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Case Report

Sublingual Lipoma – An Unusual Presentation

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Abstract: Lipomas are rare in oral cavity although they are the most common tumours of mesenchymal origin in human body. The etiology remains unclear. Different theories explain the pathogenesis of this adipose tissue tumour and also different histological variants of oral lipoma have been given in literature. Intra-Oral lipomas are likely to affect cheek, tongue, lips, gingiva and rarely the floor of the mouth. We report a case of a large sublingual lipoma, associated with difficulty in speech and mastication in a 52-year-old female. The tumour was completely excised and sent for histopathological examination, which confirmed the tumour to be a simple classical lipoma.
Keywords: Intra-oral lipoma, Sublingual lipoma.

INTRODUCTION

Lipomas are among the most frequent soft tissue neoplasm of the human body however, they are not common in the oral cavity (Dattilo, D. J. *et al.*, 1996; & Del Castillo- Pando De Vera, J. L. *et al.*, 2004). About 15 to 20% of lipoma cases involve the head and neck region, while only 1–4% affects the oral cavity, an uncommon site for the occurrence of lipoma (J. G. A. M. de Visscher. 1982; & Hatziotis, J. C. 1971).

Lipomas in the oral cavity occur more commonly in the areas of fat accumulation, especially the cheek, followed by the tongue, floor of the mouth, buccal sulcus and vestibule, lip, palate, and gingiva (Fregnani, E. R. *et al.*, 2003; & Lucas, R. B 1984). Oral cavity lipoma generally shows no gender preference while some studies have shown a female preponderance (Fregnani, E. R. *et al.*, 2003; Epivatianos, A. *et al.*, 2000; & Lawoyin, J. O. *et al.*, 2001).

Oral cavity lipomas usually present as slow-growing, painless and asymptomatic lumps. It is known that with continued growth their size may interfere with speech and mastication (Dattilo, D. J. *et al.*, 1996; & Keskin, G. *et al.*, 2002).

PRESENTATION OF CASE

A 52 year old female patient presented to department of surgery with complaints of painless swelling in sublingual region since last two years, which was gradually increasing in size associated with difficulty in mastication and speech disarticulation. There was no history of dysphagia or dyspnoea. She also complained of discomfort and feeling of heaviness in the area of the swelling. There was no other relevant medical or surgical history.

Extra oral examination was normal. Intraoral examination revealed approx 6x5 cms solitary sublingual swelling predominantly towards the right side, non-tender, firm in consistency, non-fluctuant and not adhered to lingual muscles or overlying mucosa. The covering mucosa was normal in texture without ulceration or inflammation.

Excision biopsy was planned under general anesthesia with nasotracheal intubation. With blunt dissection, the mucous membrane was undermined exposing an irregular, poorly encapsulated, and lobulated pale yellow mass (Image – 1, 2 &3). There was no involvement of the tongue musculature. The mucosal layers were closed together with absorbable sutures obliterating the dead space. Excised specimen was sent for histopathological examination.

She made an uneventful recovery from the surgery. The patient has since regained normal speech and feeding capacity with no loss of sensory or motor functions of the tongue. The patient was followed up till 02 years after surgery with no signs of recurrence.

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Image – 1

Image – 2

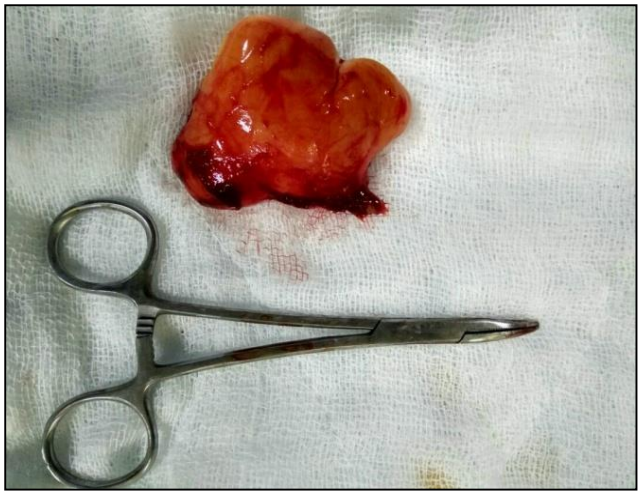

Image – 3

Histopathology of soft tissue section shows mature adipose tissue arranged in lobules separated by fibro collagenous stroma. The stroma comprises of thick irregularly placed bands of collagen with spindle shaped cells placed alongside. Lymphocyte sprinkling noted (Image – 4).

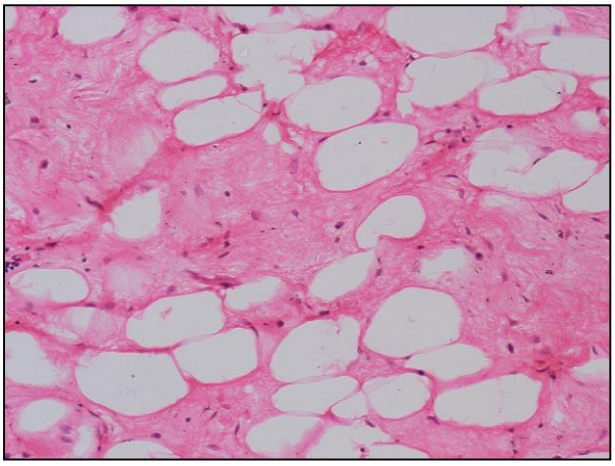

Image – 4

DISCUSSION

Lipomas are benign soft tissue neoplasm of mature adipose tissue seen as a common entity in the head and neck region, however, intraoral lipomas are rare, the statistics showing only 1 to 4% affecting these sites (de Visscher, J. G. A. M. 1982; & Hatziotis, J. C. 1971). The first ever description of an oral lesion was provided by Roux in 1848, in a review of alveolar masses which he referred to as "yellow epulis" (Rajendran, R. & Sivapathasundharam, B. 2009).

The etiology of intraoral lipoma remains unclear, but the suggested pathogenic mechanisms include the "hypertrophy theory" which states that obesity and inadvertent growth of adipose tissue may contribute to formation of these oral lesions. This theory is less convincing in explaining those lesions occurring in areas devoid of preexisting adipose (Gupta, T. K. D. 1970).

Another theory known as "metaplasia theory" suggests that lipomatous development occurs due to aberrant differentiation of in situ mesenchymal cells into lipoblast, since fatty tissue can be derived from mutable connective tissue cells almost anywhere in the body (Ashley, D. J. B. 1978).

J. J. Lin and F. Lin in their study of 459 cases of lipomas suggested that these benign entities are congenital lesions arising from embryonic multi-potential cells that remain clinically dormant until their differentiation into fat cells under hormonal influence during adolescence period (Lin, J. J., & Lin, F. 1974).

Lipomas have been reported in all parts of the body including regions of back, shoulder, neck, and extremities (de Freitas, M. A. *et al.*, 2009). Intra-oral counterparts are rare. Commonest site of intra-oral lipoma is the oral mucosa, a region rich in fatty tissue, followed by the tongue, lips, floor of the mouth, palate, and gingiva. This pattern corresponds to the quantity of fat deposits in the oral cavity (Lucas, R. B 1984; &Studart-Soares, E. C. *et al.*, 2010).

Histologically, they can be classified as simple lipoma, fibrolipoma, spindle cell lipoma, intramuscular or infiltrating lipoma, angiolipoma, pleomorphic lipoma, myxoid lipoma, and atypical lipoma. Intramuscular or infiltrating lipoma is an uncommon mesenchymal tumor, usually appearing in the extremities or trunk but rarely occurring in the oral cavity (Ayasaka, N. *et al.,* 1993).

Intra-oral lipomas may present as solitary or multiple lesions, for instance as in Gardner's or Bournville's syndrome (Del Castillo- Pando De Vera, J. L. *et al.,* 2004; Gray, A. R., & Barker, G. R. 1991) or as macroglosia or lipomatosis (Gray, A. R., & Barker, G. R. 1991; Capodiferro, S. *et al.,* 2004; Favia G. *et al.,* 2004; Nunes, F. D. *et al.,* 2002; Piattelli, A. *et al.,* 2002; & Katou, F. *et al.,* 1993).

The clinical features may vary according to the location of the lesion. Usually they manifest as slow growing, sessile round to ovoid submucosal nodules. Unless the yellow colour of the tumour appears through the overlying thin mucosa, diagnosis of these tumours clinically is not always easy (Debnath, S. C., & Saikia, A. 2010). The average duration of the lipoma before excision is 3.2 years with a range of 6 weeks to 15 years. The usual range in size is 0.5 to 8 centimeters (Furlong, M. A. *et al.,* 2004).

Signs and symptoms may include a feeling of fullness and discomfort. Rarely various functional problems like dysphagia, difficulty in speech, and mastication have also been encountered in large sublingual lipomas. Literature review has shown that 5% of the cases were multiple. Multiple lipomas have been associated with certain syndromes like neurofibromatosis, Gardner's syndrome, painful multiple subcutaneous lipomas and obesity syndrome called Dercums disease, encephalocraniocutaneous lipomatosis, multiple familial lipomatosis, Proteus syndrome and Pai syndrome (Larsen, K. *et al.,* 1984).

The differential diagnosis of intraoral lipoma includes oral dermoid and epidermoid cysts, oral lymphoepithelial cyst, benign salivary gland tumour, mucocele, benign mesenchymal neoplasm, ranula, thyroglossal duct cyst, ectopic thyroid tissue, pleomorphic adenoma and mucoepidemoid carcinoma, angiolipoma, fibrolipoma and malignant lymphoma. Lesions appearing as swelling on the dorsum of the tongue usually mimic hemangioma, lymphangioma, rhabdomyoma, neuroma, or neurofibroma (Del Castillo-Pando De Vera, J. L. *et al.,* 2004; Capodiferro, S. *et al.,* 2004; Favia G. *et al.,* 2004; Nunes, F. D. *et al.,* 2002; Piattelli, A. *et al.,* 2000).

The diagnosis of intraoral lipomas is usually clinical. The majority of intra-oral lipomas remain unulcerated. When they are ulcerated they present diagnostic dilemma. This was reported in a case report of a lingual lipoma that presented as a chronic non-healing ulcer (Del Castillo- Pando De Vera, J. L. *et al.,* 2004). Computed tomography and magnetic resonance imaging enable the diagnosis of these tumours to be made quite readily. In spite of availability of all these techniques, histopathology remains the gold standard in the diagnosis of lipoma (Epivatianos, A. *et al.,* 2000).

Histologically, the tumor is composed of adult fat cells that are subdivided into lobules by fibrous connective tissue septa. Based on microscopical features they are classified into classic lipoma, fibro lipoma, angiolipoma, spindle cell lipoma, and pleomorphic, myxoid, sialolipoma, and intramuscular lipomas. Among these variants, myxoid lipomas and angiolipomas are rarely found in the oral cavity (Fregnani, E. R. *et al.,* 2003; Dimitrakopoulos, I. *et al.,* 1990).

Main stay of treatment for intra-oral lipoma is complete surgical excision. No recurrence has been described after local excision, but infiltrative lipoma tends to recur after inadequate excision due to the fact that they are not encapsulated like simple lipomas. Recurrence rate as high as 62.5% has been recorded (Fregnani, E. R. *et al.,* 2003). Even in cases with recurrence there has been no reported incidence of malignant transformation (Ayasaka, N. *et al.,* 1993).

CONCLUSION

Intraoral lipomas are a rare entity which may be noticed only during routine dental examinations. Most of them rarely cause pain, resulting in delay to seek treatment. The patient's concerns may be regarding aesthetics or discomfort.

Simple lipomas regardless of their size are easily extirpated without recurrence. This unusually large sublingual lipoma was surgically excised uneventfully. The present case demonstrates that the size of simple sub-lingual lipoma can grow enormously if left untreated for long duration.

REFERENCES:

1. Dattilo, D. J., Ige J. T., & Nwana, E. J. C. (1996). Intraoral lipoma of the tongue and submandibular space. J Oral Maxillofac Surg, 54, 915-7
2. Del Castillo- Pando De Vera, J. L., Cebrian – Carretero, J. L., & Gomez-Garan, E. (2004). Chronic lingual ulceration caused by lipoma of the oral cavity. Med Oral, 9, 163-7
3. de Visscher, J. G. A. M. (1982). "Lipomas and fibrolipomas of the oral cavity," *Journal of Maxillofacial Surgery*, 10 (3), 177–181.
4. Hatziotis, J. C. (1971). "Lipoma of the oral cavity," *Oral Surgery, Oral Medicine, Oral Pathology*, 31(4), 511–524.
5. Fregnani, E. R., Pires, F. R., Falzoni, R., Lopes, M. A., & Vargas, P. A. (2003). Lipomas of the oral cavity: clinical findings, histological classification

and proliferative activity of 46 cases. *International journal of oral and maxillofacial surgery*, *32*(1), 49-53.
6. Lucas, R. B (1984). "Tumors of adipose tissue," in *Pathology of Tumors of the Oral Tissues*, pp. 176–179, Churchill-Livingstone, London, UK, 4th edition.
7. Epivatianos, A., Markopoulos, A. K., & Papanayonou, P. (2000). Benign tumors of adipose tissue of the oral cavity: a clinicopathologic study of 13 cases. J Oral Maxillofac Surg, 58, 1113-17.
8. Lawoyin, J. O., Akande, O. O., Kolude, B., & Agbaje, J. O. (2001). Lipoma of the oral cavity: clinicopathological review of seven cases form Ibadan. Niger J Med, 10, 189-91.
9. Keskin, G., Ustundag, E., Ercin, C. (2002). Multiple infiltrating lipomas of the tongue. J Laryngol Otol, 116, 395-7.
10. Rajendran, R. & Sivapathasundharam, B. (2009). *Shafer's Textbook of Oral Pathology*, Elsevier, India, 5th edition.
11. Gupta, T. K. D. (1970). "Tumors and tumor-like conditions of the adipose tissue," in *Current Problems in Surgery*, M. M. Ravitch, Ed., pp. 1–60, Year Book Medical, Chicago, Ill, USA.
12. Ashley, D. J. B. (1978). *Evans Histological Appearances of Turnouts*, Livingstone, Edinburgh, Scotland, 3th edition.
13. Lin, J. J., & Lin, F. (1974). "Two entities in angiolipoma. A study of 459 cases of lipoma with review of literature on infiltrating angiolipoma," *Cancer*, vol. 34, no. 3, pp. 720–727.
14. de Freitas, M. A., Freitas, V. S., de Lima, A. A. S., Bastos Pereira, J., & Nunes dos Santos, J. (2009). Intraoral lipomas: A study of 26 cases in a Brazilian population. *Quintessence International*, *40*(1). 79–85.
15. Studart-Soares, E. C., Costa, F. W., Sousa, F. B., Alves, A. P., & Osterne, R. L. (2010). Oral lipomas in a Brazilian population: a 10-year study and analysis of 450 cases reported in the literature. Medicina Oral, Patologia Oral y Cirugia Bucal, 15(5), e691-6.
16. Ayasaka, N., Chino Jr, T., Chino, T., Antoh, M., & Kawakami, T. (1993). Infiltrating lipoma of the mental region: report of a case. *British Journal of Oral and Maxillofacial Surgery*, *31*(6), 388-390.
17. Gray, A. R., & Barker, G. R. (1991). Sublingual lipoma. Report of an unusually large lesion. J Oral Maxillofac Surg, 49, 747-50.
18. Capodiferro, S., Scully, C., Maiorano, E., Lo Muzio, L., & Favia, G. (2004). Liposarcoma circumscriptum (lipoma-like) of the tongue: report of a case. Oral Dis, 10, 398-400.
19. Favia G., Maiorano E., Orsini G., Piatelli A. (2004). Myxoid liposarcoma of the oral cavity with involvement of the periodontal tissues. J Clin Periodontol, 28, 109-12.
20. Nunes, F. D., Loducca, S. V. L., de Oliveira, E. M. F., & de Araujo V. C. (2002). Well-differentiated liposarcoma of the tongue. Oral Oncol, 38, 117-19.
21. Piattelli, A., Rubies, C., Fioroni, M., & Steches, G. (2000). Spindle-cell lipoma of the cheek: a case report. Oral Oncol, 3, 495-6.
22. Katou, F., Shirai, N., Moategi, K., Satoh, R., & Satoh, S. (1993). Symmetrical lipomatosis of the tongue presenting as macroglossia. Report of two cases. J Craniomaxillofac, 21, 298-301.
23. Debnath, S. C., & Saikia, A. (2010). "Lipoma of the parotid gland extending from the superficial to the deep lobe: a rarity," *British Journal of Oral andMaxillofacial Surgery*, 48 (3), 203–204.
24. Furlong, M. A., Fanburg-Smith, J. C., & Childers, E. L. (2004). Lipoma of the oral and Maxillofacial region: Site and subclassifications of 125 cases. Oral Surg Oral Med Oral Pathol Oral Rad Endod, 98, 441-50.
25. Larsen, K., Juul, A., & Kristensen, S. (1984). Intraoral lipoma A rare cause of dysphagia. *The Journal of Laryngology & Otology*, *98*(10), 1041-1042.
26. Dimitrakopoulos, I., Zouloumis, L., & Trigonidis, G. (1990). Congenital lipoma of the tongue. Report of a case. *International journal of oral and maxillofacial surgery*, *19*(4), 208