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LIPOMA ON THE FLOOR OF THE MOUTH – A RARE CASE WITH UNUSUAL LOCALIZATION

ЛИПОМ НА ПОДОТ НА УСТАТА – РЕДОК СЛУЧАЈ СО НЕОБИЧНА ЛОКАЛИЗАЦИЈА

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Abstract

Oral lipomas as benign tumors are a rare entity in the oral cavity. However, certain locations that contain adipose tissue may be a place of origin for lipomas. This report presents a case of lipoma on the floor of the mouth in a 74-year old male patient, with evident swelling, difficulties in speech, eating, and talking. Clinical examination revealed tender oval yellowish mass located in the right sublingual space. Infection or cyst as a possible differential diagnosis were ruled out, besides the specific localization. Under local anesthesia, the mass with dimensions of 3.3×2.3×1 cm was totally extirpated from the right sublingual space and histopathological evaluation revealed a clear picture of lipoma, consisting of mature adipose cells. The follow up of six months did not reveal any recurrence or other morbidity events. It is of major importance to take into consideration the vital anatomic structures in order to avoid injuries to the salivary duct or lingual nerve during the surgical technique. A thorough differential diagnosis should be made before making the decision for treatment, especially because previously there were presented cases of malignant transformation of a buccal lipoma into liposarcoma. **Keywords:** oral lipoma, floor of the mouth, differential diagnosis. The authors declare that there is no conflict of interest in regard to this study. The study has not received any funding.

Апстракт

Оралните липоми како бенигни тумори се редок ентитет во оралната празнина. Сепак, тие може да се појават на одредени локализации кои содржат масно ткиво. Во овој приказ се презентира случај на липом на подот на устата кај 74-годишен маж, со евидентен оток, потешкотии во говорот, исхраната и зборувањето. Клиничкиот преглед покажа тврда овална жолтеникава маса локализирана во десниот сублингвален простор. Инфекција и циста како можна диференцијална дијагноза беа исфрлени, и покрај специфичната локализација. Во локална анестезија, масата со димензии 3,3×2,3×1 цм беше целосно екстирпирана од десниот сублингвален простор, а хистопатолошката анализа покажа јасна слика на липом, кој се состоеше од зрели масни клетки. Шест месеци подоцна, кај пациентот не беше забележан рецидив, ниту други знаци на морбидитет. Од големо значење е при хируршката техника да се земат предвид важните анатомски структури со цел да се избегне повреда на изводните канали на плунковните жлезди и на јазичниот нерв. Пред да се донесе одлуката за третман, потребно е детално да се проучи диференцијалната дијагноза, особено поради фактот што претходно се пријавени случаи на малигна трансформација на букален липом во липосарком. **Клучни зборови:** орален липом, под на устата, диференцијална дијагноза

Introduction

Lipomas are benign tumors with mesenchymal origin, consisting of mature adipocytes and are relatively common finding in the body. The head and neck region is affected by around 15-20% of all lipomas[1], but their occurrence in the oral cavity is rare, with a rate of 1:50,000 patients, which counts for 1-4.4% of all benign tumors in the mouth[2]. Buccal mucosa is the most affected site, because of the present fat tissue. However, lipomas may also develop on the lips, tongue, palate and rarely the floor of the mouth[3, 4]. The etiology of oral lipomas is not elucidated, but several hypotheses exist, including local chronic trauma as most speculated. Most of the studies have shown no gender preferences, although there are reports for slightly higher prevalence in males[5]. Generally, lipomas are growths resulting from developmental defects, but in the oral cavity they are most commonly associated with trauma, and they are more prevalent in adults older than 40. Clinically, lipomas are painless, well defined, slow growing submucosal or superficial lesions, encapsulated with thin fibrous

capsule and covered with intact mucosa[3, 6]. In addition, some reports show intramuscular infiltrative types[7, 8]. Literature reviews show that the mean diameter of lipomas in the oral cavity is 22 mm[9]. Although they do not pose a serious health threat, lipomas may cause aesthetic and functional disturbances, especially if located in a visible area or if they interfere with the function of mastication[10, 11].

However, the clinical diagnosis of lipoma on the floor of the mouth may be challenging, because of the unusual localization, nonspecific appearance, small incidence and other more likely differential diagnosis, such as ranulas.

This paper reports a case of lipoma on the floor of the mouth, preliminary giving a clinical appearance of ranula.

Case presentation

A 74-year old man was referred to the University Clinic for Maxillofacial Surgery in Skopje, complaining on painless growing mass on the sublingual area on the right side intraorally, which persisted for several weeks. Although it did not cause any pain, the mass was a reason for discomfort and interfered with the function of eating, speech, and swallowing. The anamnesis revealed a history of high blood pressure, controlled with appropriate antihypertensive medication, therefore not contributing to any specific issues.

The clinical intraoral examination revealed a relatively firm mass in the right sublingual area, ovoid in shape, extending from the central incisor on the second molar (Figure 1). The overlying mucosa did not show any signs of inflammation, neither a change in color or consistency. The mass was not fixed to the mucosa and it was not fluctuant. No ulcers or exophytic lesions were noted. The mass dimensions were roughly 4x2 cm, which caused discomfort during tongue movements and hence, it interfered with the function of speech and eating. There was no history of pus discharge, bleeding, pain, or huge variations in the size associated with taking food or drinks. Upon manual extraoral pressure in the submandibular area, the swelling bulged and was more evident in the mouth. However, palpation revealed that the mass was limited to the area above the mylohyoid muscle, indicating that it did not spread from the sublingual space. No significant cervical lymphadenopathy was noted. Blood analysis showed normal range of parameters important for ruling out infection or other disturbances that may be associated with the mass.

Regarding the dental status, the patient was partially edentulous, having fixed metal-composite bridge constructions on the both sites of the jaws, which lasted for more than 10 years. The remaining teeth were characterized with advanced attrition. There were no detected changes on the oral mucosa in terms of ulcers, change in color or other possible growths.

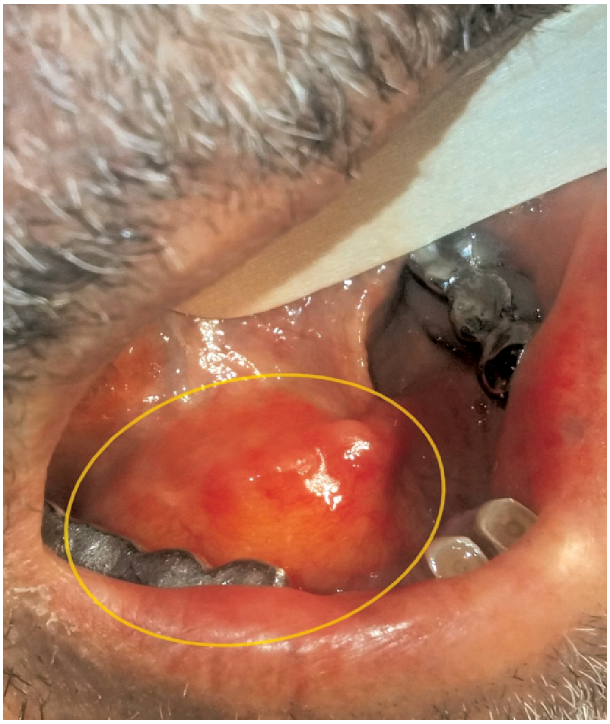

Figure 1. *Intraoral appearance of a mass in the right sublingual space. Note the metal-composite bridge constructions on both sides of the lower jaw.*

Taking into consideration the nature of the sublingual mass, its tendency not to regress, not causing any specific symptoms, except the discomfort and difficulties to speak, eat, and swallow due to the limited movement of the tongue, a decision for total extirpation of the mass was made. The procedure was performed under local anesthesia. 2 ml of solution of 2% Lidocaine hydrochloride (40 mg) and adrenaline (0,025 mg) were injected in the area of innervations of the lingual nerve, in the submucosal zone around 1 cm below the second lower molar. Additionally, a small amount of anesthetic solution was injected around the mass that was subject to extirpation. Incision was made over the highest point of the growth, taking into consideration the anatomic path of the Wharton's duct of submandibular gland and maintaining at least 1 cm distance from it. After the initial incision, a dissection of the remaining overlying mucosa was done laterally, to the extent where the mass was located (Figure 2). Then, a blunt dissection of the adjacent mucosa, connective tissue and in part of the muscle fibers was performed, taking care not to cause any injury

on the lining of the mass. The mass itself was yellowish, with smooth surface, covered with a very thin capsule. Having it carefully clumped on the periphery on its anterior aspect, with further combination of blunt dissection techniques, the mass was completely extirpated (Figures 3 and 4). The remaining surgical site was carefully examined for possible remains of the mass, and satisfactory hemostasis was achieved (Figure 5). The wound was sutured by primary intention, with non-absorbable 3-0 silk sutures. Finally, salivation was checked by manual stimulation of the submandibular gland, to make sure the Wharton's duct was intact. Once the anesthetic period of the injected solution was over, the patient was asked for any changes in sensitivity of the tongue and mucosa in the sublingual area. He confirmed no changes, approving that there was no damage to the lingual nerve.

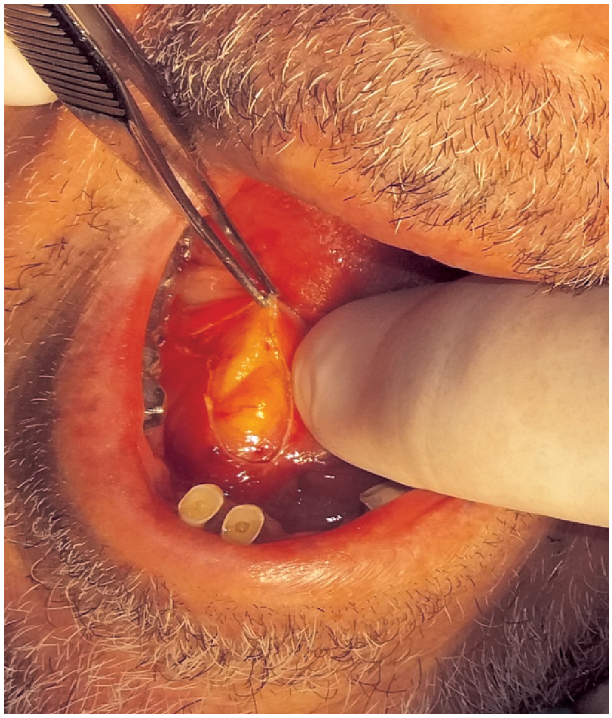

Figure 2. *After the initial incision was made, a yellowish lobulated mass just beneath the mucosa was noted.*

A specimen with dimensions 3.3×2.3×1 cm (Figure 6) was sent for histopathological evaluation and was prepared with Hematoxylin and Eosin staining. Microscopic analysis revealed mature adipose tissue lobules, comprised of benign adipocytes, separated by the connective tissue springs and incorporated with capillaries. On the surface, a thin capsule was noted. Hence, a histopathological diagnosis of lipoma was set.

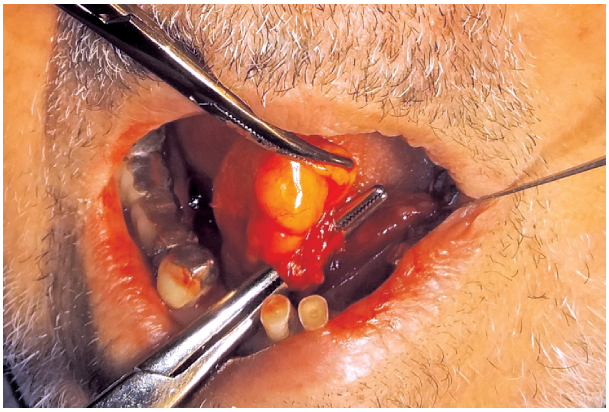

Figure 3. *Blunt dissection was performed to detach the tumor from the surrounding mucosa, connective tissue and in part the muscle fibers, in order to avoid damage of the Wharton's duct and the lingual nerve.*

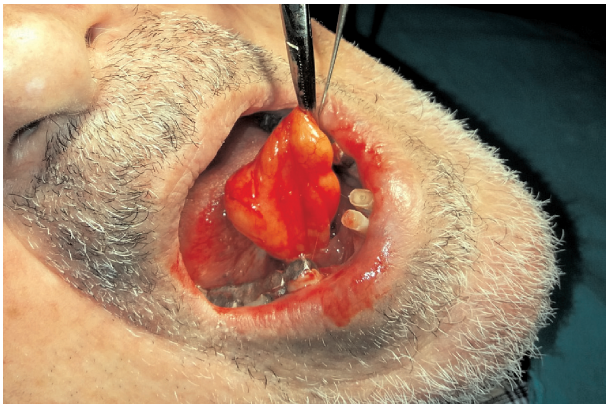

Figure 4. *Final step of tumor extirpation.*

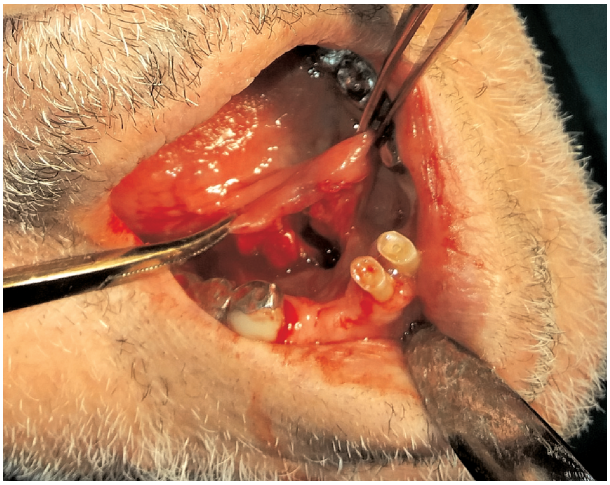

Figure 5. *Surgical site after extirpation of the tumor and after achieving satisfactory hemostasis.*

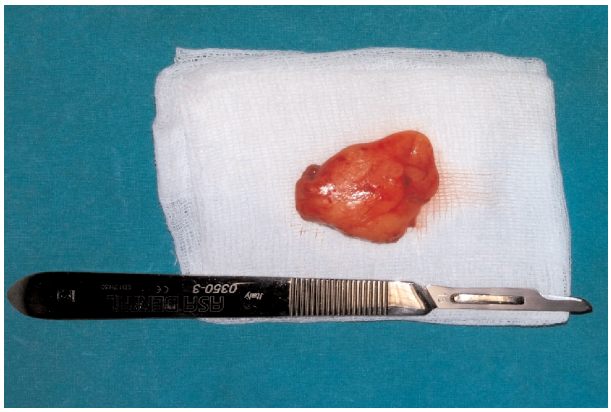

Figure 6. *The mass sent for histopathological evaluation had the following dimensions: 3.3×2.3×1 cm (tumor placed next to a surgical instrument for comparison).*

Sutures were removed one week after the surgery. The follow-up of six months did not reveal any signs of morbidity associated with the surgical procedure, nor any signs of recurrence.

Discussion and conclusion

Although oral lipomas are a rare entity, they may occur at any site where adipose tissue is present. The most common oral location is the inner side of the cheek. However, this report presents a case of a lipoma on the floor of the mouth, extending through the right sublingual space. They may occur in the lips, tongue, gingival, and palate. Lipomas are benign tumors which may remain undiagnosed until they reach certain dimensions, like in our case. Because the sublingual space may be more commonly associated with a number of pathological changes, some of which are very similar to each other, a thorough history and detailed checkup should be performed before deciding on the treatment. In terms of differential diagnosis, when such a mass is present in this area, the general dentist or surgeon should think of several possible conditions. Retention or extravasation cyst, known as ranula or mucocele, which in this zone presents as a swelling limited to the above-mylohyoid muscle space, derived from an obstruction in the ducts of the sublingual gland or the gland itself, should be ruled out. Other types of cysts may also be taken into consideration, especially developmental dermoid or epidermoid cysts. This diagnosis was ruled out because the mass in our case was not connected with the overlying mucosa, but was slightly mobile. Infection spread in the sublingual space, including abscesses may also be a possibility, but in our case, there were no signs of infection – there was no redness, pain, increased temperature, nor pus discharge or signs of damage of the mucosa. Finally, a possible tumor lesions, including benign or malignant ones, shall be taken into consideration.

A previous review shows that the mean age of patients with oral lipomas is 50.2 years[5], which is consistent with our case where the patient was over the age of 70. However, there are several reports when younger patients were diagnosed with this kind of benign tumors[5, 12]. Literature also shows that there are no predilections regarding the gender, despite the fact that lipomas are slightly more common in men. The exact reason and mechanism of occurrence of intraoral lipomas are not fully elucidated, but several hypotheses include chronic irritation or trauma, poor dental and oral status, hereditary and developmental disturbances[7].

Clinical characteristics may play an important role in deciding whether the mass will be removed or monitored. Because there were no signs of regression, the mass in our case was completely removed. The final diagnosis was set by the histopathological evaluation, which usually reveals adipocytes larger than normal, organized in lobules and encapsulated, with low vascularization. As in the case in this report, atypical changes are not seen by rule. However, a case of transformation of a lipoma on the buccal mucosa which was initially diagnosed with biopsy, into liposarcoma[13]. This possibility underlines the necessity of careful examination or deciding to remove any lesion that does not show signs of regression. In exceptional cases, imaging may be used for proper evaluation of the nature and extent of the mass. MRI may play a particularly important role since lipomas are well detected due to the high signal intensity. Sialolipomas appear on both T1- and T2-weighted images on MRI, hence being another indication for this imaging tool[14, 15]. However, different diagnostic imaging modalities such as MRI, CT, or ultrasound may not be completely accurate in distinguishing lipomas from well-differentiated liposarcomas[16]. Hence, the microscopic analysis is the gold standard for setting a final diagnosis. Histopathologically, lipomas are differentiated from liposarcoma if there are no signs of lipoblastic proliferation, adipocytes with various sizes, increased nucleous to cytoplasm ratio, signs of atypia or hyperchromatia, or bizarre stromal cells in fibrous septa, between adipocytes or in vessel walls[17, 18].

Interestingly, lipomas differ metabolically from normal fat cells even though they are histologically similar. It has been shown that the fat of lipoma is not used for energy production during starvation periods, as it happens with the normal adipose tissue[19].

Not only preliminary diagnosis may be challenging, but also certain considerations and precautions should be

taken into account when planning and performing the surgical procedure on the floor of the mouth. Important anatomical structures should be kept in mind and careful techniques should be implemented. These include the duct of the submandibular and sublingual gland and the lingual nerve, which may pass this area very superficially, posing a risk of damage. Therefore, the incision should be planned at least 1 cm away from the projected path of the duct, while sharp dissection should be avoided in order to prevent nerve damage, especially in cases where possible muscle extension of the mass is present.

Reference

1. Trandafir D, Gogalniceanu D, Trandafir V, Caruntu ID. Lipomas of the oral cavity – a retrospective study. Rev Med Chir Soc Med Nat Iasi. 2007; 111:754–758.
2. De Visscher JG. Lipomas and fibrolipomas of the oral cavity. J Oral Maxillofac Surg 1982; 10(3):177–181.
3. Fregnani E.R., Pires F.R., Falzoni R., Lopes M.A., Vargas P.A. Lipomas of the oral cavity: clinical findings, histological classification and proliferative activity of 46 cases. Int J Oral and Maxillofac Surg, 2003; 32(1): 49–53.
4. Furlong M.A., Fanburg-Smith J.C., Childers E.L.B. Lipoma of the oral and maxillofacial region: site and subclassification of 125 cases. Oral Surg, Oral Med, Oral Path, Oral Rad Endod, 2004; 98(4): 441–450.
5. Bandéca MC, de Pádua JM, Nadalin MR, Ozório JE, Silva-Sousa YT, da Cruz Perez DE. Oral soft tissue lipomas: a case series. J Can Dent Assoc. 2007; 73(5):431-434.
6. Weiss SW, Goldblum JR, editors. Benign lipomatous tumors. In: Enzinger and Weiss's soft tissue tumors. 4th ed. St. Louis: Mosby; 2001. pp. 571–639
7. Tan MS, Singh B. Difficulties in diagnosing lesions in the floor of the mouth – report of two rare cases. Ann Acad Med Singapore. 2004; 33 suppl:72-76.
8. Garavaglia J., Gnepp D.R. Intramuscular (infiltrating) lipoma of the tongue. Oral Surg Oral Med Oral Path, 1987; 63(3):348-350.
9. Manor E., Sion-Vardy N., Joshua B.Z., Bodner L. Oral lipoma: analysis of 58 new cases and review of the literature. Ann Diagn Pathol, 2011; 15(4):257–261.
10. Annibali S, Cristalli MP, Monaca GL, Giannone N, Testa NF, Russo LL, et al. Lipoma in the Soft Tissues of the Floor of the Mouth: A Case Report. The Open Otorhinolaryng J, 2009; 3:11-13.
11. Hosein AT, Razavi SM, Khabazian A. Lipoma in Oral Mucosa: Two Case Reports. Dent Res J. 2010; 7:41-3.
12. Punjabi VH, Patel S, Pathak J, Swain N. Fibrolipoma of Lip in a Young Individual: A Rare Presentation. J Cont Dent, 2017; 7(3):181-184.
13. Yamada K., Dohara Y., Nagata M., Kawashima K., Yamashita S. A case of liposarcoma of the cheek. Jpn J Clin Onc, 1979; 9(1):123–129
14. Bancroft L.W., Kransdorf M.J., Peterson J.J. O'Connor M.I. Benign fatty tumors: classification, clinical course, imaging appearance, and treatment. Skeletal Radiol, 2006; 35(10):719–733.
15. Taira Y., Yasukawa K., Yamamori I., Iino M. Oral lipoma extending superiorly from mandibular gingivobuccal fold to gingiva: a case report and analysis of 207 patients with oral lipoma in Japan. Odontol, 2012; 100(1):104–108.
16. Chikui T., Yonetsu K., Yoshiura K., Miwa K., Kanda S., Ozeki S., Shinohara M. Imaging findings of lipomas in the orofacial region with CT, US, and MRI. Oral Surg Oral Med Oral Path Oral Radiol Endodon, 1997; 84(1): 88–95.
17. Orita Y., Nishizaki K., Ogawara T., Yamadori I., Yorizane S., Akagi H, Masuda Y. Liposarcoma of the tongue: case report and literature update. Ann Oto Rhyno Laryn, 2000; 109(7):683-686.
18. Nascimento A.F., McMenamin M.E., Fletcher C.D. Liposarcomas/atypical lipomatous tumors of the oral cavity: a clinicopathologic study of 23 cases. Ann Diagn Pathol, 2002; 6(2):83-93.
19. Kumaraswamy SV, Madan N, Keerthi R, Singh Shakti. Lipomas of oral cavity: case reports with review of literature. J Maxillofac Oral Surg, 2008; 4:394–397.