

Unsuspected Odontogenic Fibromyxoma the Mandible – a Clinical Case

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Abstract

Odontogenic fibromyxoma represents a rare benign odontogenic tumor of mesenchymal origin which exhibits locally aggressive behavior and is prone to local recurrence and affects mostly the posterior region of the mandible. We present a case report of a 60-year-old male with a swelling of the left mandible. The clinical findings are atypical of a particular oral disease. Final diagnosis of odontogenic fibromyxoma is established through clinical and subsequent histopathological examination. Odontogenic fibromyxoma is a diagnostic and therapeutic challenge. The controversy in the treatment protocol for a patient with odontogenic fibromyxoma has mainly been focused on therapeutic management with recommendations varying, depending on the size of the tumor, from simple enucleation and curettage or wide excision to segmental bone resection.

Key words: mandible, myxofibroma, odontogenic myxoma

Background

The term “myxoma/ myxofibroma” is introduced by Virchow in 1871, when he described tumors that histologically resembled the mucinous tissue of the umbilical cord (1). The World Health Organization (WHO) defines myxoma as a locally invasive neoplasm consisting of rounded and angular cells that lie in an abundant mucoïd stroma. The tumor is usually poorly demarcated from the surrounding tissue with which it freely intermingles or from which it is separated by a pseudo capsule (1).

The term “odontogenic fibromyxoma (OFM) is often applied when the tumor occurs in the jaws to reflect its odontogenic origin (1,2). It is considered as a locally invasive benign mesodermal tumour found exclusively in the jaws comprising 3-6% of all odontogenic tumors. It usually occurs in the second and third decades of life (3). OFM is more common in females and located mainly in the mandible (4, 5).

Earlier theories suggest that OFM originates from the neural sheath or is a result of degeneration of fibromas, lipomas and others due to chronic irritation and the degenerative processes following tissue anoxemia. Recent studies reveal that myxomas/fibromyxomas arise from the mesenchymatous tissue of the dental follicle, thus being described as odontogenic with fibroblasts playing the major role in cell dispersal. This explanation fails to describe soft tissue myxomas. They probably arise from supportive structures of the teeth like the gingiva and the periodontal ligament (6, 7).

Clinically OFM is usually present as a slow growing asymptomatic swelling; however pain and paresthesia can occur in advanced stages. Facial asymmetry may occur due to large lesions. The lesion can be diffuse or well defined, uni- or multilocular. Displacement and mobility of teeth are relatively common. It may be associated with unerupted teeth. Cortical expansion can occur and large lesions can cause perforation (3, 8). Radiographic appearance may vary from a unilocular or multilocular radiolucent lesion to a mixed radiolucent–radiopaque lesion (3).

The clinical variables and radiological appearance make the diagnosis difficult in these tumors and histological examination reveals the diagnosis (3). Histologically these tumors are composed of a large amount of intercellular substance rich in acid mucopolysaccharides and made up of loose myxomatous connective tissue, fibroblasts and myofibroblasts. Patches of trabeculae of woven bone and capillaries are dispersed by the lesion (4).

Rare cases of OFM present with a diffuse and mottled appearance and can be mistaken for a malignant neoplasm (9).

Case Description

We report the case of a 60-year-old male (D.S. med. history 599) with history of a large lump in the mouth persisting for 20 years. In the last two years the swelling enlarged.

Clinical examination reveals a large swelling of the left mandible extending from the distal part of the second premolar to the mesial part of the third molar sized 4/5 cm. The formation is pedunculated, well defined, soft to firm in consistency on palpation, with even and reddish colour. Adjacent teeth show Grade II mobility and are migrated (Fig. 1).

Figure 1. Intraoral manifestation of OFM

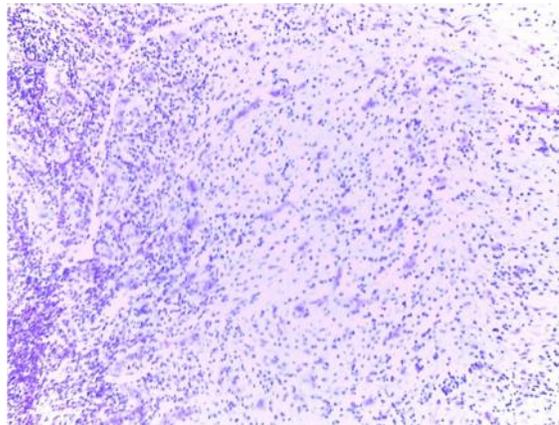


A panoramic radiograph is taken. It reveals well-defined mixed radiolucency in the area of the left molars extending from the distal root of 36 to the mesial root of 38 including the apices of the teeth.

The tumor formation is removed by electro-excision in clinically and histologically clear margins. The teeth 36, 37 and 38 were extracted. A marginal osteotomy is performed and the postoperative defect is closed with a vestibular flap.

The histological result (130903,4/19.06.2013) shows: Fibromyxoma.

Figure 2. The histopathological examination reveals a partially encapsulated myxomatous area composed of dense collagen fibres with few calcified structures and islands of odontogenic epithelium suggestive of odontogenic fibromyxoma



Discussion

There is a wide variety in the clinical and radiologic appearance of OFM, which is the most common form of presentation as an asymptomatic expansion in the jaw and a multilocular radiolucent image. Although these are slow-growing lesions, they show a persistent and destructive pattern. With growth they usually displace and resorb teeth causing pain and facial deformity (4). OFM of the maxilla behaves more aggressively than that in the mandible, as it spreads towards the maxillary sinus. Cortical expansion can occur and large lesions can cause perforations (6).

Diagnostic protocol

Usually diagnostic methods include conventional radiography and incisional biopsy with histological and histochemical investigation (8). Immunohistochemical examination uses antibodies against specific biological substances of neuronal, muscular, epithelial, and mesenchymal tissues. The evaluation of the presence of vimentin, an intermediate filament of the cytoskeleton characterizes mesenchymal tissues, thus also myxomas. Fibromyxomas also contain a high amount of hyaluronic acid (8). Less commonly ultrasound examination is performed (6), as are advanced imaging examination with Magnetic Resonance Imaging (MRI), and computed tomography (CT). These methods contribute to the differential diagnosis from other benign tumors, such as ameloblastoma (3, 8, 10, 11).

Radiological examination plays an important role for the differential diagnosis of myxomas/fibromyxomas and also for distinguishing between benign myxomas and malignant neoplasms with myxomatous tissue. In MRI, the lesion shows low-signal intensity in T1 and high-signal intensity in T2. In contrast, Kawai et al. advocate that high-signal intensity is shown in T1 and not in T2 (10). These discrepancies may be related to the ratio of fibrous/myxoid tissue, the viscosity, the concentration of proteins, the presence of hemorrhage and hypocellularity (8, 10).

In the presented case radiological examination includes only panoramic tomography. According to it the radiolucency doesn't show any peculiarities and wide differential diagnoses are possible.

Treatment procedures

The controversy has been focused mainly on therapeutic management with recommendations varying, depending on the clinical cases, from simple curettage of lesion to segmental bone resection (4).

Although curettage of the lesion is used for treatment, it is associated with a high recurrence rate of up to 25%. It typically occurs during the first 2 years after removal (4). The treatment of choice in large, destructive and expansile lesions is surgical removal with safety margins of at least 1.5 cm to prevent recurrence of the neoplasm. In most cases, tumor ablation usually involves the sacrifice of adjacent teeth (4). Recently, advanced imaging modalities such as CT and MRI are being applied to this tumor which possess the special predominance in detecting whether the adjacent bone and soft tissues are involved or not, and the exact extent of the lesion (4). Sloopweg and Wittkamp (12) comment that site of the myxoma should be taken into account when deciding on management plans.

The tumor is not radiosensitive, and surgery is the treatment of choice. The lack of a capsule and infiltrative growth pattern is responsible for the high recurrence rate (up to 25%) when conservative enucleation and curettage are performed. Recurrence is minimized with extensive partial or total resection procedures, and this method of treatment is particularly indicated in the maxilla due to the proximity of vital structures (3, 13, 14, 15).

Conclusion

The diagnosis of OFM is a correlation of clinical, radiological and histopathological features. A complete surgical excision along with proper long term follow up is essential, due to the possibility of recurrence of the myxomatous tumor and to the fact that the tumor is not radiosensitive. Surgery is the advised choice

of treatment. The primary treatment considerations include the age of the patient and the potential recurrence of the lesion.

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