

Up-to Date Review And Case Report

Intraoral fibrolipoma: case report and review of literature

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Abstract – Introduction: Lipomas are benign soft tissue tumors, characterized by the presence of mature adipocytes in histopathology. Their development is slow and their etiologies are still controversial. Frequently subcutaneous, this type of lesion is rarely present in the oral cavity. **Observation:** A 67-year-old man consulted for a nodule on the inner face of his right cheek which had been developing for several years, with severe discomfort during chewing. The typical clinical aspect of this lesion suggested a lipoma. Magnetic resonance imaging (MRI) was performed before surgical excision in order to establish a differential diagnosis with a well-differentiated liposarcoma. **Comments:** The various imaging examinations, particularly MRI, combined with the analysis of the fat component and the morphology of the non-fat component (septa) enable us to define several categories of lesions likely to have a benign or malignant character. However, some lesions remain difficult to classify, and it is the histopathological examination along with, if necessary, immunohistochemistry (anti-MDM2 and/or anti-CDK4), which allows to confirm or not the malignancy of a lesion. Histological examination suggested a fibrolipoma. **Conclusion:** Although benign, the size of a fibrolipoma may lead to significant functional and aesthetic discomfort for the patient. Surgical excision remains the treatment of choice.

Introduction

Lipoma corresponds to a benign mesenchymal tumor composed of fat tissue, with a slow development and often fortuitous finding [1]. This lesion represents 0.1–0.5% of the overall benign tumors of the oral cavity [2–4] with an identical distribution between the two sexes. This type of lesion was first described by Roux in 1848 [5].

Its pathogenesis remains quite unknown, and its development is independent from lipid metabolism and diet. Although this lesion is the most common benign soft tissue tumor [6], intraoral locations are rare [7]. The most affected anatomical sites are mucous membranes, lips, tongue, palate, floor of the mouth and gums [5,8–11].

This case report is a large intra-jugal lipoma which was treated by surgical excision. The aim of this paper was to discuss the interest of various medical imagery systems and to recall the clinical, histopathological, radiological and therapeutic features of this type of lesion.

Observation

A 67-year-old patient was referred by his dentist with a large tumefaction on the inner face of his right cheek. His medical history was marked by three myocardial infarctions, hepatic steatosis complicated by oesophageal varices, idiopathic thrombocytopenia, arterial hypertension and unbalanced type 2 diabetes. The medical follow-up revealed a non-compliance of the patient. Concerning the lesion, an intra-jugal right tumefaction had been present for several years and had become quite uncomfortable during.

The extraoral examination revealed no specific abnormality. The intraoral examination revealed a single nodule measuring four centimeters long, mobile, soft and painless on palpation, yellowish in color, highly vascularized, homogeneous, circumscribed and asymptomatic (Fig. 1).

The panoramic radiograph, except for the presence of carious lesions revealed no specific abnormality. Owing to the localization and the size of the lesion, magnetic resonance imaging (MRI) was recommended. Septa were clearly identified with this imaging system, hypo-intense on T1 and T2 weighted sequences (*versus* fat component with high signal intensity), and they enhanced barely after gadolinium injection (Fig. 2).

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Fig. 1. Intraoral view showing the lesion located on the inner face of the right cheek and interfering with the dental arch.

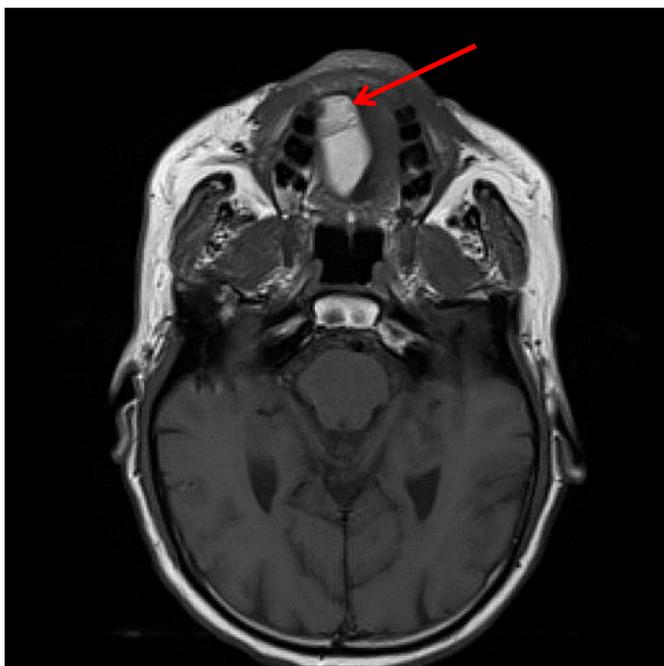


Fig. 2. Facial MRI in axial section, T1 sequence of the lesion.

A complete surgical resection was performed under a potentiated local anesthetic with anesthetic monitoring. Sutures were performed at the basis of the lesion to limit the bleeding risk. The histological analysis of the surgical specimen concluded to a fibrolipoma without cellular atypia and no sign of malignancy. Surgical margins were negative. The absence of immunohistochemical staining using the anti-MDM2 antibody confirmed the non-malignancy of this lesion. The postoperative control at fifteen days showed a good wound healing of the oral mucosa.



Fig. 3. Macroscopic view of the lesion.

Comments

A Medline (Pubmed) search was performed to investigate the current state of knowledge on intraoral lipomas. The keywords were: "lipoma", "oral", "fibrolipoma", "MRI", and "immunohistochemistry". Only the most relevant articles related to this case report were included in our review of the literature.

Lipoma is a benign mesenchymal tumor, composed of mature lobulated adipocytes and limited by fibrous septa. In a macroscopic view, lipoma looks like of a well-defined and homogeneous yellow fat mass, easy to identify due to its characteristic features, except when the lesion is located more deeply [2]. This tumor is rare in the oral cavity (1–4%) [3,4,9,10] unlike other locations, including the cervicofacial region (15–20%) [4]. The second most commonly described variant in the oral sphere is fibrolipoma, [4] (Fig. 3).

On an epidemiological aspect, the average age of onset for lipomas is 54.6 years with an equal distribution between the two genders [6,10]. This type of lesion is mainly diagnosed between 40 and 60 years old. Children are rarely affected [9]. Often unique, this lesion may be part of the clinical picture described in general pathology, such as the Gardner's and Bourneville's syndromes, lipomatosis and neurofibromatosis [6,8].

Lipomas are often correlated with chronic trauma [3,5,7,12], family history, and recently described in relation to diabetes [2]. In this case report, the patient only experienced discomfort during chewing. Most often, this type of lesion remains asymptomatic [1,3]. However, some authors associate intraoral lipomas with other symptoms, such as dysphagia and dyspnea, depending on the size and anatomical location of the lesion [6,10]. The main differential diagnosis is atypical lipomatous tumor/well-differentiated liposarcoma (ALT/WDLs). This malignant form frequently involves an intraosseous component [1].

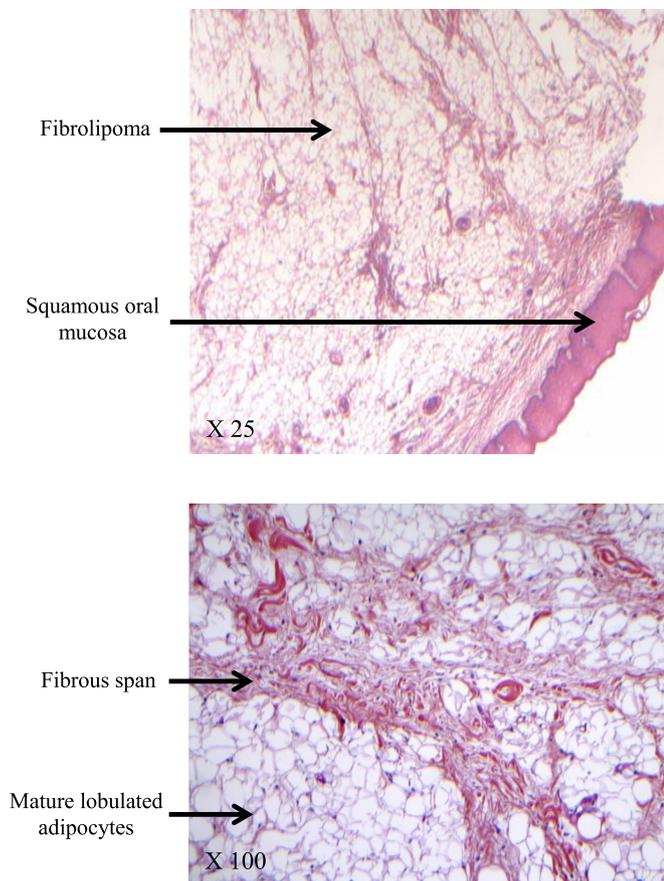
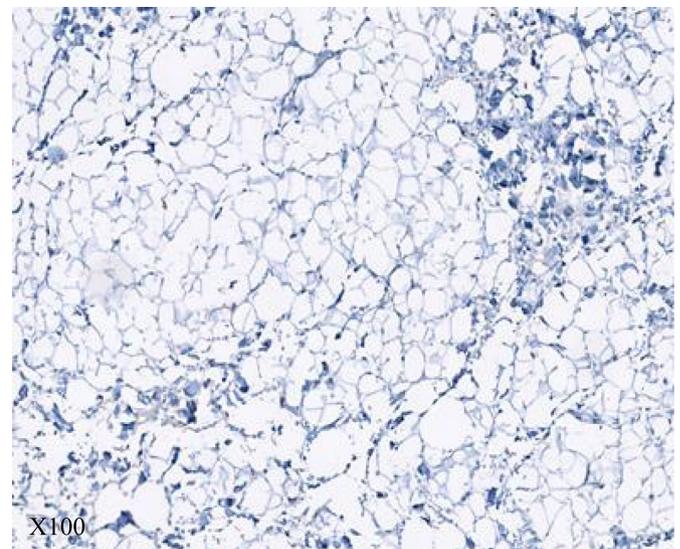


Fig. 4. Histological analysis of the lesion (hematoxylin eosin saffron staining): microscopic appearance in favour of a fibrolipoma and presence of numerous adipocytes separated by fibrous septa.

Radiologically, when the lipoma is superficial, it may be difficult to differentiate subcutaneous fatty tissue, using both computed tomography (CT) and MRI. A fibrous capsule should always be searched and MRI can differentiate an infiltrating lipoma from a well-limited lipoma [9,13]. Within the lesion, the morphology of the non-fat component (septa) as well as the fat volume allow to define several categories of lesions likely to have a benign or malignant character. In the presence of numerous septa, with irregular thickness or with high signal intensity, the tumor is regarded as suspicious [3]. Some lesions, however, remain difficult to classify. A lesion greater than 5 cm in its axis must also suggest some malignancy [3]. Regarding CT, this imaging technique determines the infiltrative nature of the lesion into bone tissue, which can provide some information on the malignancy or not of the lesion [1,6]. Medical ultrasound is also useful for the characterization and the follow up of a typical superficial lipoma [1]. Easily available and inexpensive, ultrasound will be the first-line examination in this context [10], showing a homogenous and hypoechoic mass [1,10].

On a histological point of view, lipomas can be classified as simple lipomas or variants, such as fibrolipomas, spindle cell lipomas, intramuscular or infiltrating lipomas, angioliipomas, salivary gland lipomas, pleomorphic lipomas, myxoid lipomas,



Anti-MDM2 Immunohistochemistry

Fig. 5. Immunohistochemical analysis of the lesion using anti-MDM2 antibodies. No labeling of this antibody is observed, thus eliminating a malignant tumor.

and atypical lipomas. Fibrolipoma is the second most common form after the simple lipoma, and is characterized by more abundant and thicker fibrous spans [4,5,11]. The submucosal lesion was well-defined, consisting of mature adipocyte lobules separated by fibrous septa with various thickness (Fig. 4). The lesion was highly vascularized by fine vessels (with no histological feature). The non-keratinized squamous surface epithelium showed focal parakeratosis and rested on lamina propria.

ALT/WDLs is distinguished from benign adipose lesions by the presence of mature adipocytes, but with significant anisocytosis, as well as nuclear atypia, such as hyperchromasia or multinucleation of stromal cells in fibrous septa [13]. Immunohistochemistry (marking with anti-MDM2 and/or anti-CDK4 antibodies) and molecular biology (looking for amplification of MDM2 and/or CDK4 genes by FISH or RT-PCR) provide decisive arguments in favour of malignancy. The overexpression of MDM2 (nuclear phosphoprotein capable of inhibiting the activation domain of p53, a protective protein against malignant transformation) and/or CDK4 (having a catalytic kinase function) related to an amplification of their genes is observed in ALT/WDLs [13,14]. For this patient, the benign nature of the lesion was confirmed using the anti-MDM2 antibody, which revealed no labeling (Fig. 5). The diagnosis of oral fibrolipoma was therefore established.

The treatment of choice consists of a complete surgical resection with systematic histological analysis. Medical treatment may be considered, consisting in the injection of corticosteroids to achieve a lesion atrophy [8,10]. In the article published by Volpato *et al.* on a lipoma of the palate, a low-intensity laser therapy in several sessions was used to improve the post-operative healing and comfort of the patient [9]. In superficial locations, the lipoma with fibrous septa makes it a

well-limited and easily accessible lesion. However, in the presence of contact with the salivary glands, the facial nerve and the floor of the mouth, as well as a deeper anatomical location, the clinician must re-evaluate the benefit-risk balance and the surgical indication [2].

Conclusion

This case report on fibrolipoma reveals the importance of complementary examinations. MRI, along with the analysis of the fat component and of the fibrous septa, is used to help the clinician in the diagnosis of malignancy of this type of lesion, particularly the well-differentiated liposarcoma [3].

The complete excision of the lesion and the histological analysis allow to establish the diagnosis with (if necessary) the use of immunohistochemistry (anti-MDM2 and/or anti-CDK4). Surgical treatment is the treatment of choice associated with a follow-up, although recurrence is rare [2,6-8,10,12].

Conflicts of interest: The authors declare that they have no conflicts of interest in relation to this article.

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