

原文題目(出處)：	Sialolipoma of minor salivary glands. Annals Diag Pathol 2011;15:6-11
原文作者姓名：	Cassiano Francisco Weege Nonaka, Karuza Maria Alves Pereira, Pedro Paulo de Andrade Santos, Roseana de Almeida Freitas, Márcia Cristina da Costa Miguel
通訊作者學校：	Federal University of Rio Grande do Norte, Natal, RN, Brazil
報告者姓名(組別)：	黃靜瑜 intern J 組
報告日期：	100/06/13

內文：

1. Introduction

■ Lipomas

- (1) Benign neoplasms of mature adipose tissue
- (2) Relatively uncommon in the oral cavity, corresponding to 0.1% to 5.0% of all benign tumors at this anatomical site.
- (3) Several histopathologic variants of lipoma have been identified in the oral cavity, including :
 - Fibrolipoma
 - Angiolipoma
 - Chondrolipoma
 - Chondroid lipoma
 - Pleomorphic lipoma
 - Spindle cell lipoma

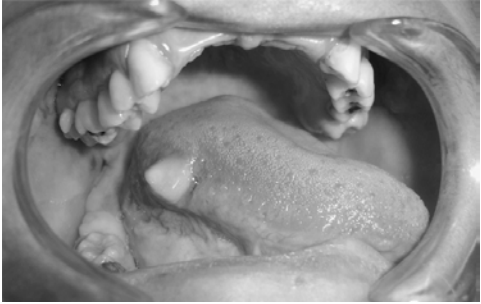
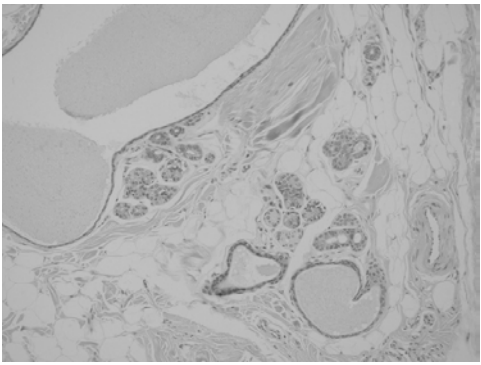
■ Sialolipomas

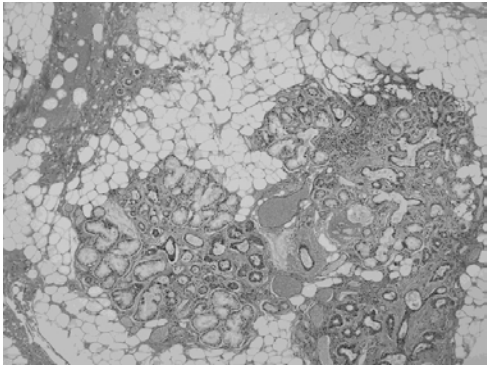
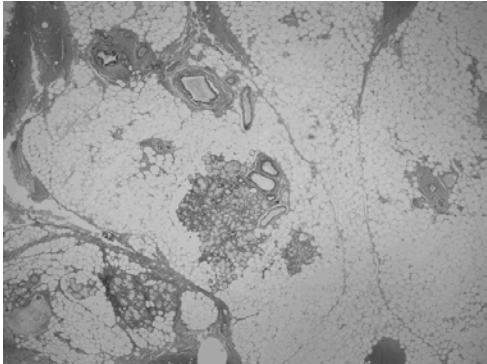
- (1) Recognized as a distinct entity by Nagao et al.
- (2) A well-demarcated proliferation of mature adipocytes with secondary entrapment of salivary gland elements.
- (3) In both the major and minor salivary glands.
- (4) More than 20 cases of sialolipoma have been reported in the English literature (Pubmed database).

■ The present article

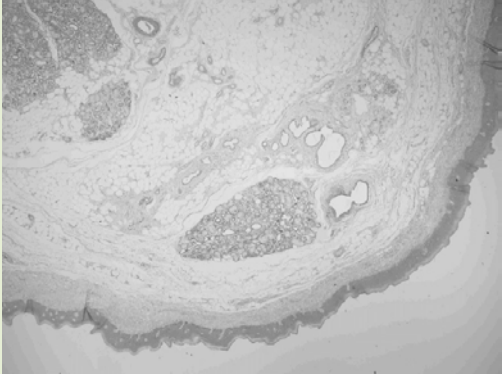
- (1) Reports 4 additional cases of sialolipoma, all of them affecting the minor salivary glands
- (2) Reviews the literature regarding clinicopathologic aspects, differential diagnosis, and therapeutic management of this recently recognized histologic variant of lipoma.

2. Case reports

	Case 1	Case 2
Age	A 27-year-old woman	A 73-year-old woman
Site	Posterior region of the lateral margin of the tongue	Left side of the floor of her mouth
Feature	<ul style="list-style-type: none"> • Painless, firm, pinkish nodule 	<ul style="list-style-type: none"> • Painless mass • Well demarcated • Covered with a nonulcerated mucosa of normal color
Size	1.0 × 1.0 cm	4.0 × 1.0 cm
Duration	5 years earlier	(not mentioned)
		(no picture)
Medical history	unremarkable	noncontributory
Clinical diagnosis	Fibroma	Ranula
	Excisional biopsy	Excisional biopsy
	<ul style="list-style-type: none"> • Proliferation of mature adipocytes • Entrapped salivary gl. parenchyma showing atrophic mucous acini & markedly dilated ducts. 	<ul style="list-style-type: none"> • Proliferation of mature adipocytes • Entrapped mucous acini and ducts of the minor salivary glands. • The glandular parenchyma exhibited fibrosis, acinar atrophy, dilated ducts with foci of squamous metaplasia and oncocytic change • A moderate lymphoplasmacytic inflammatory infiltrate
Microscopic analysis	 <ul style="list-style-type: none"> • The adipose tissue accounted for about 50% of the tumor volume • The glandular elements presented as sparsely distributed epithelial 	

	<p>islands of variable size.</p> <ul style="list-style-type: none"> • Fibrosis, squamous metaplasia of duct cells • A discrete lymphoplasmacytic inflammatory infiltrate. • A sharp demarcation from the surrounding tissue was noted at the periphery of the tumor. 	 <ul style="list-style-type: none"> • The adipose tissue accounted for most of the tumor volume (65%) • The glandular component presented as epithelial islands of variable size distributed throughout the tumor.
		 <ul style="list-style-type: none"> • A thin fibrous capsule was at the periphery of the tumor.
Definitive diagnosis	Sialolipoma	Sialolipoma
Recovery	Uneventful	Uneventful

	Case 3	Case 4
Age	A 65 y/o female	A 68 y/o woman
Site	Buccal mucosa	Retromolar pad
Feature	<ul style="list-style-type: none"> • Painless nodule • The overlying mucosa was intact and of normal color. 	<ul style="list-style-type: none"> • Symptomatic, • Well-demarcated • Firm, pinkish swelling
Size	2.0 cm in max. diameter	0.9 cm in max. diameter
Duration	2 years earlier	(not mentioned)
Medical history	(not mentioned)	(not mentioned)
Clinical diagnosis	Fibroma	Fibroma

	Excisional biopsy	Excisional biopsy
Microscopic analysis	<ul style="list-style-type: none"> • A well-circumscribed tumor • Consisting of mature adipose tissue • The presence of entrapped salivary gland parenchyma showing fibrosis, acinar atrophy, and dilated ducts with foci of oncocytic change. • The glandular component presented as sparsely distributed epithelial islands • A discrete lymphoplasmacytic inflammatory infiltrate was superimposed on the glandular parenchyma. • The adipose tissue accounted for most of the tumor volume (60%). • A sharp demarcation of the tumor from the surrounding connective tissue 	<ul style="list-style-type: none"> • Proliferation of mature adipocytes enclosing the salivary gland parenchyma. • The glandular component presented acinar atrophy and ductal dilatation • The glandular component presented as sparsely distributed epithelial islands of variable size. • A discrete lymphoplasmacytic inflammatory infiltrate. • The adipose tissue accounted for 50% of the tumor volume • A thin fibrous capsule was at the periphery of the tumor.
definitive diagnosis	Sialolipoma	Sialolipoma
Recovery	(not mentioned)	(not mentioned)

3. Discussion

■ **Sialolipoma**

- (1) A histologic variant of lipoma
- (2) Well-demarcated proliferation of mature adipocytes with secondary involvement of salivary gland parenchyma.
- (3) Share similar clinical features with **conventional lipomas**, such as :
 - A wide range in patient age

- The presence of a slow-growing and asymptomatic mass .

● **Review of the previous 30 studies**

case	Author	Age	Sex	Location	Duration	Size	Treatment	Follow-up
1	Nagao et al	20	M	Parotid	4 mo	3.5 cm	Superficial parotidectomy	91 mo
2	Nagao et al	45	F	Parotid	10 y	6 cm	Superficial parotidectomy	85 mo
3	Nagao et al	67	M	Parotid	2 mo	1.7 cm	Superficial parotidectomy	37 mo
4	Nagao et al	66	F	Parotid	5 mo	6 cm	Superficial parotidectomy	35 mo
5	Nagao et al	48	M	Parotid	10y	6 cm	Superficial parotidectomy	20 mo
6	Nagao et al	66	M	Soft palate	72 mo	2.2 cm	Surgical excision	11 mo
7	Nagao et al	75	M	Hard palate	3y	1 cm	Surgical excision	NA
8	Hornigold et al	0	F	Parotid	2.5 mo	3 cm	Superficial parotidectomy	2 y
9	Lin et al	67	F	Floor of mouth	1y	3 cm	Surgical excision	2 y
10	Michaelidis et al	44	M	Parotid	18 mo	3.5 cm	Total parotidectomy	2 y
11	Sakai et al	60	F	Hard palate	10 y	1.8 cm	Surgical excision	No evidence of disease
12	Walts and Perzik	48	M	Parotid	NA	3.5 cm	Superficial parotidectomy	No evidence of disease
13	Walts and Perzik	65	M	Parotid	2 mo	2.6 cm	Superficial parotidectomy	No evidence of disease
14	Baker et al	44	M	Parotid	2 mo	1 cm	Superficial parotidectomy	30 mo
15	Fregnani et al	NA	NA	Tongue	NA	NA	Surgical excision	No evidence of disease
16	Fregnani et al	NA	NA	Buccal sulcus	NA	AN	Surgical excision	No evidence of disease
17	Ramer et al	84	F	Buccal mucosa	NA	1 cm	Surgical excision	11 mo
18	Ramer et al	43	F	Soft palate	NA	2 cm	Surgical excision	NA
19	Parente et al [77	F	Submandibular	Months	3x2x1.8 cm	Surgical excision	22 mo
20	Ponniah et al	60	M	Floor of mouth	NA	2 cm	NA	2 y
21	Kadivar et al	3	F	Parotid	7 mo	3 cm	Surgical excision	NA
22	Bansal et al	11	M	Parotid	11 y	7x7 cm	Surgical excision	1 y
23	de Freitas et al	38	M	Lower lip	NA	1 cm	Surgical excision	NA
24	Okada et al	66	F	Hard palate	10 y	1.2x1x1 cm	Surgical excision	NA
25	Jang et al	62	F	Submandibular	3 y	5 cm	Surgical excision	17 mo
26	Dogan et al	33	M	Parotid	1 y	2x2 cm	Surgical excision	No evidence of disease
27	Present study	27	F	Tongue	5 y	1 cm	Surgical excision	1.5 mo
28	Present study	73	F	Floor of mouth	NA	4 cm	Surgical excision	NA
29	Present study	65	F	Buccal mucosa	2 y	2 cm	Surgical excision	NA
30	Present study	68	F	Retromolar pad	NA	0.9 cm	Surgical excision	14 mo

(1) *Age* : A wide range in patient age

Affect from newborns to the elderly (mean age, **50.7 years**).

(2) *Sex/ Gender* :

Although previous studies have reported a male preference, a review of the cases published so far revealed a slightly higher frequency of sialolipomas among females (53.6%).

(3) *Site* :

a) With respect to salivary gland type,

- 15 (50%) of the 30 cases involved the minor salivary glands
 - 15 (50%) affected the major salivary glands.
- b) **Minor salivary gland** sialolipomas : no preference for a specific site
- Soft palate
 - Hard palate
 - Tongue
 - Buccal mucosa
 - Floor of the mouth
 - Buccal sulcus
 - Retromolar pad
 - Lower lip
- c) **Major salivary gland** sialolipomas
- 13 (86.7%) cases were observed in the parotid gland.
 - Only 2 (13.3%) affected the submandibular gland.
- (4) *Clinically feature* :
A slowly growing, asymptomatic swelling
- (5) *Size* :
- In the **minor salivary glands**, tumor size usually ranges from 0.9 to 4.0 cm in maximum diameter (mean, 1.7 cm).
 - In the **major salivary glands** usually measure 1.0 to 7.0 cm in diameter (mean, 3.7 cm).
- (6) *Duration* :
Quite variable, ranging from 2 months to 11 years
- (7) *Initial clinical diagnosis* :
- a) In the **minor salivary glands** :
- Benign salivary gland neoplasms, such as:
 - ⊕ Pleomorphic adenoma
 - ⊗ Low-grade mucoepidermoid carcinoma.
 - ✧ Our cases :
 - 3 of our cases the clinical diagnosis suggested was fibroma.
 - 1 in the floor of the mouth was clinically diagnosed as ranula.
 - ✧ Similarly, *Lin et al* reported a case of sialolipoma involving the floor of the mouth that raised a clinical diagnostic hypothesis of ranula.
- b) In the **major salivary glands** :
- ⊕ Salivary gland neoplasm
 - ⊗ Lipoma
- (8) *Method of diagnosis* :
- a) Computed tomography(CT) & magnetic resonance imaging (MRI)
- Might be useful for the diagnosis of sialolipomas
 - Sialolipomas appear as a well-circumscribed tumor presenting a low-intensity CT signal and high MRI intensity.
 - ✧ *Sakai et al* reported a case of sialolipoma located in the **palate** :

- Hyperintensity on T1-weighted images & isointensity on T2-weighted images
- Features resembling those of **subcutaneous fat**.
- ✧ According to these authors, MRI suggested that the tumor apparently had extended into the subcutaneous fat.

(9) *Histological* :

- a) Well-circumscribed lesions
- b) Proliferation of mature adipocytes with secondary involvement of salivary gland parenchyma.
- c) **The glandular components** :
 - Consisted of regularly organized epithelial & myoepithelial elements
 - Possessed normal specific cellular phenotypes and no proliferative activity, features seen in normal salivary gland tissue.
 - ➔ Thus, the glandular components probably become entrapped during lipomatous proliferation rather than representing true neoplastic elements.
 - Presents variable degrees of acinar atrophy and ductal dilatation.
- d) Slight differences in the proportion of adipose and glandular components are observed according to the type of salivary gland affected.
 - In the **major salivary glands**, adipose tissue accounts for 75% to 90% of the neoplasm.
 - In the **minor salivary glands**, lipomatous tissue accounts for 50% to 80% of their volume.
 - ✧ Coherently, in the present cases, adipose tissue accounted for 50% to 65% of the tumor volume.
- e) The glandular component consists of **epithelial islands**
 - Sparsely distributed throughout the tumor.
 - Occasionally, found at the periphery of the tumor.
 - In some cases of sialolipoma located in the palate, the epithelial components were found to be clustered.
 - ✧ In the present cases :
 - The glandular component consisted of epithelial islands of variable size, sparsely distributed throughout the tumor.
- f) **Fibrous capsule** :
 - One of the histologic criteria for the diagnosis of sialolipoma is the identification of a fibrous capsule around the tumor.

- Almost all parts of the oral cavity contain salivary glands. Thus, any intraoral lipoma that occurs at these sites might be intermingled with adjacent salivary gland elements.
- However, to diagnosis a sialolipoma in the context of minor salivary glands :
 - ⊕ The amount of adipose tissue and glandular elements should be in equal proportions
 - ⊕ Limited peripherally by a fibrous capsule.
- g) Lymphocytic infiltration
- h) Fibrosis, myxoid change in adipose tissue
- i) Squamous and oncocytic metaplasia in ductal cells.
- j) Sialolipomas encircling **small nerve bundles**
 - Particularly sialolipomas affecting the major salivary glands.
 - According to *Michaelidis et al*, this finding may suggest
 - ⊕ A more aggressive mode of growth of these tumors than that attributed to ordinary lipomas
 - ⊕ A multicentric proliferation of adipose tissue, ultimately entrapping interstitial normal structures.
- k) Sialolipomas compared with common lipomas
 - *Fregnani et al* :
 - ⊕ Increased expression of proliferating cell nuclear antigen
 - ⊕ Faster growth.
 - Both showed no recurrence during the follow-up period.

(10) *Differential diagnosis* :

The microscopic differential diagnosis of sialolipoma should include lesions containing extensive adipose tissue, such as

- Lipomatous pleomorphic adenoma
- Lipomatosis
- Lipoadenoma

a) **Lipomatous pleomorphic adenoma** :

- Although adipose tissue can account for up to 90% of the tumor mass, the adenomatous tissue in these lesions shows typical features of pleomorphic adenoma, including ducts and sheets or strands of (dark-staining) epithelial cells.

b) **Lipomatosis** :

- A nonmalignant overgrowth of adipose tissue throughout the salivary gland parenchyma, resulting in the diffuse enlargement of the latter.
- Associated with diabetes mellitus, liver cirrhosis, chronic alcoholism, malnutrition, and hormonal disturbances.
- Lipomatosis vs. Sialolipomas :
Sialolipomas has a **fibrous capsule**.

c) **Lipoadenomas** :

- Well-circumscribed tumors with distinct adipose and glandular components, called **adenolipomas** or **lipoadenomas**, have been described in the breast, thyroid, and skin.
- These lesions consist of an admixture of mature fat cells and branching narrow epithelial tubules lined with columnar cells and supported by basal cells without myoepithelial differentiation.
- Lipoadenomas vs. Sialolipomas :
The lack of normal acinar structures in lipoadenomas

d) **Sclerosing polycystic adenosis** :

- Some cases of sclerosing polycystic adenosis may present a prominent lipomatous stroma.
- In contrast to sclerosing polycystic adenosis :
→ Sialolipomas :
 - ⊕ No proliferation of ductal or acinar cells.
 - ⊖ No cellular atypia in the glandular component.

(11) *Therapeutic management* :

- In the parotid gland → Superficial parotidectomy.
- In minor salivary gland → Simple surgical excision.

(12) *Prognosis* :

- Despite the lack of follow-up data in some cases, no recurrence or malignant transformation has been reported so far.

4. Conclusion

- (1) Sialolipoma is a rare histologic variant of lipoma, commonly observed in adults.
- (2) When involving the minor salivary glands, these tumors show no preference for any site in the oral cavity.
- (3) Although the diagnostic criteria and microscopic features of sialolipomas have been relatively well established, permitting the differential diagnosis with

other important lesions and/or conditions, many aspects regarding the histopathogenesis of these tumors are still unclear.

- (4) Thus, further studies regarding this newly recognized histologic variant of lipoma should be performed, particularly using molecular biology techniques.

題號	題目
1	The following statements of lipoma, which is true? (A) Most are in 20-years-old or younger patient (B) More common in obese people (C) Most are more than 3 cm in size (D) If the caloric intake is reduce, the size of lipoma decreases as normal body fat loss
答案(B)	出處：Brad W. Neville et al, Oral and Maxillofacial PATHOLOGY 3 rd edit, St. Louis, Missouti, USA: Saunders, Elsevier; 2009. 523-524
題號	題目
2	Where is the most common site of intra-oral lipoma ? (A) Tongue (B) Soft palate & the posterior part of the hard palate (C) Buccal mucosa & buccal vestibule (D) Floor of the mouth
答案(C)	出處：Brad W. Neville et al, Oral and Maxillofacial PATHOLOGY 3 rd edit, St. Louis, Missouti, USA: Saunders, Elsevier; 2009. 523-524