Sialolipoma of Minor Salivary Glands: Presentation of Five Cases and Review of the Literature with an Epidemiological Analyze

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Received: 13 August 2014 / Accepted: 22 October 2014 / Published online: 1 November 2014
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Abstract Sialolipoma is rare benign neoplasm arise from salivary glands (majors and minors) characterized by neoplastic adipose tissue with scattered non-neoplastic salivary gland acinus. To date 60 cases (including 5 cases reported in the present paper) have been reported in scientific literature. This article presents 5 new cases of sialolipoma affecting minor salivary glands (MiSG) and additionally reviews and analyzes the previously published cases to assess possible demographical differences between sialolipoma from minor and from major salivary glands. Case reports: 5 cases (3 females; 2 males; age means 63.8 years), of sialolipoma from MiSG, are reported. 2 of them were located in buccal mucosa, 1 in upper lip mucosa, 1 in floor of the mouth and 1 in retromolar area. All tumors were composed by neoplastic adipocytes cells interlaced

with normal salivary gland acinus cover it by a fibrous tissue capsule. Analyzes of literature showed that MiSG sialolipoma is most frequent in females over 60 years old, therefore and in conclusion this article assess different demographical profile of sialolipoma in respect to their topography.

Keywords Sialolipoma · Adenolipoma · Minor salivary glands · Salivary gland tumor

Introduction

Sialolipoma is an uncommon benign tumor of salivary glands characterized by normal adipocytes interlaced with normal salivary gland tissue [1]. To date and in our knowledge 58 cases (including the present cases) have been reported in scientific literature [1–34]. Recently, it has been propose some clinical and histopathological differences between sialolipomas affecting the major salivary glands (MaSG) and minor salivary glands (MiSG) [19]. With the aim of better understanding of this neoplasm we present 5 new cases of sialolipoma from MiSG and additionally we analyzed the scientific literature to establish if demographical differences exist between patients suffering sialolipoma affecting the minor and MaSG.

Case Reports

From January of 2004 to December of 2013, 5 cases of sialolipoma were referred to the Oral Medicine Clinic and Oral Pathology Service, Postgraduate Division, Dental School, National Autonomous University of México. In all cases, excisional biopsies were performed in the Maxillofacial and Oral Surgery service at same institution.

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Fig. 1 Macroscopic and macroscopic aspect of sialolipoma of minor salivary gland. **a** Macroscopic aspect. This picture of the surgical sample of case 1 show a gross specimen displaying typical lobulation and yellowish aspect due to fat predominance. **b** Whole-mount section showing the characteristic salivary gland structures distributed in an adipocytic background. *Arrows sign* fibrous tissue septa forming a well-circumscribed nodule, produce a "nodule-in-a-nodule" pattern.

Case 1; Hematoxylin–Eosine stain $\times 40$. **c** Sialolipoma showing islands of normal salivary gland acinus (*arrow*), surrounding of neoplastic adipocytes of normal aspect (*asterisk*). Case 3, Hematoxylin–Eosine stain $\times 100$. **d** Photomicroscopy of sialolipoma of minor salivary gland show neoplastic adipocytes of normal aspect (*asterisk*) and seromucous acinus (SMA) of minor salivary gland. Case 3, Hematoxylin–Eosine stain $\times 400$

From 5 cases, 3 were female (age mean 62.3 years) and 2 male (age mean 66 years); the age mean of all cases was 63.8 years. In respect to localization 3 were from buccal mucosa, 1 from floor of the mouth and 1 from retromolar area. The time of evolution before their diagnosis was from 10 to 3 years (mean 5.6 years; in two cases was not specified). Clinically in all cases sialolipoma was observed like a mobile, well-circumscribed, firm, nodule. In two cases the oral mucosa that covers the nodule showed yellowish color, the rest 3 cases were cover by oral mucosa with normal color.

Macroscopically all surgical samples had an oval shape with smooth or irregular surface and a yellowish color with bluish or brownish areas and the average of the maximum dimension was 16 mm (range 5–34 mm) (Fig. 1A). All

surgical samples were fixed in 10 % formalin for minimum 48 h, embedded in paraffin and cut at 5 μ m to be stained with Hematoxylin–Eosin technique.

Microscopically, all cases were well-circumscribed lesions, surrounded by a capsule of fibrous tissue, composed of neoplastic adipose tissue of normal aspect interlaced with glandular seromucous acinus and intercalary or secretory ducts (Fig. 1B–D). The proportion of adipose varied from 40 % (cases 2 and 4) to 60 % (cases 3 and 5). Additionally lymphoid tissue (case 1), nervous fibers (cases 2, 3 and 4), lymphocyte infiltration (cases 3 and 4), and muscular fibers (case 2) were identified. In none case it was possible to identify oncocyctic cells. A diagnosis of sialolipoma in all cases was emitted. The demographical, clinical and microscopically data of each case is show in Table 1.

Table 1 Demographical and clinical data of 5 new cases of sialolipoma of minor salivary glands

Case number	Gender	Age (years)	Evolution (years)	Clinical aspect	Localization	Size (cm)
1	Male	62	4	Nodule	Buccal mucousa	3.4
2	Female	68	10	Nodule	Buccal mucousa	1.4
3	Female	61	3	Nodule	Floor of the mouth	1.8
4	Male	70	N/S	Nodule	Buccal mucousa	0.9
5	Female	58	N/S	Nodule	Retromolar area	0.5

Discussion

Sialolipoma is an uncommon neoplasm of the salivary glands. In the best of our knowledge 60 cases (including the 5 present here) have been reported in the scientific literature [1–34]. From the 60 cases of sialolipomas; 33 cases were from MaSG (Table 2) [1–14, 21–26, 34] and 27 cases were from MiSG (Table 2) [1, 15, 20, 25, 27–30, 32, 33]. From 27 cases of sialolipoma of MiSG 8 were from palate (6 from hard palate; 2 from soft palate) [1, 16, 18, 19, 25, 28, 31]; 4 from the floor of the mouth [17, 20, 26]; 5 from the buccal mucosa [15, 16, 29], 4 from labial mucosa (3 lower; 1 upper) [27, 30, 33]; 3 from the tongue and 2 from the retromolar area [29]. It has been suggested that MiSG sialolipomas showed male predilection and an average age of 64 years [19]. However, from the 5 cases describes here 3 were from females, even more, the analyses of scientific literature showed a female predilection ($p < 0.05$) of MiSG sialolipoma with a ratio f:m of 1.8:1. Okada et al. [19] also suggested that sialolipomas from

MiSG had an average of size of 17 mm (greatest dimension). The average greatest dimension from our 5 cases was 16 mm.

The histological characteristics of sialolipoma include the presence of neoplastic mature adipocytes and non-neoplastic salivary gland tissue [1]. Recently periductal fibrosis was suggested as a particular histological image of sialolipomas from MiSG [19]. All the cases included in the present report were histological characterized by neoplastic mature adipocytes interlaced with non-neoplastic salivary gland tissue. However in none of our cases periductal fibrosis or duct dilatation was identified. Histological differential diagnosis of sialolipoma includes lipoma and lipomatosis of salivary glands and lipoadenoma [10]. The lipoma of salivary gland is a well-delimited and/or encapsulated neoplasm composed by mature adipose tissue and foci of atrophic non-neoplastic glandular acini. The glandular acini are sparsely distributed including occasionally the periphery of the tumor [9]. The lipomatosis of the salivary gland is a histological variant of the lipoma,

Table 2 Demographical data and topography of 60 sialolipoma cases reported in scientific literature (including the present 5 cases)

Demographical data/localization	Total ($n = 60$)	MaSG ($n = 33$)	MiSG ($n = 27$)	p
Age (years)	50.5 (± 22.9)	42 (± 25)	60.3 (± 15.5)	0.003*
Gender				
Female	26 (48.1 %)	11 (35.5 %)	15 (65.2 %)	0.05†
Male	28 (51.8 %)	20 (64.5 %)	8 (34.8 %)	
n/s	4		4	
Topography				
Parotid gland	26 (44.8 %)	26 (83.9 %)		
Submandibular gland	5 (8.6 %)	5 (16.1 %)		
Floor of the mouth	4 (6.8 %)		4 (14.8 %)	
Palate	8 (13.7 %)		8 (29.6 %)	
Buccal mucousa	6 (10.3 %)		6 (22.2 %)	
Tongue	3 (5.2 %)		3 (11.1 %)	
Lip (upper and lower)	4 (6.9 %)		4 (14.8 %)	
Retromolar area	2 (3.4 %)		2 (7.4 %)	

MaSG Major salivary gland, MiSG minor salivary gland, Age age mean at diagnosis, n number of cases, (%) relative frequency, ($\pm$) standard deviation, p statistical significance ($p < 0.05_{1C95\%}$), n/s no-specified

* t student test

† χ^2 test ($p < 0.05_{1C95\%}$)

Table 3 Histological differential characteristics of lipomatous lesions from salivary glands characterized by adipose tissue

	Sialolipoma	Lipomatosis	Lipoma	Adenolipoma	Lipomatous pleomorphic adenoma
Capsule/well-circumscribed	+	–	+	+	±
Normal non-neoplastic acinar cells	+	+	–	–	–
Ductus	+	–	–	–	+
Myoepithelial cells	±	–	–	–	+
Neoplastic adipocytes	+	–	+	+	–
Neoplastic glandular epithelial	–	–	–	+	+
Metaplastic epithelial	–	–	–	–	+

(+) Present, (–) absent, (±) present or absent

and is characterized by a non-well circumscribed (absence of a fibrous capsule) proliferation of mature adipocytes [1, 15]. The lipoadenoma is characterized by both neoplastic tissues: glandular acini/ductus and adipose [21]. In the present cases, we discarded lipoma due to the absence of atrophic non-neoplastic salivary gland tissue; while lipomatosis was discarded because the presence of the fibrous capsule; and lipoadenoma was discarded due to the absence of neoplastic (glandular or adipocytes) tissue [15]. Another differential diagnosis could include pleomorphic adenoma with lipometaplasia [22]. In the sialolipoma all the salivary gland tissue are non-neoplastic without any evidence of metaplasia. The Table 2 shows the histological differential parameters of sialolipoma and other lipomatous lesions from salivary glands.

Recently, it has been suggested demographical differences between patients suffering sialolipomas affecting the MaSG and MiSG [19]. To confirm or reject this suggestion we analyzed the reported cases of sialolipoma in the scientific literature, including the 5 cases of the present report. MaGS Sialolipoma is more frequent in males than females (ratio 2:1.3); and their age average at moment of the diagnosis is 42 years (± 25). MiSG Sialolipoma show female preference; 15 females; 8 males (1.8:1 f:m ratio). The result of the statistical analyse, using χ^2 test ($p < 0.05_{IC95\%}$), of the gender distribution in respect to topography was Chi square = 3.62 ($p < 0.05$) (Table 3). Four cases not specify gender. The age mean at moment of the diagnosis of patients suffering MiSG Sialolipoma is 60.3 years (standard deviation ± 15.5) while in patients suffering MaSG Sialolipoma is 42 years (standard deviation ± 25). The patients suffering sialolipoma of MiSG are older than patients suffering MaSG sialolipoma with female predominance. The difference of age from sialolipoma of MaSG and from MaSG was statistical significant ($p < 0.003$) using the “*t*” student ($p < 0.05_{IC95\%}$) test.

In conclusion, our results support the assumption that the demographical profile of patients suffering sialolipoma

from MiSG is different to patients suffering sialolipoma from MaSG [19]. The clinical practitioner must be aware to include the sialolipoma as a differential diagnosis of minor salivary gland tumors, specifically when a unique, mobile, asymptomatic nodule from minor salivary gland, especially from palate, is observed in females older than 60 years. The treatment of sialolipoma is surgical excision. The entire patient reported here has not shown signs of recurrence.

Acknowledgments This research received no specific grant from any funding agency in the public, commercial, or not for-profit sectors.

Conflict of interest The authors declare that there is no conflict of interest.

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