Case Report

Odontogenic myxoma in an 8-year-old girl: A case report with review of literature

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Abstract

Aggressive enlargements of maxilla in pediatric patients are uncommon and present with diagnostic and therapeutic dilemma. The decision on therapeutic modality is based on an early and accurate diagnosis, minimizing disease-associated morbidity which is of utmost importance considering the young age and thereby resulting in better prognosis. Odontogenic myxoma is a locally aggressive lesion which is primarily seen in relation to odontogenic apparatus in mandibular posterior region in association with an impacted tooth. This presentation describes a unique case of odontogenic myxoma of anterior maxilla in an 8-year-old girl with emphasis on its diagnosis and treatment planning.

Keywords: Maxilla, odontogenic myxoma, pediatric neoplasm

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INTRODUCTION

The term myxoma was coined by Virchow in 1863 to describe a group of tumors that had histologic resemblance to the mucinous substance of the umbilical cord. Myxomas are benign, slow-growing and locally aggressive mesenchymal neoplasms. They can be found in various body parts such as skin, subcutaneous tissue and the heart (left atrium), but rarely in the head and neck region.[1] Myxoma of the head and neck region can occur either in the subcutaneous tissue or the facial bones with increased predilection to occur in relation to odontogenic apparatus.[2]

Thoma and Goldman first described odontogenic myxoma of the jaw in 1947.[3] It is a nonencapsulated benign tumor of the jaws which can be divided into two groups: (1) tumors that arise specifically in jaw bones (most common type) and (2) those that arise in soft tissues of that area. The origin of odontogenic myxoma is believed to be from the ectomesenchyme of a developing tooth or from undifferentiated mesenchymal cells in the periodontal ligament.[4]

Odontogenic myxoma mostly occurs in the second or third decade of life and is common in mandibular posterior region.[5] Smaller lesions are asymptomatic while large lesions cause painless expansion of bone. Radiographically, myxoma shows unilocular or multilocular radiolucency with irregular or scalloped margins. Microscopic picture shows stellate-shaped cells dispersed in a loose myxoid stoma.[6] We report a unique case of odontogenic myxoma in an 8-year-old girl affecting the anterior maxillary area which is a rare occurrence, both in terms of age and site.

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CASE REPORT

An 8-year-old girl reported with painless, gradually enlarging swelling in anterior maxilla for 3 months. On examination, a well-circumscribed growth was seen in the anterior maxillary region measuring 4.1 cm × 4 cm in size extending from labial vestibule to the palatal bone in the region deciduous first molar teeth. Overlying mucosa was inflamed with area of ulceration due to incisional biopsy [Figure 1].

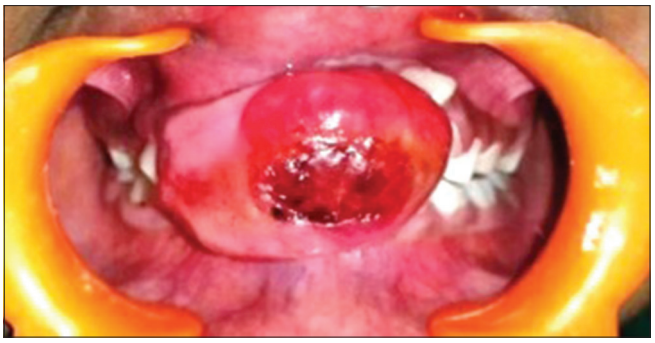

Figure 1: Inflammed overlying mucosa

On palpation, growth was firm in consistency, nontender without any bleeding on probing. Lymph nodes were not palpable. Radiographic picture showed radiolucency with defined borders in the anterior maxillary area, buccal and lingual cortical plates were destroyed and developing permanent central and lateral incisors were seen to be displaced. Based on the radiographic evaluation, provisional diagnosis of Juvenile ossifying fibroma and osteosarcoma was considered [Figures 2 and 3].

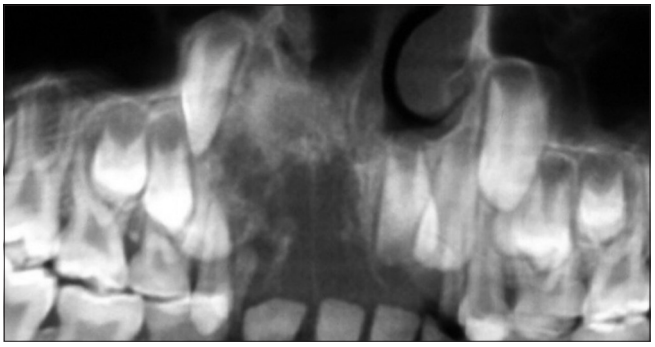

Figure 2: Orthopantomogram view

Incisional biopsy performed at another institute gave a histopathological diagnosis of aggressive fibrous lesion.

Considering the extent of lesion and incisional biopsy report, complete surgical excision with partial maxillectomy was performed and the excised tissue was sent for histopathological analysis.

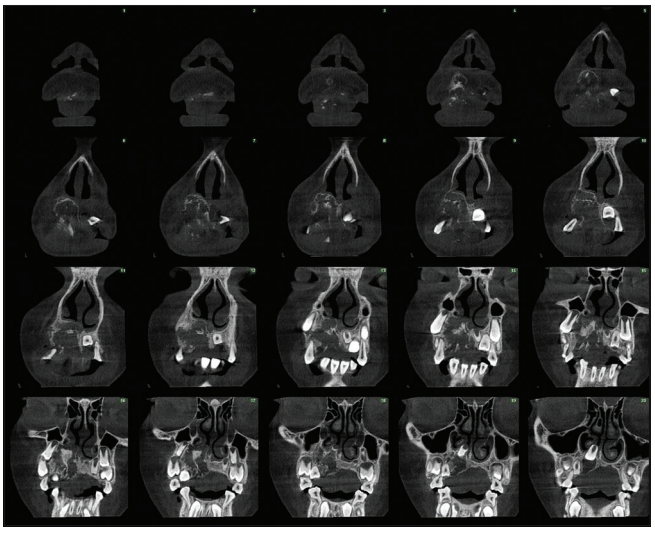

Figure 3: Computed tomography scan view

Macroscopically, the specimen received in our laboratory measured 3 cm × 4 cm × 2 cm and composed of both soft and hard tissue. The macroscopic appearance of the lesion showed slimy surface and firm in consistency. Deciduous teeth and impacted permanent teeth were present within the lesion.

Microscopic examination showed a well-circumscribed lesion consisting of myxoid areas composed of sparsely distributed stellate-shaped fibroblasts and odontogenic islands in a loose stroma [Figure 4]. The presence of inactive odontogenic islands in the deeper connective stroma was evident [Figure 5]. The peripheral capsule was densely fibrous with reactive bone formation.

Based on the above findings, the lesion was diagnosed as odontogenic myxoma. On 6-month follow-up, uneventful healing was observed.

DISCUSSION

Odontogenic myxoma is a rare aggressive intraosseous lesion. Myxomas are most commonly associated with unerupted teeth and derived from mesenchymal tissue of the tooth germ consisting of a myxomatous ground substance with scattered undifferentiated spindled mesenchymal cells.[7] Although it is a benign neoplasm, it may be infiltrative, aggressive and may recur.[8]

The tumor occurs across an age group that varies from 22.7 to 36.9 years. It is rarely seen in patients younger than 10 years of age.[9] In our case, it was seen in an 8-year-old girl which is an unusual finding. The mandible appears to be more frequently affected than the maxilla, especially the posterior region. However, cases have been

reported of odontogenic myxoma appearing in posterior maxilla.[8] Anterior maxillary involvement in our case is a rare occurrence.

The aggressive nature of odontogenic myxoma is well documented in the literature. Odontogenic myxoma of the maxilla is less frequent, but behaves more aggressively than that of the mandible, as it has tendency to spread to the maxillary sinus.[10]

Odontogenic myxoma grows without symptoms as a painless swelling and gets noticed only after reaching a considerable size. Radiographic picture can show either unilocular or multilocular appearance with defined or diffuse borders. A unilocular radiolucency is generally seen in children and in lesions present in the anterior region of the jaw.[11] It is a benign neoplasm without encapsulation. A spectrum of fibrous connective tissue stroma is present ranging from myxoid to densely hyalinized and from relatively acellular to cellular appearance.[12] Calcification may or may not be present. It is distinguished by the presence of sparse cords and islands of inactive odontogenic epithelium.[13] Sivakumar and Kavitha suggested that odontogenic myxoma is a tumor of a dual fibroblastic-histiocytic origin and the cells comprising odontogenic myxoma are of myofibroblastic origin.[14]

It is not radiosensitive, and surgery is the treatment of choice.[15] The infiltrative growth pattern is responsible for high rate of recurrence when conservative enucleation and curettage are performed.[16] However, Bravo-Burguillos *et al.* stated that conservative surgical excision done in infants did not show any clinical or radiographic evidence of recurrence for 3-year posttreatment follow-up.[17] Recurrence is minimized with extensive partial or total resection of the lesion. While doing resection particularly in the maxilla, proximity of vital structures should be considered.

A summary of odontogenic myxoma documented in the literature during the period from 2010 onward is presented in Table 1.

CONCLUSION

A unique case of odontogenic myxoma is reported in an 8-year-old girl involving the anterior maxillary region, and according to the literature, occurrence of this lesion in patients <10 years of age is rare. Odontogenic myxoma shares common clinical and radiographic features with various other odontogenic and fibrous tumors. To establish accurate diagnosis and treatment plan, histopathological diagnosis is of utmost importance.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form the patient(s) has/have given his/her/their consent for his/her/their images and

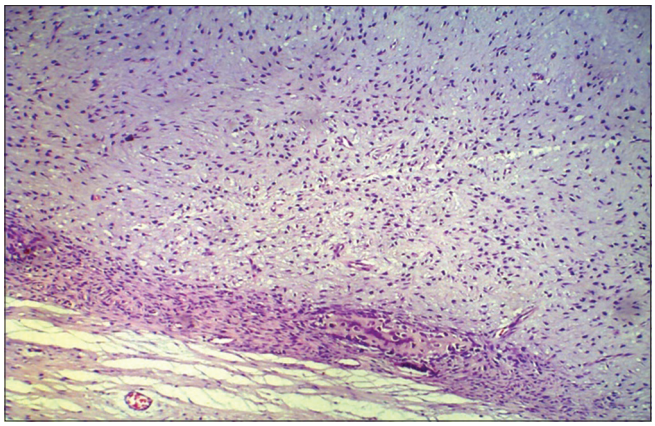

Figure 4: Microscopic view (×10)

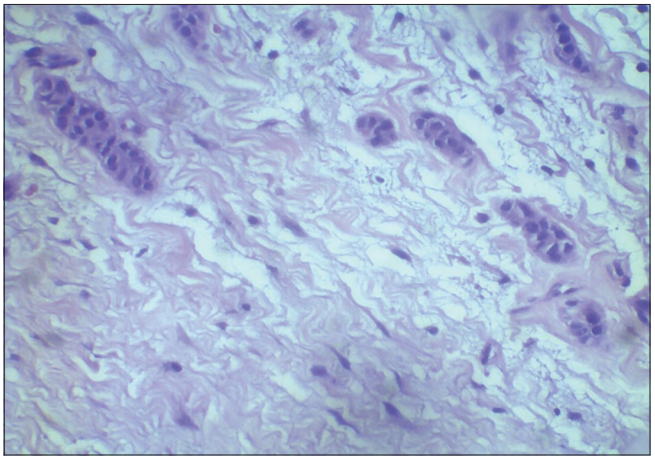

Figure 5: High-power magnification

Table 1: Summary of odontogenic myxoma affecting the maxilla documented in the literature

Author name	Site	Treatment	Age of patient/gender
Sasidhar Singaraju *et al*.	Middle third of maxillary region	Complete resection of lesion	7-year-old boy
Ajaz Shah *et al*.	Middle third of maxillary region	Complete resection of lesion	37-year-old female
Karuna Jindwani *et al*.	Posterior maxilla	Complete resection of lesion	10-year-old boy
Vijeev Vasudevan *et al*.	Posterior maxilla	Complete resection of lesion	13-year-old boy
Mohammad Asif Kiresur and Sathyavanthan Hemavathy	Posterior maxilla	Complete resection of lesion	17-year-old male
Akash P. Nehete *et al*.	Posterior maxilla	Complete resection of lesion	21-year-old male
Iqbal Ali, Ram Kumar Srivastava *et al*.	Posterior maxilla	Complete resection of lesion	35-year-old male

other clinical information to be reported in the journal. The patients understand that their names and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

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Conflicts of interest

There are no conflicts of interest.

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