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ONLINE CASE REPORT

Large Complex Odontoma
A report of a rare entity

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ورم سني مركب كبير
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أودري ديكروز، سوشميني هدج، أرفاشي شيتي، أي بي شيتي

الملخص: الأورام السنية تعتبر من أمراض الورم العابي والتي تتكون من مركبات ناضجة من الميناء والعاج واللب السني وتنقسم إلى أورام مركبة أو معقدة نسبة إلى التمايز الشكلي أو التشابه مع السن الطبيعي. هذه الأورام تعتبر من أكثر الأورام الحميدة سنية المنشأ، وتشكل22% من جميع الأورام سنية المنشأ في الفك. عادة تعتبر هذه الأورام ذات طبيعة غير عدوانية وبطيئة النمو ويتم تشخيصها عادة عن طريق فحوصات الأشعة الاعتيادية في العقد الثاني من العمر. نعرض هنا تقريراً لحالة استثنائية لورم سني مركب كبير غير مؤلم. كان موقع الورم في الجهة الخلفية اليسرى من الفك السفلي وكان متعلق بفقدان الرحى الأولي والثانية من الفك السفلي. تم تأكيد التشخيص بعد الإزالة الجراحية للورم وفحص أمراض الأنسجة.

مفتاح الكلمات: ورم سني؛ عتامة شعاعية؛ ورم سني المنشأ؛ الهند؛ تقرير حالة.

ABSTRACT: Odontomas are hamartomatous lesions composed of mature enamel, dentin, and pulp, and may be compound or complex depending on the extent of morphodifferentiation or on their resemblance to normal teeth. They are the most common benign odontogenic tumours, constituting 22% of all odontogenic tumours of the jaw. They are often non-aggressive and slow growing in nature, and are usually diagnosed on routine radiological examinations in the second decade of life. We report the case of an unusually large, painless, complex odontoma, which is a rare entity. It was located in the left posterior mandible and was associated with missing 1st and 2nd left mandibular molars. The diagnosis was confirmed following surgical excision and histopathological analysis of the lesion.

Keywords: Odontoma; Radiopacity; Odontogenic tumor; Cysts; Case report; India.

The term odontoma has been used to describe any tumour of odontogenic origin. They are known as mixed odontogenic tumours because they are composed of both epithelial and ectomesenchymal components.[1] The enamel and dentin are laid down in an abnormal pattern because the organisation of the odontogenic cells fails to reach a normal state of morphodifferentiation.[2] Broca was the first to coin the term odontoma in 1867.[3] Although the aetiology of the condition is unknown, several theories have been proposed, including local trauma or infection. It has also been proposed that odontomas are inherited from a mutant gene or possible postnatal interference with the genetic control of tooth development.[4] Odontomas are often non-aggressive and slow growing in nature.

The World Health Organization (WHO) classifies odontomas into compound and complex odontomas. In a compound odontoma, all the dental tissue is in a more orderly pattern so that the lesion consists of many tooth-like structures. When this calcified dental tissue is simply an irregular mass, bearing no morphologic similarity even to rudimentary teeth, it is termed a composite complex odontoma.[2,3] Odontomas have also been classified as central or intraosseous, which present inside the bone; peripheral or extraosseous, which occur in the soft tissue covering the tooth-bearing portions of the jaws, and erupted odontomas according to clinical presentation.[5,6]

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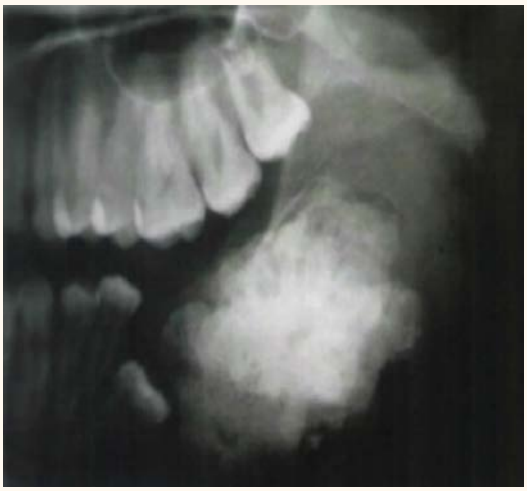

Figure 1: Orthopantomograph showing a well-defined radiodense, multilocular lesion in the lower left molar region extending from mandibular left second premolar to the angle of the mandible.

Case Report

A male patient aged 18 years reported to a private dental clinic in Bangalore, India, with a complaint of swelling in the lower left side of his face. A medical history revealed the presence of a painless swelling which had gradually increased in size over the previous two years. The patient had undergone an uneventful extraction of two teeth in that region 4 years before.

On extra oral examination, there was a solitary, ill-defined swelling measuring 4.5 x 5.5 cm, extending 2.5 cm from the midline anteriorly to the angle of the mandible posteriorly and superioinferiorly one cm above and below the border of the mandible. There were no secondary changes on the skin. An intraoral examination revealed a well-defined lesion extending from the lower left second premolar region to the retromolar area. Laterally, the lesion extended into the buccal vestibule causing vestibular obliteration. The mandibular left first and second permanent molars (36 and 37) were missing. The 1st and 2nd mandibular left premolars were displaced but the teeth were firm and non-mobile. The overlying mucosa appeared smooth and erythematous. On palpation, the swelling was non-tender and bony, hard in consistency, associated with buccal and lingual cortical plate expansion. Considering the above clinical findings, a provisional diagnosis of a residual cyst in the mandibular left molar region was made, with a differential diagnosis of ameloblastoma and periapical cemental dysplasia. The patient was advised to undergo an orthopantomograph (OPG) and cross-sectional occlusal X-ray of the mandible.

The OPG revealed a well-defined, radiodense, multilocular lesion in the lower left molar region extending from the mandibular left 2nd premolar to the angle of the mandible. The lesion was surrounded by a radiolucent border, which was ill-defined superiorly [Figure 1]. On the occlusal radiograph, the expansion of buccal and lingual cortical plates could be clearly seen. On the basis of the radiological features, a differential diagnosis of calcifying epithelial odontogenic cyst, cement-ossifying fibroma, and complex odontoma was made. Surgical excision via an intraoral approach was done under general anaesthesia at a private hospital and the specimen was sent for histopathological examination.

Grossly, the specimen showed a lobulated, yellowish-white, large, hard tissue mass, measuring 4.5 x 3 x 3 cm. An additional brownish-yellow soft tissue mass measuring 2.5 x 3 x 2.5 cm was attached to this hard tissue mass [Figure 2]. The ground section of the tissue showed the presence of enamel and cemental globules with irregularly arranged dentinal tubules around a pulp-like tissue [Figure 3]. On histological examination, decalcified sections showed the presence of eosinophilic masses of haphazardly arranged dentine and pulp-like tissues [Figure 4]. An area of fibrous connective tissue capsule was also noted. Irregular empty spaces representing the enamel space lost due to decalcification were also seen. A histopathological examination of the soft tissue mass that was attached to the hard tissue showed the presence of connective tissue, comprised mainly of acute and chronic inflammatory cells, and numerous dilated

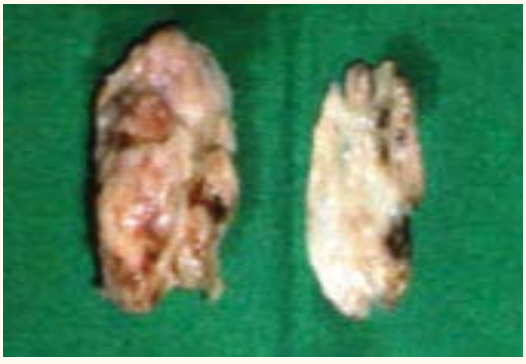

Figure 2: Gross specimen showing a hard mass (left) along with the soft tissue (right) attached to it.

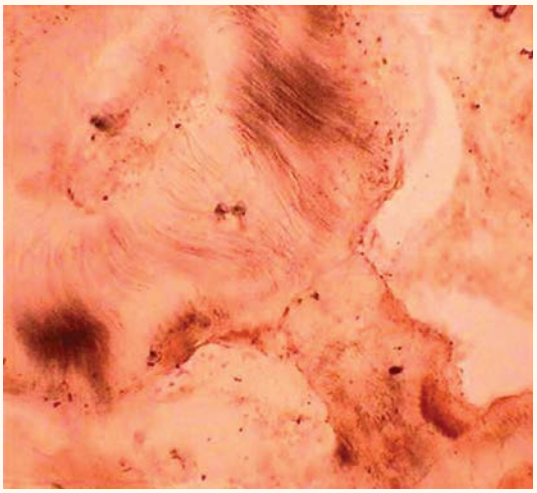

Figure 3: Ground section showing presence of enamel and cemental globules with irregularly arranged dentinal tubules around a pulp-like tissue (x 40 magnification).

vascular spaces with red blood cells interspersed between mature collagen fibres. Some odontogenic islands with cohesive cells and hyperchromatic cells were also seen. The histopathological diagnosis was of a complex odontoma.

Discussion

Odontomas are hamartomatous lesions or malformations rather than true neoplasms. They are the most common benign odontogenic tumours, constituting 22% of all odontogenic tumours of the jaw.[7,8] The occurrence of complex odontomas is rare, with a prevalence of 5–30%.[9] Most of the complex odontomas reported in the literature usually measure 1–2 cm in diameter. However, a large complex odontoma such as the one in the present case, measuring 4.5 x 5.5 cm, is very rare. Compound odontomas show a predilection for the anterior maxilla, while complex odontomas are typically found in the posterior mandibular region as seen in the present case. Complex odontomas are frequently found in the right side of the jaw.[10] However, in this case, in contrast to the usual reported findings, the complex odontoma was seen on the left side of the jaw. Complex odontoma are less common compared to the compound variety in a ratio of 1:2. There is no gender predilection and odontomas can occur at any age, but most are found in the second decade of life, as in the present case.[9]

The aetiology of complex odontomas is unknown. Several theories have been proposed, including local trauma, infection, family history, and genetic mutation. It has also been suggested that odontomas are inherited from a mutant gene or interference, possibly postnatally, with the genetic control of tooth development.[11]

Odontomas are commonly asymptomatic and are usually detected on routine radiography. A clinical indication of odontomas may include retention of deciduous teeth, non-eruption of permanent teeth, pain, expansion of the cortical bone, and tooth displacement. Other symptoms that may be present include numbness in the lower lip, frontal headaches, and swelling in the affected areas.[4] Pain is a rare symptom and is usually caused by a secondary infection due to the invasion of oral microorganisms between the bone and odontoma. The infection is likely to occur due to the absence of the periodontal ligament and lack of adequate adhesion between them. In the present case, the patient presented with swelling and expansion of the bucco-lingual cortical plate.

The radiological appearance of complex odontomas depends on their stage of development and degree of mineralisation. The first stage is characterised by radiolucency due to the lack of dental tissue calcification, followed by an intermediate stage characterised by partial calcification of odontogenic tissue. This stage is characterised by radiolucent-radiopaque images. In the third stage, the lesion usually appears radiopaque with amorphous masses of dental hard tissue surrounded by a thin radiolucent zone

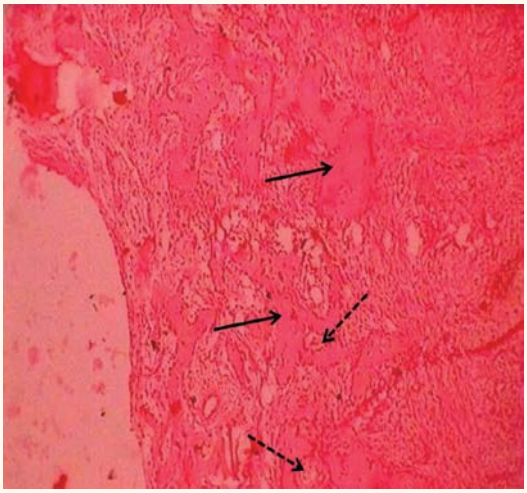

Figure 4: Decalcified hematoxylin & eosin stained section showing irregularly-arranged dental tissues, principally dentine (solid arrows) and pulp (dash arrows) (x 40 magnification).

corresponding histologically to the connective capsule.[12]

Since the present case had a mixed radiodense radiolucent lesion, we considered the present lesion to be not completely mature—that is, at the intermediate stage. Histopathologically, complex odontomas are often spherical in shape and consist primarily of a disordered mixture of odontogenic tissues. Cementum or cementum-like substances are often admixed with dentinoid structures. Small spaces with pulp tissue, enamel matrix, and epithelial remnants may be observed within the calcified mineralised masses of dentin of different qualities. A thin fibrous capsule or, occasionally, a cyst wall is seen surrounding the lesion.[1] Similar histological findings were noted in the present case.

The present case should be differentiated from periapical cemental dysplasia. Periapical cemental dysplasia occurs at the periapical region of the mandibular anteriors, whereas odontomas are commonly found in the posterior mandible occlusal to the tooth, although some may be located periapically. Also, the presence of a radiolucent border is not a constant feature in periapical cemental dysplasia and, when present, it is surrounded by sclerotic margins, whereas a radiolucent rim is a constant finding with odontomas. Periapical cemental dysplasia is uniformly radiopaque whereas an odontoma has mixed radiopaque-radiolucent features.

Mixed radiolucencies can also occur in adenomatoid odontogenic tumours, calcifying epithelial odontogenic tumours, or odontoameloblastomas. The presence of disorderly-arranged, well-formed, odontogenic tissues on histopathological examination will confirm the diagnosis of complex odontoma.

The treatment of complex odontomas varies depending on the clinical situation. The treatment options comprise surgical extraction, fenestration, and posterior orthodontic traction, or simple observation with periodic clinical and radiological controls.[13] Most often, the treatment of choice is surgical excision of the odontoma followed by histological analysis.

Conclusion

The authors present a case report of a complex odontoma that was painless, unusually large, and associated with missing 1st and 2nd left mandibular molars. Although rare in occurrence, it is important to diagnose complex odontomas that form in association with missing teeth. The use of panoramic radiography will aid in the early detection of such dental lesions, followed by surgical excision and histopathological analysis, will prevent complications. The prognosis of these tumours is very favourable, with a minimal tendency towards relapse.

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