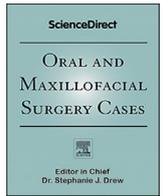




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Huge erupted complex odontoma in maxilla

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ABSTRACT

Objective: Odontoma is the most common odontogenic origin benign tumor considered to be hamartoma. It is characterized by its slow-growth and non-aggressive behavior. Those erupting to oral cavity are called “erupted odontomas” are rarely seen.

Methods: Here we report a rare case of a large erupted complex odontoma in maxilla, presenting clinical and radiographic features and treatment.

Results: Most odontomas are asymptomatic, but those erupting to oral cavity may cause severe infection. Bone loss and teeth impaction will greatly influence patients’ function.

Conclusions: Routine X-ray examination is necessary in mixed dentition period. Once odontoma is diagnosed, early surgical removal is the best choice.

Odontomas are the most common odontogenic origin benign tumor considered to be hamartoma. They are composed by enamel, dentin, cementum and pulp. According to the World Health Organization classification of 2017, there are two types of odontomas: complex and compound odontomas. Patients suffered odontomas are always symptomless, but often associated with teeth eruption disturbances. It shows no aggressive behavior and slow-growth. Eruption of complex odontoma in the oral cavity is extremely rare. The case happened in maxilla was first reported in 1985 by Liu [1]. We find less than 15 cases happened in maxilla reported till date by reviewing literature [Table 1]. Huge erupting odontomas sometimes cause acute or chronic infection of paranasal sinus and maxillofacial space. The treatment involves surgical excision of entire lesion and extraction of the impacted teeth that have little value for reserving. It has a very low recurrence rate. Teeth and bone reconstruction, prevention of fistula between oral cavity and maxillary sinus, nasal cavity are the difficulty during therapy. The present study reports our experience with a rare case of erupted complex odontoma in maxilla as an example of this infrequent entity.

1. Case reviewed

A 14-year-old girl without systemic disease came to our clinic with chief complaint of a slowly enlarged asymptomatic hard lesion in posterior region of left maxilla referred by her local dentist 3 months ago. The panorama X-ray showed a huge radiopaque irregular lesion occupied part of the left maxillary sinus (Fig. 1). It had a surrounding radiolucent zone. The size of it was about 3cm * 2.5cm. There was a presence of an impacted molar overlapping with the mass. The distal part of the second premolar root is exposed because of bone resorption. The retentive deciduous tooth replaced an absent left lower second premolar. Intraoral examination revealed a partially erupted hard, rough lesion on the alveolar ridge distal to the left upper second premolar (Fig. 2). The lesion was covered by dental calculus with yellow color. The mobility of the second premolar was normal.

Computed tomography showed the lesion sized 2.6cm*2.4cm*4.2cm, protruding to maxillary sinus (Figs. 3–5). The floor of the sinus

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Table 1

A summary of erupted complex odontoma in maxilla from 1985 to 2018.

Authors	Presenting Time	Age/sex	Accomplishment	Impacted Teeth	Extraction	Size(cm)	Wound Repair
Liu,etc. [1]	1985	20/ male	None	17	Unknown	2*1*1	
C. C Ragalli etc. [18]	2000	59/ male	Enlarged	None	Adjacent tooth extracted	3.5	
Ferrer Ramírez MJ etc. [19]	2000	23/ female	Pain Swelling	27	Yes	unknown	
Ilief-Ala MA, Eisenberg E, Mathieu G [11]	2008	2/ female	bleeding	First and second deciduous molars	no	unknown	
Luis Junquera etc. [20]	2005	23/ male	None	27	Yes	3	
Gabriel Serra-Serra etc. [17]	2009	26/ male	None	None	Adjacent tooth extracted	1	
Vijeev Vasudevan etc. [7]	2009	19/ female	Pain Swelling	None	No	3*3.5	
Cyntia Helena Pereira de Carvalho etc. [10]	2011	25/ male	Pain Swelling Sinusitis	26	Yes	7	buccal fat pad flap
SantoshPatil etc. [21]	2012	25/ male	None	26	Yes	5	
K.V.Arunkumar etc. [15]	2012	22/ male	Pain Swelling Nasal obstruction Bleeding	26 27	No(pain)	3.5*4	Palatial flap
Chao etc. [9]	2013	24/ male	None	27	Yes	3*2.5	
Yumi Ohtawa etc. [12]	2013	10/ female	None	16, 17	No, 16 erupted after 12 months,17 was still impacted	1	
ShubhaRanjan Dutta etc. [16]	2016	13/ male	Pain Swelling	23	No	2	Hydroxyapatite and bio-giude membrane
Rana Fuad Swaidan etc. [8]	2017	16/ male	Cellulitis	26	Yes	2	
Nupoor Deshpande etc. [3]	2017	18/ male	Swelling	16, 17	Yes, adjacent 15 was extracted too	3*3.5	

**Fig. 1.** Orthopantomography shows a irregular mass with huge high density locates in the posterior region of left maxilla. It lies beneath the maxillary sinus floor.

existed partially. Maxillary sinus was compressed more than a half. Chronic infection can be seen in the sinus. The well-defined radiopaque mass grew around the crown of the impacted teeth. The mass was surrounded by a fine radiotransparent zone, and was separated from the normal bone by a well define corticalization line. The clinical diagnosis was erupting complex odontoma.

After preoperative evaluation excluding the surgical contraindication, we remove the entire lesion through an intraoral access under general anesthesia. Half an hour before surgery the patient was given antibiotics through vein. A mucoperiosteal flap was raised with a intrasulcular incision, from the maxillary left second premolar to tuberosity. A releasing incision was performed on the mesial of the second premolar to allow the sufficient exposure of the mass. Osteotomy was performed to facilitate its removal. The lesion was entirely removed as well as the impacted tooth which had a short root in saggital direction (Fig. 6). The apical was closed. The space between the lesion and the crown is extremely narrow. After the lesion was removed, we found there was no fistula to the maxillary sinus. In some area lacking sinus floor bone, the maxillary sinus mucosa could be seen clearly. A bio-guide membrane is used to cover the bone,



Fig. 2. Oral examination find the loss of both the first and second molars. A hard, yellow, rough mass is erupting into oral cavity.



Fig. 3. Computed tomography shows the mass protrudes to maxillary sinus. The floor of the sinus exists partially.

especially the alveolar ridge. Adjacent buccal flaps were repositioned by using a releasing incision. We used gauze to compress the defect area which could decrease bleeding in alveolar and mucosa.

The postoperative follow-up was satisfactory, with the patient developing no oral sinus fistula and showing no sign of maxillary sinusitis. The histopathological analysis showed an irregular arrangement of dental tissues such as enamel, dentine, and cementum, findings that confirmed the diagnosis of complex odontoma (Fig. 7). After three months' follow up, the patient showed no evidence of recurrence.

Now the patient suffers great loss of left posterior alveolar bone. The reconstruction of bone and teeth is difficult in adolescent period. We plan a two-stage operation to reconstruct bone and finish implanting in the late three to five years.

2. Discussion

Odontoma is a generally asymptomatic, slowly progressing tumor. It is hamartomatous malformation composed of an irregular mass of both hard and soft tissues of odontogenic origin. It constitutes 22%–67% of all odontogenic tumors [2]. Paul Broca is the first to use the

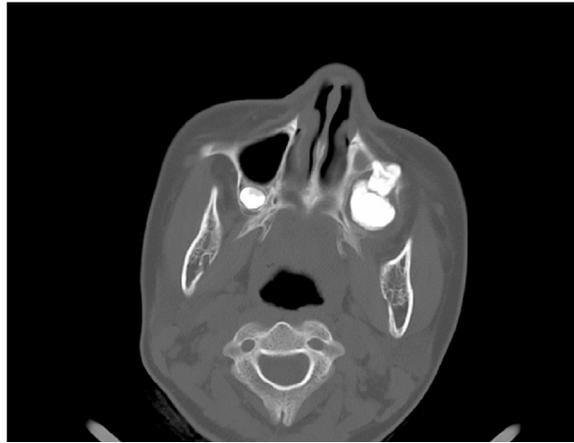


Fig. 4. CT shows the mass grows around the crown of the impacted teeth.



Fig. 5. CT shows the maxillary sinus shrinks more than a half. Chronic infection can be seen in the sinus.



Fig. 6. The surgical specimens include an impacted tooth and a irregular hard mass with a clear boundary.

term “odontoma” in 1867 [3]. Currently, the WHO has placed odontomas under the benign mixed epithelial and mesenchymal odontogenic tumors, along with ameloblastic fibro-odontoma, which is regarded as an immature precursor of complex odontoma [4].

WHO classified it based upon its gross and radiographic features into compound and complex odontomas. Compound odontomas are recognizable as orderly tooth like structures. Adjacent fibrous connective tissue consistent with dental follicle is often present. Complex odontoma appears as a disorganized amorphous mass of calcified hard tissues. Compound odontoma is twice as frequent as complex

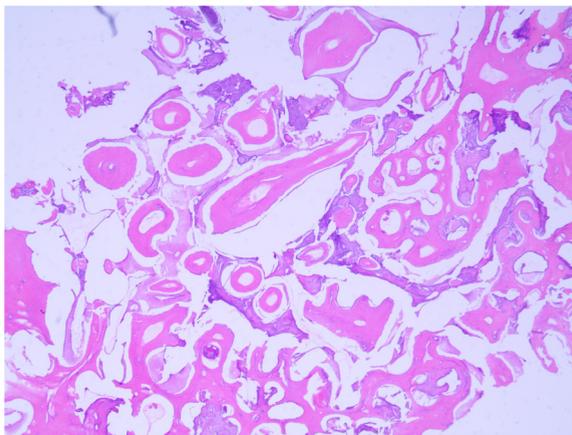


Fig. 7. The pathological diagnosis is complex odontoma. Calcified structures not resembling normal anatomical arrangement of dental tissues were detected by microscopy.

odontoma. Compound odontoma is more often seen in the anterior maxilla, while complex odontoma occurs more often in the molar region of either jaw.

It is rare for odontomas to emerge into the oral cavity, and they are first termed “erupted odontomas” by Rumel in 1980 [5]. Amado reported in a review that erupted odontoma occurred on only 1.6% of cases [6]. Comparing to the eruption of tooth, the possible mechanism of odontoma eruption appears to be the force of impacted teeth, increasing size, bone resorption, remodeling of jaws [7]. Periodontal ligament does not exist in complex odontoma. The movement does not link to the pulling of ligaments. There is inadequate growing ability without normal formation of root. The increasing size and gingivitis may induce the resorption of the overlying bone. The size of odontoma varies from 1cm to 7cm. The enlarged size is not the only factor to induce eruption. Impacted teeth exist in most of the erupted complex odontomas in maxilla. Erupting of the impacted teeth may contribute to the eruption of odontoma in their early age of normally growth and development period of teeth. But we found 3 cases without impacted teeth. The special case reported by Gabriel Serra-Serra was a young man sized about 10mm without impacted teeth and severe bone resorption [17]. In these cases the remodeling and resorption of overlying bone plays an important part in erupting. The different absorption stages of alveolar bone can be seen in those cases. It means that the possible etiology of erupting is multifactorial. The etiology of erupting is not so clear now.

The etiology of odontoma is not clear. The most accepted etiology is associated with trauma during primary dentition, infection, growth pressure, and hereditary anomalies [8].

Liu reported the first erupted complex odontoma in maxilla in 1985. There are two cases reported in our country [1,9]. We found about 30 erupting odontomas reported until now. Nearly half of these cases happened in maxilla (Table 1). The largest one presenting in posterior region of maxilla was approximately 7 cm in diameter [10]. Our case is the largest reported in our country.

Those erupting into oral cavity may cause severe oral and maxillofacial space infection. The rough surface makes dental plaque adhering and growing easily. Eruption of an odontoma through the mucosa could allow invasion of oral microorganisms into the bone due to lack of adequate adhesion between bone and odontoma because of the absence of periodontal ligament. Some patients suffered apnea because of the severe swelling of soft tissue in mouth floor and anterior portion of neck [8]. The situation is more serious than that with unerupted odontomas. Because of the complicated and numerous anatomical spaces, odontomas in maxilla may lead to severe infection more quickly than those happen in mandible. In literature, we found half of 15 cases had infection. Facial deformation can be seen more frequently in mandible without concealing of rich soft tissue in buccal fat pad region and high cheek bones.

Odontomas are mostly diagnosed during the first two decades [4]. The youngest patient suffered erupted complex odontoma is 2-year-old [11]. Our patient is a 14-year-old girl who is still in junior middle school lack of expressing ability. Her parents ignored her eruption disorder in the past 2 years.

Delayed eruption of permanent teeth is seen in 37–87% patients with odontoma [12]. The probability rises to 86% in erupted-odontomas [12].

Eruption of impacted tooth itself is hard in those with apical closure. Enlarged of the odontoma may cause the resorption of the adjacent impacted permanent tooth [13]. The reservation of the impacted teeth becomes difficult in those patients with larger odontomas, longer medical histories and older ages.

It reminds us that this disease that mostly happens in puberty should be paid special attention by parents and doctors in situations that finding unilateral deciduous teeth retention, permanent teeth deficiency, teeth shifting, no matter there is swelling and pain. Odontomas can be removed in early stage which will produce a better prognosis of the impacted teeth.

Radiographic appearance of complex odontoma depends on the stage of development and degree of mineralization [2]. The first stage is characterized by radiolucent due to lack of calcification. In the second stage, partial calcification is seen. In the third stage, lesion appears radiopaque with amorphous mass of dental hard tissue surrounded by a thin radiolucent halo. Our case is in stage three.

Although odontoma has mild biological characteristic, cyst and tumor may happen in some situation [14]. Early excision is recommended. General anesthesia is under taken in those located in the posterior region of alveolar bone with large size and close

relationship with sinus and inferior alveolar nerve. The most common approach is transoral approach. Labial and palatal mucoperiosteal flaps may be raised to allow adequate exposure. Neighboring bone should be preserved as much as possible. Le Fort I type osteotomy is used in removing those giant and highly located mass [14].

Fistula between oral and sinus should be prevented by gentle operating, tight and retention suturing with the help of palatal rotation flap, buccal mucosal flap, pedicled buccal fat pad graft or tongue flap [15]. Excessive curettaging should be avoided when sinus mucosa and inferior alveolar nerve expose into the wound. It is not rare to see fistula and dehiscence if we just simply suture the incision in those cases with maxillary sinus perforation. Fenestration in the inferior meatus is recommended. Infection and dehiscence will happen because of high tension if there is a dead space filled with blood. Bone exposure caused by wound dehiscence will lead to huge scars and pain. Infection will increase the possibility of oral and maxillary sinus perforation and nerve damage. Compressions in the area and drainage rubber tissue are useful in decreasing bleeding and pressure.

Osteoinduction active materials can be used to cover the wound which are of advantage to bone regeneration [16]. We used a bio-guide membrane to protect the bone and soft tissue. The bio-guide membrane will limit the ingrowth of Sharpey's fiber to the new bone and alleviate the contracture of the scars which will be adverse to the augmentation of alveolar.

The impacted teeth can be preserved when they locate near alveolar ridge crest. Most of them can erupt by themselves or with the help of orthodontic traction [17]. Those have abnormal shape and direction can be removed during surgery. Adjacent tooth with resorption of root could be extracted, too.

Reconstruction of dentition is hard because of bone defect. Inflammation will accelerate the absorption of bone. The adjacent teeth will be influenced by the pressure of enlarged odontoma and the infection of surrounding soft tissues. So an early excision is advised once diagnosed.

Our girl is arranged to receive bone graft surgery after the rapid growth period. Bone and teeth reconstruction will be done by free iliac bone grafting and implanting.

3. Conclusion

Odontomas are common tumors that can be easily diagnosed and treated. Erupted complex odontomas are rarely seen, it shows aggressive features, comparing with unerupted odontomas, that can lead to serious disorders in patients. Thus, there is consensus that, once diagnosed, odontomas should be surgically removed. Parents and doctors should pay more attention to their patients in puberty. The impacted teeth should be reserved if possible.

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