REVIEW ARTICLE

Odontogenic myxoma: A review with report of an uncommon case with recurrence in the mandible of a teenage male

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Abstract We describe a 13-year-old boy with recurrence of an odontogenic myxoma of the mandible. We review the existing published literature on the lesion, emphasizing the similarities and differences among lesions in the differential diagnosis. Odontogenic myxoma is an uncommon benign tumor that mainly affects the mandible, with a peak incidence in the second to fourth decades of life and predilection for the female sex. Clinical, radiological, and histopathological features should be considered when making a diagnosis. Several of these characteristics overlap with those of other benign and some malignant tumors. Odontogenic myxoma is known for recurrence. The treatment plan should consider the age and sex of the patient and the site and size of the lesion. Reconstructive surgery may be required, but should be delayed until after an adequate follow-up to rule out recurrence.

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1. Introduction

Myxomas are rare tumors of the hard sclerous and soft tissues in the body. Odontogenic myxomas comprise only a small fraction of myxomas. Clinical and radiological features of odontogenic myxomas are variable and mimic those of other tumors of the region. The tumor may be an incidental finding or may cause symptoms, including pain, paresthesia, and tooth mobility (Gonzalez-Garcia et al., 2006; Li et al., 2006). Virchow has been credited with the coining of the word “myxoma” (Melo et al., 2008; Moore et al., 2008). Myxomas of odontogenic origin were first described by Thoma and Goldman (1947), on the basis of site of occurrence, age at occurrence, association with missing teeth, and histopathological examination, which showed structural resemblance with dental mesenchyme and the sporadic presence of islands of odontogenic epithelium.

Odontogenic myxoma is a benign but invasive tumor that has a high rate of recurrence after surgical removal (Speight, 2013). We describe the case of a 13-year-old boy who developed recurrence of an odontogenic myxoma of the mandible. We review the literature on the features, differential diagnosis, and treatment modalities of this lesion.

2. Case report

2.1. History and clinical examination

A 13-year-old boy reported to the outpatient clinic with complaints of a painless, gradually progressive firm swelling on the lower left side of the jaw of 9 months’ duration. He gave a history of a similar swelling in the same location 2 years before, for which he was treated intraorally at a dental clinic under local anesthesia 15 months prior. The patient had no documents pertaining to the previous treatment.

Clinical examination revealed a diffuse swelling, measuring about 2 x 4 cm with no clear borders, over the left angle of the mandible. Skin over the swelling was normal with no local rise of temperature, ulceration, or redness (Fig. 1). Intraoral examination did not show any swelling or breach in the mucosa on the left posterior region. On palpation, the swelling was diffuse, nontender, firm, and resilient to touch. Superoinferiorly, the swelling extended from near the left ear lobe to the angle of the mandible. Anteroposteriorly, it extended 4 cm anteriorly from the posterior border of ramus of the mandible. Expansion of the buccal and lingual cortical plate was palpable near the ramus of mandible.

2.2. Investigations and differential diagnosis

Orthopantomography (OPG) showed a poorly defined, multilocular, radiolucent lesion of 2.0 x 4.5 cm involving the left angle of the mandible, with a thickened bony rim (Fig. 2). Routine laboratory investigations (hematological and serological) were normal. Differential diagnosis included keratocystic odontogenic tumor, ameloblastoma, central giant cell granuloma, and odontogenic myxoma.

2.3. Surgical management

Given the patient’s age, the recurrent and progressive nature of the lesion, and the observed buccal cortical expansion, we recommended radical surgery, which was rejected by the patient’s parents due to domestic circumstances. After obtaining written informed consent from the patient and his parents, conservative surgical excision of the tumor with curettage was performed. Under general anesthesia, the lesion was exposed, together with the coronoid process, sigmoid notch, and condylar neck, through a submandibular approach. The lesion was completely excised and properly curetted. Searification of the cavity was done with an acrylic bur. The surgical wound was irrigated with normal saline, povidone-iodine, and hydrogen peroxide solutions. The patient’s parents were counselled and...
advised to consider radical surgery with later reconstruction in the case of a further recurrence.

Excised tissues were gel-like pieces with a mucinous appearance and a gray-white color. The tissue was sent for histopathological examination. A capsule was not evident on gross examination of the tissue (Figs. 3 and 4).

2.4. Histopathology

Histopathological examination of the resected specimen with hematoxylin and eosin staining revealed spindle-shaped cells dispersed in a background of loose and abundant mesenchyme. Cells had long cytoplasmic processes at either end. Inflammatory cells were occasionally seen. All cells were uninucleate. Nuclei did not show any abnormality or variation in number or structure. Collagen fibers were loosely arranged. Vascularity was sparse, mainly of fine capillaries. Nests or islands of odontogenic epithelium were not seen. Cellular atypia or mitotic cells were not seen (Fig. 5). Considering the clinical, radiological, and histopathological findings, we made a final diagnosis of odontogenic myxoma.

2.5. Follow-up

The patient’s 1-year follow-up clinical examination was uneventful and revealed no evidence of recurrence.

3. Discussion and review of related literature

Myxomas are very rare benign tumors of mesenchymal origin. These tumors are locally invasive (McFarland et al., 1996; Simon et al., 2004; Martinez-Mata et al., 2008; Zarbo, 2010; Speight, 2013) and occur in various tissues, such as the heart,
Odontogenic myxoma, also termed as odontogenic fibromyxoma or myxofibroma, is a subtype of myxoma occurring mainly in the hard, bony tissues of the face (Shafer et al., 2003), although the lesion may also occur in the surrounding soft tissues (Moore et al., 2008). We use the term “uncommon” to describe the present report of recurrence of odontogenic myxoma in a 13-year-old boy because these lesions are most prevalent in adult women (Shafer et al., 2003).

Odontogenic myxoma is usually a slow-growing mass with late-appearing symptoms, primarily due to the mass effect. Symptoms include pain, paresthesia, ulceration, and tooth mobility (Gonzalez-Garcia et al., 2006), although none of these symptoms were found in the present case. Odontogenic myxomas crossing the midline are rare (Simon et al., 2004; Li et al., 2006) probably because the symptoms cause the patient to present to a clinician before the tumor is too large. Myxomas crossing the midline are usually found in the mandible (Landa et al., 2002).

On gross examination, odontogenic myxoma appears as a grayish white, nodular heterogeneous mass of variable consistency (Canalis et al., 1976), with a glistening gelatinous cut surface (Landa et al., 2002). The tumor may have a minimal true capsule (Li et al., 2006) or may be uncapsulated and poorly demarcated from surrounding tissues (Rius et al., 2013). In the present case, there was no capsule.

The lesion mimics dental pulp in its histological features (Regezi, 2002). Microscopically, the tumor has a mucoid-rich ECM, with scattered cells, connective tissue fibers, bony trabeculae, irregular calcifications, scant blood vessels, and sparse capillaries. Nests of odontogenic epithelium are occasionally seen but not essential for diagnosis (Simon et al., 2004; Li et al., 2006; Melo et al., 2008; Chrcanovic et al., 2010). The ECM comprises eosinophilic mucoid tissue, which resembles connective tissue of the umbilical cord. Spindle-shaped or stellate cells with small hyperchromatic nuclei and cytoplasmic processes are interspersed in collagen or reticulin fibers (Stout, 1948; Lo Muzio et al., 1996). Cellular atypia is rare, and the presence of mast cells has been reported (Martinez-Mata et al., 2008). Fibers are oriented toward the tumor periphery. All of these classical features were seen in our case. Immunohistochemistry studies of odontogenic myxomas are thought to be of little value in differentiating these lesions from other nonodontogenic cell tumors (Raubenheimer and Noffke, 2012).
Table 1  Comparison of a few earlier studies on odontogenic myxoma.

| Researcher       | Type of study                              | Sample size          | Range of age in yrs (peak incidence in brackets) | Peak in_decade | Sites | Tooth displacement | Root resorption |
|------------------|--------------------------------------------|----------------------|-------------------------------------------------|----------------|-------|--------------------|-----------------|
| Kaffe et al.     | Systematic review with two case reports    | 164 (96-radiological)| 01–73 (most cases in 2nd to 5th decade)         | 2nd            | 55    | 109                | 26%             |
|                  | Retrospective                              | 62                   | 09–71 (most in 2nd to 4th decade)               | 3rd            | 25    | 37                 | 12 (19.3%)      |
| Martinez-Mata et al. | Retrospective-radiological              | 41                   | 04–63 (most cases in 1st to 5th decades)        | 3rd            | 17    | 24                 | Not mentioned   |
| Zhang et al.     | Retrospective-radiological                | 41                   | 11–70 (most cases in 2nd to 3rd decade)         | 3rd            | 11    | 19                 | Number not mentioned |
| Simon et al.     | Prospective                                | 33                   | 03 months–64 years (majority in 2nd to 4th decade) | 3rd            | 8     | 24                 | 10 out of 21 avbl cases |
| Noffke et al.    | Retrospective                              | 30                   | 11–70 (peak in 4th decade)                      | 4th            | 13    | 19                 | 13 (43%)        |
| Ajike et al.     | Retrospective                              | 27                   | 06–66 (peak between 2nd and 5th decade)         | 3rd            | 13    | 12                 | Not mentioned    |
| Li et al.        | Retrospective                              | 25                   | 08–45                                           |                | 5     | 9                  | 11 (48%)        |
| Friedrich et al. | Retrospective-radiological                | 14                   | 15–65                                           | 4th            | 4     | 6                  | 2               |
| Lo Muzio et al.  | Retrospective                              | 10                   | 10–40 (All between 2nd and 5th decade)          | 3rd            | 4     | 6                  | Number not mentioned |
| Abiose et al.    | Retrospective                              | 10                   |                                                 |                | 2     | Number not mentioned |

Sex predilection-mainly female predominance. One study – almost equal (Li et al.); Very slight male predominance-(Zhang). Age range – 03 months to 73 years. Site predilection-Mandible more affected than maxilla in all studies except one (Li et al.).

* In six cases-no information. Also excludes missing teeth in relation to the lesion.
Table 2  Comparative features of some lesions which may be considered as differential diagnoses of odontogenic myxoma. (Shafer et al., 2003; Altug et al., 2011; Soames and Southam, 2005; Neville et al., 2002; Whaites, 2002).

| Lesion                  | Odontogenic myxoma | Radicular cyst | Odontogenic keratocyst | Ameloblastoma | Central giant cell granuloma | Aneurysmal bone cyst | Osteosarcoma |
|-------------------------|--------------------|----------------|------------------------|--------------|------------------------------|---------------------|--------------|
| **Usual age of incidence** | 2nd–4th decade    | 2nd–5th decade | 2nd–4th decade         | 3rd–4th decade | Adolescents; usually below 30 yrs | Adolescents; usually below 20 yrs | Young, under 30 years |
| **Usual site of lesion**  | More in the mandible than the maxilla. | Apex of any non vital tooth. Especially upper lateral incisors | Posterior mandible/canine region of maxilla | Mainly the mandible. Maxilla—very occasional | Mandible. Often crosses the midline | Mandible. Occasionally in maxilla | Usually the mandible |
| **Sex predilection**     | Female predominance | Not significant | Slight male predilection | Not significant | Equal. Slight female predilection | Not significant | Male predominance |
| **Radiological appearance** | Not well defined | Smooth, well defined, well corticated if long-standing & if not infected | Smooth, well defined. Little mediolateral expansion | Smooth, scalloped, well corticated | Smooth, rarely perforates the cortical bone | Smooth, well defined. Cortex usually retained even when large. Buccal and Lingual expansion of cortex | Poorly defined—‘moth eaten’ appearance. Unilocular, widening of periodontal ligament space. Classical but rare is sunray appearance Usually unilocular |
| **Loculation**           | May or may not be seen | Unilocular | Pseudo/multilocular | Multilocular. May be unilocular in initial stages | Multilocular. May be unilocular in early stages | Seen | Faint |
| **Trabeculae/septae**    | Occasional. If present, very fine. Calcification is seen occasionally | Not seen. Calcification may be present | Not described | Seen | Faint | Sclerosing form shows irregular spicules and trabeculae-Sunray appearance |
| **Root resorption**      | Common | Rare | Common | Common | Sometimes | Rare | Spiking resorption |
| **Tooth displacement**   | Very common | Rare | Rare/minimal | Common | Often | Often | (Widening of periodontal ligament space is very characteristic) |
| **Histopathology**       | Matrix/cavity      | Bland appearance | Inflammatory infiltrate in the connective tissue just adjacent to the wall is a characteristic feature. Cavity usually contains fluid with low protein content | Cavity filled with cheesy material or with clear fluid | Cystic changes & Squamous metaplasia is seen | Characteristic is few to many multinucleate giant cells. In a loose fibrillar connective tissue stroma | Blood-filled spaces of varying sizes separated by fibrous tissue | Irregular new osteoid formation seen. Widely variable atypical osteoblasts are seen |
| Capsule | Fibrous CT wall lined by stratified squamous epithelium. Hyaline/rushton bodies may be present |
|---------|------------------------------------------------------------------------------------------|
| Cellular atypia | Not seen | Occasional dysplasia present | Not seen | Multinucleated giant cells | Multinucleated giant cells | Atypical neoplastic osteoblasts arranged irregularly around bony trabeculae |
| Nucleus | Single, Occasionally hyperchromatic | Single | Single | Few or several dozen nuclei in each giant cell | Single | Large deeply staining |
| Fibers | Not present in lumen | Not seen | Varying amount seen | Collagen fibers not usually in bundles | Not seen | Anaplastic fibroblasts are seen in the Fibroblastic variety of the tumor |
| Recurrence | More than 25% | No. But a follow-up for a minimum of two years is strongly advised | 30%. Most recurrences are in mandibular lesions | 50–90%. Five year disease free period is not indicative of cure | 15–20%. But recurrence rates of 50% have also been reported | Very variable. 8–60% | Known. More often in maxillary tumors |
In our case, the radiological appearance was of a unilocular, mixed radiolucent-radiopaque type with corticated margins. The radiological appearance of odontogenic myxoma has been variably described as always radiolucent, usually radiolucent, or mixed radiolucent-radiopaque (Kaffe et al., 1997; Li et al., 2006; Melo et al., 2008; Altug et al., 2011). Margins of the lesion are classified as corticated, noncorticated, poorly defined, or diffuse (Noffke et al., 2007). The tumor may be uni- or multilocular on radiographs (Lo Muzio et al., 1996; Altug et al., 2011), with multiloculated lesions being larger than unilocular ones (Kaffe et al., 1997). The appearance is variably described as mottled, soap-bubble (Zarbo, 2010), tennis racquet (Noffke et al., 2007), or honeycombed (Shafer et al., 2003). The honeycomb appearance seems to be restricted to mandibular lesions (Friedrich et al., 2012). The lesion shows “wispy” bony trabeculae within radiolucent areas (Li et al., 2006). Zheng et al. (2007) classified the radiological appearance of odontogenic myxoma into six groups.

The tumor will usually displace adjacent teeth, and root resorption has been infrequently reported (Shafer et al., 2003; Noffke et al., 2007; Chrcanovic et al., 2010). Accurate diagnosis is difficult based on routine radiographs alone, as the features of odontogenic myxomas overlap with those of other lesions. Differential diagnoses suggested based on radiological appearance of odontogenic myxoma include ameloblastoma, intraosseous hemangioma, aneurysmal bone cyst, glandular odontogenic cyst, central giant cell granuloma, cherubism, metastatic tumor, simple cysts, odontogenic keratocyst, and osteosarcoma (Abiose et al., 1987; Li et al., 2006; Chrcanovic et al., 2010). Fibromyxoid sarcoma, myxoid chondrosarcoma, and rhabdomyosarcoma should also be ruled out (Speight, 2013). Some differentiating features of the lesions are described in Table 2. Diagnosis is made on the basis of clinical features, radiographic appearance, and histopathology. Computerized tomography, magnetic resonance imaging, immunohistochemistry, and ultrastructural studies can aid in the correct diagnosis. PET scans may help rule out metastasis (Guo et al., 2014).

Surgery is the treatment of choice, with the treatment protocol depending on the site and size of the tumor. Complete extirpation of the tumor is difficult because infiltration may be more extensive than that observed clinically. Surgery types vary from enucleation and curettage, wide excision, and resection, to radical surgeries involving resection of adjacent tissues (Halfpenny et al., 2000). Allphin et al. (1993) recommended an initially conservative approach, followed by radical surgery if required. When radical surgery is performed, delayed reconstruction has been advised because of the high recurrence rate (Leiser et al., 2009). Odontogenic myxoma is radioresistant (Shafer et al., 2003). Although a few researchers advised pre- or postoperative radiotherapy (Attie et al., 1966; Cuestas-Carneiro et al., 1988), the present consensus is that radiotherapy has no role in the management of odontogenic myxoma. In the present case study, an excisional biopsy was preferred over incisional biopsy to give the patient the best possible treatment under conditions beyond our control. The tumor was situated on the posterior-most part of ramus; therefore, an extroral approach was chosen for reasons of accessibility and extension of exposure if required.

Odontogenic myxoma is notorious for a high recurrence rate of up to 25% after curettage (McFarland et al., 1996; Speight, 2013). A minimum follow-up period of 5 years without recurrence is recommended by some researchers before performing reconstructive surgeries. (Leiser et al., 2009). Rocha et al. (2009) reported a case of recurrent odontogenic myxoma 30 years after surgical treatment, which they treated by combining excision and curettage with cryotherapy.

4. Conclusion

Odontogenic myxoma is an uncommon entity occurring mainly in the bones of the face and jaws. Its clinical, radiological, and histopathological features mimic those of several other tumors and cysts. A high index of suspicion is to be maintained for accurate diagnosis and to rule out potentially malignant lesions. There is no consensus on the treatment protocol to be followed. In the present case of a growing patient, conservative surgery with strict clinical follow-up was recommended because the lesion has a significant chance of recurrence. Wider surgical excision should be employed in the case of recurrence. We recommend that large multicenter studies and systematic reviews of odontogenic myxoma be performed.

Conflict of interest

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