Ossifying chondrolipoma of the tongue

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Abstract

Introduction. Chondrolipomas and osteolipomas are uncommon variants of lipomatous tumors. Case report. We presented a 60-year-old woman with ossifying chondrolipoma of the tongue. Clinical examination revealed a firm nodular mass, located in the midline of the posterior region on the dorsal surface of the tongue. Histologically, the lesion was well-delimited showing areas of mature adipocytes arranged in lobules and separated by fibrous connective tissue septa, islands of mature cartilaginous tissue and osseous metaplasia. Trabeculae of lamellar bone within a fibro-fatty background were visible throughout the tumor. The cartilaginous areas merging centrally with bone formation and fatty marrow tissue were present, as well as the hematopoietic elements in the fatty marrow. The bone forming was found to be through both membranous and enchondral mechanisms. Conclusion. Ossifying chondrolipoma with hematopoietic elements is extremely unusual lesion. This interesting entity should be kept in mind in the differential diagnosis of lingual lesions.

Key words: tongue neoplasms; neoplasms, complex and mixed; chondroma; osteoma; lipoma; mesenchymoma; diagnosis; diagnosis differential.

Introduction

Lipomas are very common benign soft tissue tumors. They may occur anywhere in the body, but usually present as slow-growing, solitary and asymptomatic subcutaneous or superficial lesions. Histologically, lipomas are composed of mature adipocytes arranged in lobules that are separated by fibrous connective tissue septa, and occasionally associated with one or more secondary mesenchymal elements. Different variants of lipoma have been described, such as fibrolipoma, angiolipoma, myxolipoma, spindle cell lipoma, chondroid lipoma, chondrolipoma, and osteolipoma. Although lipomas are one of the most common soft tissue tumors, with about 20% of cases affecting the head and neck region, only 1%–4% of these neoplasms involve the oral cavity. Oral lipomas usually develop in patients over the age of 40. The buccal mucosa is the most affected site, and lipomas and fibrolipomas are being the most frequently observed in the oral cavity. Among the other histopathological variants, lipomas with cartilaginous or osseous metaplasia, called chondrolipomas or osteolipomas, respectively, are rare in the oral cavity. Only 10 cases of chondrolipoma have been reported in the literature, with a total of 15 cases in the international literature.
Oral osteolipomas are less common than chondrolipomas. A review literature revealed 8 cases of this variant located in the oral cavity. Lipomas with cartilaginous and osseous metaplasia have been described in the subcutaneous and deep soft tissues, particularly in the parosteal localization. To our knowledge, there is no case of this variant located in the oral cavity reported in the literature. We present an unusual case of ossifying chondrolipoma of the tongue containing hematopoietic elements and discuss the pathogenesis and the differential diagnosis.

**Case report**

A 60-year-old female was referred to the Department of Otorhinolaryngology with a painless mass on the dorsum of her tongue which caused discomfort during chewing. The lesion was first noticed about 5 years earlier and had not grown remarkably so far. The patient reported no previous trauma to the affected region. Her medical history was non-contributory.

Clinical examination showed a well-defined nodular mass of firm consistency, which was located in the midline of the posterior region on the dorsal surface of the tongue (Figure 1). The remainder of intraoral examination was otherwise unremarkable, with no evidence of submandibular and cervical lymphadenopathy. The lesion was completely excised, under local anesthesia, and sent to the Department of Pathology for examination.

Grossly, the excised specimen consisted of firm, yellowish-white oval nodule, measuring 20 x 17 mm, covered by normal mucosa. Histologically, the tumor showed areas of mature adipocytes arranged in lobules and separated by septa of fibrous connective tissue (Figure 2). The amount of fibrous septa was increased. Islands of mature cartilaginous tissue were identified in the proximity to the fibrous connective tissue septa (Figure 3). These cartilage islands containing lacunae filled with chondrocytes were variable sizes and surrounded by adipose tissue. Trabeculae of lamellar bone within a fibro-fatty background were visible throughout the lesion, especially toward the central areas (Figure 4). They were surrounded by spindle-shaped mesenchymal cells, and a small rim of osteoblasts was observed. A limited myxoid change was present but no necrosis was observed. In addition, partly cartilaginous areas merging centrally with bone formation and fatty marrow tissue were found (Figure 5), suggesting that a part of the bone had been formed through the enchondral mechanism. There were hematopoietic elements also present in the fatty marrow tissue (Figure 5). No prominent inflammatory reaction was detected. The tumor showed both enchondral and membranous ossifications. The bone forming di-

Fig. 1 – Clinical view of nodular lesion in the midline on the dorsum of the tongue

Fig. 2 – Well-demarcated collection of mature adipocytes arranged in lobules separated by septa of fibrous connective tissue (HE, ×40)

Fig. 3 – Islands of mature cartilage in the proximity to the fibrous connective tissue, surrounded by mature adipocytes (HE, ×40)

Fig. 4 – Bone trabeculae surrounded by thick fibrous connective tissue septa and areas of mature adipose tissue (HE, ×40)
rectly from the fibrous background (membranous ossification) predominated in this lesion. No nuclear atypia or mitotic figures were observed. The tumor was well-delimited and covered by normal surface mucosa. The lesion was diagnosed as ossifying chondrolipoma with hematopoietic elements. The recovery course was uneventful, and after a 2-year follow-up, the patient showed no signs of recurrence.

**Discussion**

Lipomas of the oral cavity are uncommon and represent about 1%-5% of all neoplasms of the oral cavity. They are slow-growing tumors and usually presenting a mean diameter of 2.0 cm at the time of diagnosis. Variants of lipoma with cartilaginous or osseous change have been called as chondrolipoma, osteolipoma, lipoma with cartilaginous or osseous metaplasia, or ossifying lipoma. These tumors are rarely observed in the oral cavity. Osteolipomas are less common than chondrolipomas, similar to finding in the extraoral soft tissue. There currently are no more than 8 reported cases of osteolipomas in the oral cavity. While oral lipomas are believed to be more common in men, it seems that there is no clear sex preponderance in patients with osteo/chondrolipomas in the oral cavity.

Chondrolipomas are characterized by the proliferation of mature adipocytes with additional mature cartilaginous tissue formation. Cases of chondrolipoma of the oral cavity have been diagnosed in patients aged from 2 to 72 years, and most of the tumors are found in the tongue and in the lower lip. Osteolipomas, in turn, are characterized by formation of bone trabeculae scattered among proliferating mature adipocytes. Most cases of oral osteolipoma are diagnosed after the fifth decade of life. The most affected sites are the buccal mucosa and vestibulum, tongue, and alveolar mucosa. Osteolipomas have been reported in patients with a long history measured in years, as in our patient.

The case presented in this report shows three rare alterations. First, mature cartilaginous tissue was a prominent component of the lesion. Usually the cartilage in lipoma represent a small part of the tumor, but lipoma of the tongue with marked cartilaginous metaplasia and chondrolipoma of the tongue with two main components (adipose and cartilage) have been reported. Second, this lesion exhibited osseous metaplasia. The bone was found to be formed through the membranous and the enchondral mechanisms. Lipomas showing cartilaginous and osseous metaplasia were observed in the subcutaneous and deep soft tissues, often in the parosteal localization. Rau et al. reported a case of parosteal lipoma with extensive areas of cartilaginous and osseous differentiation, considered as an osteochondrolipoma. This authors identified bone formation through both enchondral and membranous mechanisms. Bone forming directly from the fibrous background (membranous ossification), typically seen in heterotopic bone formation, predominated in our case. Third, in the fatty marrow tissue of the lesion, hematopoietic elements were identified. Extramedullary hematopoiesis is known to be an integral part of myelolipoma arising in the adrenal glands most commonly, but occasionally in other sites including pelvic soft tissue. This phenomenon is rarely seen in the other lipoma variants, and to our knowledge none of the intraoral cases reported to date contained hematopoietic elements.

The differential diagnosis of osteo/chondrolipomas depends on their location. When osteolipomas affecting alveolar mucosa, lesions such as tori and exostoses have to be taken into consideration, but a histological finding of mature ossification with fatty tissue usually can help with diagnosis. In the cases arising in the tongue, such as the presented case, osteo/cartilaginous choristomas should be considered, as they commonly affect the tongue. However, these lesions consist almost entirely of cartilage and/or bone and usually contain less fatty tissue. In addition, mature cartilaginous areas in a lipoma should be distinguished from chondroid lipoma. This lipoma variant is uncommon in the oral cavity, consisting of mature adipocytes and multivacuolated cells in a myxohyaline matrix that has a chondroid appearance. Chondroid lipoma, a newly described lesion, having an immature aspect may be mistaken for malignancy, while chondroidlipoma can easily be identified as benign tumor. As chondroid lipoma is integrated into the newest classification of lipomatous lesions, Rau et al. proposed that chondrolipomas, osteolipomas, and osteochondrolipomas should also be included as one subvariety of lipomas.

The nomenclature of osteo/chondrolipomatous lesions is controversial. Hietanen and Makinen reported an example of chondrolipoma of the tongue and stated that the tumor meets the criteria of benign mesenchymoma, as well. According to the inclusion criteria by Jones et al., benign mesenchymoma is an unencapsulated tumor composed of two or more mature mesenchymal tissues (excluding fibrous connective tissue), with no single mesenchymal element predominating in respect to others. In contrast, in osteo/chondrolipomatous tumors a predominating component is a mature fatty tissue, while chondroidlipoma can easily be identified as benign tumor. As chondroid lipoma is integrated into the newest classification of lipomatous lesions, Rau et al. proposed that chondrolipomas, osteolipomas, and osteochondrolipomas should also be included as one subvariety of lipomas.

amount of fibrous septa predominated, although areas of cartilage were prominent and osseous metaplasia was notable.

The pathogenesis of these lipomatous tumors is uncertain. Different theories have been proposed to explain the formation of cartilaginous and osseous tissues in lipomas. One theory suggests that adipose, cartilaginous and osseous components originate from multipotent undifferentiated mesenchymal cells. Alternatively, cartilaginous and osseous components may represent a metaplastic process in pre-existing lipoma. According to Katzer, the formation of cartilage and bone within lipomas can be explained by combination of local trauma with a special reactivity of the mesenchymal cells, which is possibly influenced by localization (i.e. proximity to periosteum). Mesenchymal cells can be modified by local or systemic factors such as permanent mechanical stress, repeated microtraumas, and reduced blood supply. These factors can act as cartilaginous and osseous differentiation inducing agents within lipoma. The origin of these lesions from multipotent cells seems attractive, as these cells have been identified in adult differentiated fat tissue. In vitro and animal models showed a multidirectional differentiation capacity of adipose tissue-derived stem cells. This permits formation of bone, cartilage, fat, muscle, blood vessels and fibrous tissue form the same precursor cells.

Rau et al. well documented two ways of bone formation in their case of parosteal osteochondroplasia, suggesting that a more pleomorphic differentiation capacity of deeply located lipomatous tumors might reflect the existence of more multipotent stem cells. In this regard, osteo/chondroplasmas arising from the oral soft tissues presumably reflect the multipotent nature of undifferentiated mesenchymal cells of this region. The presented case had no history of trauma preceding the appearance of the lesion. Thus, it might be speculated that the chronic irritation of the lesion combined with the local microenvironment was a significant inducer of osseous metaplasia with hematopoiesis.

**Conclusion**

Despite controversies on nomenclature and pathogenesis of osteo/chondroplasmas of the oral cavity, complete surgical excision is the treatment of choice. There have been no reports on recurrence. These rare tumors must be kept in mind both clinically and pathologically when evaluating lingual lesions. The presented ossifying chondroplasia with hematopoietic elements is extremely unusual lesion. Further studies are necessary for more detailed characterization of this interesting entity.

**REFERENCES**


