 cm).

The histopathological description of reported sialolipomas was consistent with the original description by Nagao [2]. The tumor is always encapsulated and consisted of mature adipose tissue with islets of epithelial salivary gland cells with the normal ductuloacinar structure of a salivary gland.

The amount of fatty tissue ranges in the 14 cases where this information was provided between 40 and 90% with a mean value of 68.7 %. No atypia of the epithelial element was noticed in any case. Atrophy of the glandular elements is also a common finding. Other findings that were reported were ductal ectasia, periductal fibrosis, inflammatory cell infiltrate, oncocytic ductal metaplasia and myxoid change [16]. The low staining for Ki67 reflects a low proliferative activity.

However, our case was composed of 90% oncocytes thus justifying its characterization as “oncocytic lipoadenoma”. The presence of oncocytic metaplasia is mentioned in 10 of previously reported cases [2, 12, 14-17, 19, 20]. Pusiol et al [17] characterized oncocytic metaplasia in a sialolipoma as “uncommon but not unexpected” and proposed the use of the term “oncocytic sialolipoma”. However, it still remains unclear, if the non-oncocytic sialolipoma and the oncocytic lipoadenoma represent different manifestations of the same disease process.

Regarding the pathogenesis of the lesion, Nagao [2] suggested that the glandular component had become entrapped during lipomatous proliferation and believed that it was not of neoplastic origin. This is why he suggested the term sialolipoma, to signify that the tumor is a type of lipoma and not of hamartomatous origin. Interestingly, in one case reported by Parente et al [12], a disorganized proliferation of neural and vascular structures was noticed, which led the authors to suggest that sialolipoma might be of hamartomatous origin. Akrish et al [19] disregarded Parente’s suggestion of hamartomatous nature and proposed the hypothesis that a dysfunction of a salivary gland, leading to a modification of the normal gland function underlies the formation of this tumour. To support this hypothesis Akrish et al referred to clinical characteristics such as the complete absence of recurrence capacity, the prolonged natural history of the lesion, as well as histopathological features like the noted duct ectasia, periductal fibrosis, oncocytic metaplasia, the replacement of glandular tissue with mature adipose tissue as well as the inflammatory infiltrates.

The clinical differential diagnosis should include all the benign salivary gland lesions. A CT can give hints regarding the presence of a

lipoma, since a density of 50-150 Hounsefield Units is typical for such a tumour [13]. MRT may also suggest diagnosis and allows for assessment of fatty and non-fatty components.

In the histopathological differential diagnosis of the sialolipoma and oncocytic lipoadenoma, lipomatosis, and pleomorphic adenoma with lipometaplasia should be included. Pleomorphic adenoma can be excluded by the presence of normal salivary tissue. In contrast to sialolipoma, oncocytic lipoadenoma is characterized by lack of predominant organoid salivary gland elements and paucity or absence of normal ductuloacinic structures [3, 15, 18, 29]. Microscopic differential diagnosis of sialolipoma should include also myoepithelioma and other salivary gland lesions with extensive lipometaplasia [11]. Lipomatosis is characterized by intensive shrinkage of acinar cells and it is a variant of interstitial lipomatosis [15, 30]. Sialolipoma has a distinct enlarged main mass, which helps its separation from lipomatosis. Fibrolipoma, the second most common oral lipoma contains also mature adipose tissue [29]. However, in this case there exist no identifiable salivary elements, the fat tissue being contained within dense connective tissue.

Spindle cell lipoma is another lesion that needs to be distinguished from sialolipoma. It may affect the parotid region and present typically as a well-circumscribed fatty tumour with bland spindle cells, ropey collagen, and adipocytes [29].

In conclusion, oncocytic lipoadenoma is probably not as uncommon as was initially believed and this entity should be included in the differential diagnosis of slow growing mass of the salivary glands. Although oncocytic metaplasia has been recognized in several of the reported sialolipomas, we believe that the term “oncocytic sialolipoma” is more appropriate for oncocyte-predominant lesions.

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