Intraoral lipomas: Review of literature and report of two clinical cases

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Abstract
Background: Lipomas are benign mesenchymal tumors composed of mature adipocytes. They are classified according to their histological pattern and their etiology remains unclear. Objectives: To present two cases and review the literature.

Material and Methods: A search was conducted in the Medline / PubMed and Scielo data bases of the last 10 years (2004-2014) with the keywords “intraoral lipoma OR oral cavity lipoma”.

Results: 46 articles with 95 cases (56 women and 39 men) were reviewed. The average age was found to be 52.28 years (52.28 ± 18.55); and most of them occurred between the 4th and 6th decade of life. Lipomas occur mostly in the buccal mucosa (n = 36, 37.9%), followed by the tongue (n = 23, 24.2%) and other locations (n = 36, 37.9%). The most common histologic pattern was simple lipomas (n = 40, 42%), followed by fibrolipomas (n = 18, 18.9%) and other types (n = 37, 39.1%). The average tumor size was 19.77 ± 16.26mm.

Conclusions: Lipomas are a relatively rare finding in the oral cavity. Surgical excision is the treatment of choice and recurrence is not expected.

Key words: Benign oral tumor, oral lipoma, lipoma, oral cavity.
Introduction
Lipoma is a benign mesenchymal tumor composed of mature adipocytes (1,2). They are common in the head and neck region, but their appearance in the oral cavity is uncommon. Only 1-5% of the lesions are located in this area (3,4); representing 2.2% of all lipomas (3). The most common areas are the buccal mucosa, lips, tongue, palate, vestibule, floor of the mouth and retromolar area (5).
Clinically they are well circumscribed, painless and slow growing tumors (6-8). Their etiology and pathogenesis are not clear, even though factors like mechanical, endocrine, inflammatory (6,8-10), hypercholesterolemia and obesity (11,12), radiation (11) as well as chromosomal abnormalities (3,9,13) have been considered. Histologically they can be classified as simple lipoma, fibrolipoma, spindle cell lipoma, intramuscular lipoma, chondrolipoma, pleomorphic lipoma, myxoid lipoma, angiolipoma and sialolipoma (3,6,14).
The most accepted treatment is surgical excision, but medical management has also been proposed (4,5). Recurrence is rare (4).
In this paper we present a review of the literature of the past 10 years and two clinical cases.

Material and Methods
A literature review of the last 10 years (2004-2014) in the Medline_Pubmed database and ScIELO (Scientific Electronic Library Online) was done using the keywords "intraoral lipoma OR oral cavity lipoma". Selection criteria included literature reviews, case series and case reports in humans; in English and in Spanish. Articles which did not include intraoral lipomas or information about sex, age and/or size and lipomas localized in the parotid gland were eliminated. The papers reviewed specifically analyze the number of cases, sex, age, location, size and histological pattern.
In this review we present two cases of lipoma.

Results
157 articles were initially found, which after the application of the selection criteria were reduced to 40 case series and clinical case reports which included a total of 95 intraoral lipomas (Fig. 1). Table 1 summarizes the most significant data obtained from each article (1-40).
With regards to sex distribution, 56 cases were found in women (58.9%), 39 in men (42.2%). The average age was found to be 52.28 years (52.28 ± 18.55); and it is noteworthy that the majority of lesions occurred in the fourth and sixth decade of life.
The most common region for the occurrence was the buccal mucosa (n=36, 37.9%), followed by the tongue (n=23, 24.2%), lip (n=10, 10.5%), palate (n=7, 7.4%), floor of the mouth (n=7, 7.4%), vestibule (n=6, 6.3%), retromolar area (n=4, 4.2%) and gingiva (n = 2, 2.1%) (Table 2).
Regarding the histological type, the most common pattern was simple lipomas (n=40, 42%), fibrolipomas (n=18, 18.9%), followed by spindle cell lipoma (n=12, 12.6 %), sialolipomas (n=9, 9.5%), chondrolipomas (n=5, 5.3%), intramuscular lipomas (n=3, 3.2%), angiolipomas (n=2, 2.1%), osteolipomas (n=2, 2.1%), myxolipomas (n=2, 2.1%), angiofibrolipomas (n=1, 1.1%) and angiomyxolipomas (n=1, 1.1%) (Table 2).
With reference to the size of lipomas reviewed, the average size was recorded to be 19.77 ± 16.26 mm and two of the lesions were multiple.

Fig. 1. Flow chart with the selection criterion for the inclusion of selected publications.
Intraoral lipomas

Table 1. Articles reviewed with their most significant data.

| Author and year of publication | N° of cases | Sex | Age | Location | Size (mm) | Predominant histological pattern |
|---------------------------------|-------------|-----|-----|----------|-----------|----------------------------------|
| Darling et al., 2005 (27)       | 1           | Male| 35  | Lower lip| 40        | Chondrolipoma                     |
| Billings et al., 2006 (17)      | 7           | 4 Female 3 Male| 31 a 88 (avg: 64)| 4 in tongue 1 in buccal mucosa 1 in floor of the mouth 1 in lip | 3 a 10 (avg: 7,6) | 7 Spindle cell lipomas |
| Coimbra et al., 2006 (19)       | 1           | Female| 29  | Floor of the mouth | 15 | Spindle cell lipomas |
| Chalazona et al., 2006 (20)     | 1           | Female| 35  | Tongue | 110 | Lipoma |
| Ramesh et al., 2007 (22)        | 2           | 2 Female| 43 a 84 (avg: 63.5)| 1 in soft palate 1 in buccal mucosa | 10 a 20 (avg: 15) | 2 sialolipoma |
| Srinivasan et al., 2007 (13)    | 1           | Female| 34  | Tongue | 35 | Lipoma |
| Bandeča et al., 2007 (15)      | 6           | 3 Female 3 Male| 28 a 78 (avg: 50.2)| 4 in buccal mucosa 1 in Lip 1 in tongue | 15 a 50 (avg:30) | 4 lipoma 1 fibrolipoma 1 intramuscular lipoma |
| Imai et al., 2008 (18)          | 1           | Male| 72  | Tongue | 12-15 (multiple) | Spindle cell lipomas |
| Ahlg et al., 2008 (28)          | 1           | Female| 22  | Buccal mucosa | 40 | Angiofibrolipoma |
| Vecchio et al., 2009 (14)       | 1           | Male| 52  | Buccal mucosa | 25 | Spinolipid cell lipomas |
| Okada et al., 2009 (23)         | 1           | Female| 66  | Hard palate | 12 | Sialolipoma |
| de Freitas et al., 2009 (6)     | 26          | 20 Female 6 Male| 29 a 91 (avg:54.6)| 9 in buccal mucosa 7 in tongue 4 in lower lip 3 in floor of the mouth 2 in retromolar area 1 in vestibule | 5 a 28 (avg:11,9) | 15 lipoma 7 fibrolipoma 2 intramuscular lipoma 1 spindlecell lipomas 1 sialolipsa |
| Nonaka et al., 2009 (25)        | 1           | Male| 30  | Tongue | 14 | Chondrolipoma |
| Kumarasamy et al., 2009 (3)     | 4           | 1 Female 3 Male| 30 a 60 (avg: 45,8)| 3 in buccal mucosa 1 in vestibule | 5 a 30 (avg:16,2) | 3 lipoma 1 fibrolipoma |
| Brkić et al., 2010 (11)         | 1           | Female| 59  | Buccal mucosa | 12 | Angiofibrolipoma |
| Manjunatha et al., 2010 (21)    | 3           | 3 Male| 55 a 75 (avg:66.7)| 3 in buccal mucosa | 10 a 30 (avg:18.3) | 3 Fibrolipoma |
| Studart-Soares et al., 2010 (5) | 10          | 6 Female 4 Male| 21 a 73 (avg: 53.4)| 5 in buccal mucosa 3 in vestibule 1 in gingiva 1 in retromolar area | 10 a 30 (avg: 19) | 4 lipoma 4 fibrolipoma 1 angiofibrolipoma 1 myxolipoma |
| de Moraes et al., 2010 (7)      | 1           | Female| 71  | Hard palate | 20 | Sialolipoma |
| SY et al., 2010 (29)            | 1           | Female| 43  | Soft palate | 30 | Lipoma |
| de Castro et al., 2010 (30)     | 1           | Female| 47  | Buccal mucosa | 15 | Osteolipoma |
| Hoşeni et al., 2010 (8)         | 2           | 2 Male| 50-63 (avg: 56.5)| 1 in palate 1 in tongue | 10-15 (avg:12,5) | 2 lipoma |
| Nonaka et al., 2011 (24)        | 4           | 3 Female 1 Male| 27 a 73 (avg: 58.3)| 1 in tongue 1 in buccal mucosa 1 in floor of the mouth 1 in retromolar area | 9 a 40 (avg:19.8) | 4 sialolipoma |
| Martínez-Mata et al., 2011 (31)| 1           | Male| 12  | Buccal mucosa | 60 | Angiomyxolipoma |
| Venkateswarlu et al., 2011 (32)| 1           | Female| 6   | Lowerlip | 30 | Chondrolipoma |
| Ono et al., 2011 (33)           | 1           | Male| 52  | Tongue | 15 | Myxolipoma |
| Adhihyyi et al., 2011 (34)      | 1           | Female| 37  | Palate | 32 | Osteolipoma |
| Santos et al., 2011 (10)        | 1           | Male| 58  | Buccal mucosa | 50 | Lipoma |
| Motagi et al., 2012 (12)        | 1           | Male| 36  | Buccal mucosa | 40 | Lipoma |
| Lee et al., 2012 (10)           | 1           | Male| 71  | Tongue | 10-20 (multiple) | Lipoma |
| Channadi et al., 2012 (26)      | 1           | Female| 34  | Lower lip | 6 | Sialolipoma |
| Khubebhandari et al., 2012 (35)| 1           | Female| 10  | Buccal mucosa | 30 | Fibrolipoma |
| Taira et al., 2012 (1)          | 1           | Female| 65  | Gingiva | 21 | Lipoma |
| RaviKiran et al., 2013 (9)      | 1           | Female| 53  | Buccal mucosa | 10 | Lipoma |
| Pattipati et al., 2013 (2)      | 1           | Male| 37  | Palate | 20 | Lipoma |
| Chandik et al., 2013 (36)       | 1           | Male| 75  | Tongue | 90 | Lipoma |
| Junior et al., 2013 (37)        | 1           | Female| 64  | Tongue | 20 | Spindle cell lipomas |
| Kumar et al., 2014 (4)          | 1           | Male| 77  | Vestibule | 20 | Lipoma |
| Raj AA et al., 2014 (38)        | 1           | Male| 72  | Floor of the mouth | 37 | Lipoma |
| Raj V et al., 2014 (39)         | 1           | Male| 35  | Tongue | 10 | Chondrolipoma |
| Kamakshi et al., 2014 (40)      | 1           | Female| 6   | Lower lip | 15 | Chondrolipoma |

-Case report nº 1

We report a case of a 61 year old man with an accidental finding of a lesion; a single painless swelling in the right lower vestibule. Clinical examination revealed a mass of 1.6x1.7cm, soft, mobile, not attached to deeper planes and covered by mucosa which appeared normal but with a slight yellowish color. No neurological defects were demonstrated despite its location near the mental foramen, and cervical lymphadenopathy couldn’t be palpated (Fig. 2). A differential diagnosis of lipoma was given, which was confirmed with fine needle aspiration cytology (FNAC). The lesion was enucleated under local anesthesia with articaine 1: 100,000 4%, followed by incision, dissection, excision and suture using Vicryl® 3/0. Macroscopically it was an encapsulated lesion, easily enucleable and yellowish in color (Fig. 3a). Histological examination at 40x magnification (H & E, Hematoxylin & Eosin) showed adipocytes and a well circumscribed tumor with a thin fibrous capsule.
Table 2. Location and histologic pattern.

| Location          | %     | Histologic pattern          | %     |
|-------------------|-------|----------------------------|-------|
| Buccal mucosa     | 37.9  | Simple lipoma              | 42    |
| Tongue            | 24.2  | Fibrolipoma                | 18.9  |
| Lip               | 10.5  | Spindle cell lipomas       | 12.6  |
| Floor of the mouth| 7.4   | Sialolipoma                | 9.5   |
| Palate            | 7.4   | Chondrolipoma              | 5.3   |
| Vestibule         | 6.3   | Intramuscular lipoma       | 3.2   |
| Retromolar area   | 4.2   | Angiolipoma                | 2.1   |
| Gingiva           | 2.1   | Osteolipoma                | 2.1   |
|                   |       | Myxolipoma                 | 2.1   |
|                   |       | Angiofibrolipoma           | 1.1   |
|                   |       | Angiomyxolipoma            | 1.1   |

The postoperative period was uneventful and no recurrence was seen after 1 year of follow-up. The definitive diagnosis was confirmed to be lipoma, similar to clinical diagnosis (Fig. 3b).

-Case report n° 2

The second case is of an 89-year-old female with a nodular lesion on the left lingual border with a diameter of 1.2 cm and unknown time of evolution. The lesion was asymptomatic, well defined, smooth with soft consistency and with anormal lingual mucosal surface.

A presumptive diagnosis of fibroma was made. An excisional biopsy and enucleation of the lesion was performed. Macroscopically an encapsulated and yellowish lesion was observed. The definitive diagnosis was lipoma and there was no recurrence.

Histological examination with H & E staining revealed mature fat cells that differed little in microscopic appearance from the surrounding normal fatty tissue. The epithelium was found to be stratified squamous parakeratinized epithelium and fibrocellular connective tissue stroma, having abundant groups of oval cells with vacuolated peripheral nuclei planes, resembling adipocytes, characteristic of lipoma.

Discussion

While intraoral lipomas are relatively uncommon (1,4,7,8,15,16), their clinical diagnosis is easy due to their yellowish color, and their usual location superficially near the mucosa (1). The differential diagnosis includes fibroma, dermoid cyst, minor salivary gland tumors, mucocele, hemangioma, lymphangioma, rhabdomyoma or neuroma (4,17) (Table 3). Depending on its location, a herniated buccal fat of pad should also be kept in mind while performing a differential diagnosis (3).

The majority of cases are solitary tumors. The literature shows that only 5% occur as multiple lesions (4,13,16,18). We found 2 cases of multiple lipomas in
Table 3. Differential diagnosis of intraoral lipoma.

| Age  | Sex | Most frequent location | Predominant clinical features | Histology |
|------|-----|------------------------|-------------------------------|-----------|
| Fibroma | >20 | M=F | Buccal mucosa | Pink and firm consistency. May appear ulcerated. < 1 cm. | Stratified squamous epithelium covering a mass of dense fibrous connective tissue |
| Lymphangioma | Congenital | M=F | Anterior 2/3 of tongue | Hypochromatic, sometimes purple mass. It can cause difficulty in swallowing and interfere in phonation. | Dilatation of the lymphatic vessels |
| Rhabdomyoma | 40-50 | F>M | Lip Tongue Buccal mucosa | Slow growing. Usually asymptomatic. Firm consistency. | Numerous thin-walled vessels, calcification, muscle bundles. |
| Neuroma | 20-50 | M=F | Tongue Palate Buccal mucosa | Semi elastic consistency, firm, mobile. Painful on palpation. | Remnants of nerve endings. Double cell distribution histological pattern (Antoni A, Antoni B) |
| Dermoid cyst | Congenital | M=F | Tongue Lip Palate | Mobile, slow growing and painless. It can cause problems in speech, swallowing and breathing. | Keratinized stratified squamous epithelium. May contain hair follicles, sweat and sebaceous glands. |
| Benign tumor of minor salivary glands | 40 | F>M | Palate | Mobile, slow growing and painless. Occurs as a single lesion. | Histological variety. Epithelial component with a large number of patterns that can join stromal mixtures of myxoid tissue |
| Mucocele | 10-20 | M=F | Lip Buccal mucosa Floor of the mouth | Sessile, fluctuating, painless, rapidly growing mass. | Cyst wall without epithelial lining, with granulation tissue |
| Hemangioma | Congenital | M=H | Lip Tongue Buccal mucosa | Reddish or bluish lesions. The compression of the lesion usually produces its blanching due to ischemia. | -Capillary Hemangioma: capillaries of endothelial origin. superficial -Cavernous Hemangioma: poorly defined dilated vascular channels, affecting deeper structures -Mixed Hemangioma |

our review (16,18). When they occur as multiple lesions they may be associated with syndromes such as neurofibromatosis, Gardner's syndrome, Dercum's disease, familial multiple lipomatosis, Proteus syndrome or Pai syndrome (4).

We have already mentioned that the etiology is unknown, but two main theories have been established. (i) The “Hypertrophy theory”, which states that obesity and inadvertent growth of adipose tissue may contribute to their formation. This theory is less convincing at explaining the lesions that occur in areas lacking pre-existing adipose tissue (4). (ii) The “Metaplasia theory” suggests that the lipomatous development occurs due to aberrant differentiation of mesenchymal cells in lipoblasts (3,4,19). Other mechanisms such as trauma, infection, chromosomal abnormalities or hormonal imbalances have also been proposed (3,6,11).

According to the literature, the most common sites of this type of tumor are the buccal mucosa and the tongue (20,21). Studart-Soares et al. (5), revised 450 intraoral lipomas between 1966 and 2009, and the most common site was found to be buccal mucosa (n=174; 38.7%), followed by vestibule (n=35; 7.8%), retromolar area (n=21; 4.7%), and other sites (n=220; 48.8%). Taira et al. (1), studied 207 cases published between 1987 and 2004, and also found the buccal mucosa to be the most prominent
site \((n=84, 40.6\%)\), followed by tongue \((n=37, 17.9\%)\), lip \((n=26, 12.6\%)\), and other areas \((n=60, 28.9\%)\). With respect to the histology, the sialolipoma variety is generally encountered in minor salivary glands \(22-24\). Our review also found the buccal mucosa to be the most common site for lipoma \((n=38, 34.9\%)\), in accordance with the literature.

With respect to the sex distribution in 450 cases studied by Studart-Soares et al. \(5\), 256 were males \((52.2\%)\) and 234 were females \((47.8\%)\). The tendency was similar in the review conducted by Taira et al. \(1\). On the other hand, in the study of 26 cases by Freitas et al. \(6\), 20 were females \((76.92\%)\) and 6 were males \((23.8\%)\). In our revision, we found a female predilection with 56 cases in females \((58.9\%)\), as compared to 39 males \((41.1\%)\). Thus, if we rely on the literature, there is practically no difference in distribution between the sexes, with a male to female ratio of \(1:1.2\) \(4\). With respect to the age of distribution, all the articles we studied seemed to coincide with our revision, with the majority of the lipoma cases occurring between the 4th and 6th decade of life \(1, 5\).

Histologically, lipomas are classified based on the matrix and the properties of tumor cells: Simple lipoma, fibrolipoma, spindle cell lipoma, intramuscular lipoma, angiolipoma, chondrolipoma, pleomorphic lipoma, myxoid lipoma and sialolipoma \(3, 17, 25, 26\). Studart-Soares et al. \(5\) investigated the histological type of 390 cases, in which the most common histologic pattern was simple lipoma \((n=229, 48.7\%)\), followed by fibrolipomas \((n=103, 26.4\%)\), myxolipoma \((n=9, 2.3\%)\), angiolipomas \((n=4, 1\%)\) and others \((n=45, 11.5\%)\). Taira et al. \(1\), analyzed the histological pattern of 113 cases in their study; and the prevalence of various histological types in descending order were simple lipomas \((n=78, 69\%)\), fibrolipomas \((n=31, 27.4\%)\) and others \((n=4, 3.5\%)\). This trend coincides with the one we found in this review; we found predominance of simple lipomas \((n=40, 42\%)\), followed by fibrolipomas \((n=18, 18.9\%)\), spindle cell lipomas \((n=12, 12.6\%)\) and finally other histological types \((n=25, 26.3\%)\).

The size of lipomas varies greatly, although most of the lesions are less than \(10\) \(mm\) \(3\), reaching up to \(11\) \(cm\) in diameter \(20\). In this review, the average size was \(19.77\) \(mm\), the largest recorded lesion being \(110\) \(mm\). The treatment of choice is surgical excision. No recurrence has been described, although it may occur in the case of infiltrating lipomas basically due to an inadequate excision combined with a non-encapsulated lesion. Malignant transformation hasn’t been described either \(4\). Medical management of lipomas has also been proposed which involves injecting steroids to cause atrophy of adipose tissue. Lesions which are less than \(2.5\) \(cm\) in diameter show a better prognosis. The injection of a mixture of \(1:1\) parts lidocaine with triamcinolone acetate is repeated once a month. The average volume used ranges from \(1\) to \(3\) \(ml\) depending on the size of the tumor. Liposuction is also used using a \(16\)-gauge needle in average \(4\) to \(10\) \(cm\) or large-sized \((>10cm)\) tumors \(4\). In this review all lipomas were treated by surgical excision and none of them showed any recurrence.

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Conflict of Interest
The authors declare that they have no conflict of interest.