CASE REPORT

Bilateral coronoid hypoplasia and complex odontoma: a rare concurrence of developmental pathology and odontogenic tumour of the mandible

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SUMMARY

We present a rare case of concurrent bilateral coronoid hypoplasia and complex odontoma in the mandible, with replacement of missing posterior teeth in both sides of the lower jaw. A 20-year-old woman was diagnosed with bilateral occurrence of coronoid hypoplasia and unerupted complex odontoma after radiographic and histopathological examination. The patient was surgically treated with complete removal of the unerupted complex odontoma and prosthetic replacement of the missing teeth.

BACKGROUND

Coronoid hypoplasia is a defective or underdeveloped coronoid process. Coronoid hypoplasia is considered to be extremely rare and, so far, only two cases have been identified in the previously published literature.¹ The first report was associated with Melnick-Needles syndrome and the second was an isolated case of coronoid hypoplasia.^{1 2} Thus it can be suggested that coronoid hypoplasia can occur in isolation or in conjunction with other pathological disorders. The two previously published cases were identified in women between 18 and 30 years of age.² Coronoid hypoplasia is categorised to be a developmental abnormality of the oral and maxillofacial region. Based on embryology, it has been suggested that the posterior muscle group in the temporal region helps in the development of a coronoid process.³ The mandible and temporal bone develop based on temporalis muscle activity before and after birth. In contrast, coronoid hypoplasia may also be accompanied by unchanged temporalis muscle activity and therefore the condition may be considered congenital in origin.⁴

Odontomas are the most common odontogenic tumours and considered to be a hamartomatous condition rather than true neoplasms.⁵ Paul Broca, in 1867, coined the term odontomas to describe all odontogenic tumours. However, today, the term is restricted to the odontogenic tumour, and originates from odontogenic epithelium with ectomesenchyme and contains components of dental hard and soft tissues such as enamel, dentin and pulp. The prevalence of these odontomas was reported as 22–67% in the general population, with a higher predilection in the maxillary bone in comparison with the mandible.⁶ They have no sex predilection, and are usually diagnosed in the first and second decade of life, and hence the prevalence rate is

noted to be higher in child and adolescent populations.⁷ The precise aetiology of odontomas is still not clear, however, trauma during deciduous dentition, inflammatory and infectious diseases in oral cavity, hereditary anomalies such as Gardner's syndrome and Hermann's syndrome, and genetic perturbations during odontogenesis, have been postulated.⁸ The WHO, in 2005, classified odontomas into two types: compound odontomas, usually unilocular lesions associated with multiple radiopaque tooth-like structures, termed as denticles; and complex odontomas, usually demonstrating an irregular mass of dental hard and soft tissues.⁹ The occurrence of compound odontomas is twice as common as that of complex odontomas. Odontomas are usually asymptomatic and are often discovered during routine radiographic investigation. However, a large odontoma may cause bony expansion. Compound odontomas are most frequently seen in anterior maxillae. In contrast, complex odontomas are most frequently seen in the posterior/molar region of either jaw.¹⁰

To the best of our knowledge, this is the third documented case of bilateral coronoid hypoplasia in the literature. In this paper, we present a case report of concurrent bilateral coronoid hypoplasia and complex odontoma. The diagnosis was made on the basis of radiographic and histopathological features.

CASE PRESENTATION

A 20-year-old woman of Indian origin required replacement of missing posterior teeth in both jaws. Clinical examination of the intraoral cavity revealed the absence of 4, 8, 17, 19, 20, 22, 29, 32 (Universal numbering system) (figure 1A, B). The patient had no history of extraction, thus suggesting these to be clinically missing teeth. She had no signs of occlusal derangement and no history of trismus; her mouth opening appeared to be normal (approximately 42 mm).

INVESTIGATIONS

Panoramic radiographs revealed the bilateral presence of large radiopaque tooth-like structures posterior to the mandibular second molars, with slight encapsulation of radiolucent areas. The radiopaque areas were irregular in shape, and demonstrated abnormal distribution of enamel, dentin and pulpal structure within the impacted teeth. Bilateral coronoid hypoplasia was also evident on panoramic

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Figure 1 (A) Partially erupted odontoma in posterior molar area (right). (B) Left posterior molar area showing clinically missing third molars.

radiographic examination. Panoramic radiography also revealed impacted maxillary right second molar (4), mandibular left canine (22) and left and right mandibular third molars (17 and 32) (figure 2). A postoperative radiograph showed the socket area in the posterior molar region (figure 3).

Based on the radiographic investigation, the patient was provisionally diagnosed as having partially edentulous maxillary and mandibular arches, as well as bilateral coronoid hypoplasia and mandibular complex odontoma(s). The patient was advised extraction of 4, 17, 22 and 32 due to these being impacted teeth, and because of the presence of bilateral complex odontoma.

Microscopic examination of the H&E-stained decalcified sections of the teeth showed a disorganised mass of dentin with pulpal tissue. The dentin was transverse with a longitudinal section of dentinal tubules with focal areas of fused dentinal tubules having a lack of structural architecture. The pulpal tissue showed diffuse bundles of collagen fibres interspersed with numerous endothelial lined blood vessels. The histopathological evidence confirmed the radiological diagnosis of the case (figure 4).

DIFFERENTIAL DIAGNOSIS

The initial diagnosis was multiple unerupted teeth, namely, 4, 8, 17, 19, 20, 22, 29 and 32. There were no local or general sign(s) or symptom(s) to make a possible syndromic association.

TREATMENT

Following the radiographic investigation, extraction of the maxillary second premolar (4), mandibular left canine (22) and impacted odontomas bilaterally at the mandibular jaw in the third molar position (17, 22) was recommended (figure 4). Prosthodontic rehabilitation was advised in 2–3 months after surgical procedures.

Figure 2 Preoperative panoramic radiograph showing bilateral complex odontoma and coronoid hypoplasia.

Figure 3 Postoperative panoramic radiograph showing extraction socket area in the posterior molar region (bilaterally).

OUTCOME AND FOLLOW-UP

A 1-year postoperative visit showed a good prognosis with no symptoms of recurrence of complex odontoma.

DISCUSSION

Bilateral coronoid hypoplasia is a rare entity with an unclear aetiopathogenesis. It is usually considered as a developmental abnormality, and has been suggested to be due to activity of the temporalis muscle in prenatal and early postnatal life. However, it has also been suggested that coronoid hypoplasia may occur in individuals with unchanged muscle activity.^{1 3} The present case was diagnosed during routine radiographic investigation. Gorlin, in 1978, reported coronoid hypoplasia in a patient with Melnick-Needles syndrome. The radiographic examination of Melnick-Needles syndrome also showed hypoplasia of mandibular angle, cyst-like alteration in the molar ramus area and absence of mandibular second and third molar teeth.² Melnick-Needles syndrome is considered as osteodysplasia, which is usually characterised as generalised bony alterations. The present case could not be concluded as Melnick-Needles syndrome because of isolated hypoplasia of a coronoid process.

Odontomas are considered to be the commonest odontogenic tumours, and most authors believe them to be hamartomas rather than true benign neoplasms.¹¹ Complex odontomas are more prevalent than compound odontomas.¹² The aetiology of odontomas is due to gene mutations or interference, or possibly postnatal genetic control of odontogenesis. Persistent dental

Figure 4 Histophotomicrograph (x20) showing the transverse and longitudinal section of dentinal tubules, with lack of structural architecture; pulpal area showing diffuse collagen fibre bundles. The inset showing dentinal tubules.

lamina is an important factor in the aetiology of complex odontomas.^{10 13}

In most cases, odontomas are asymptomatic in presentation; however, certain clinical predictors may be identified in a few cases. Clinical predictors are retention of deciduous teeth, failure to erupt of permanent teeth, asymptomatic enlargement of cortical bone and adjacent tooth displacement. Pain and swelling may be seen in cases of erupted odontomas.⁸ Eruption of odontomas in the oral cavity is controversial. Eruption failure is due to lack of eruptive forces in these malformed teeth, resorption of the alveolar ridge and resulting exposure of the odontoma, and sequestration of overlying bone.¹¹ In the present case, the odontoma on the right showed partial eruption, while the odontoma on the left was completely located in the jaw bone. In the present case, the lesion was asymptomatic and diagnosed on routine radiological examination.

Odontomas are the commonest odontogenic tumours, representing 70% of all odontogenic tumours. However, multiple odontomas are less common, however, cases have been reported.^{14 15} Recently, 12 cases of multiple odontoma were reported, including isolated cases as well as those involving two or four quadrants of the jaws.¹⁶ A concurrent case of multiple complex composite odontoma and dentigerous cyst has been reported.^{17 18} Another case has been reported on multiple compound odontomas involving the four quadrants of the jaw.¹⁹ A bilateral complex odontoma in the mandibular jaw in a patient with Pierre Robin syndrome has been previously reported.²⁰ However, the present case did not show any syndromic features during clinical or radiological examination. The present case is unique due to the concurrent occurrence of bilateral complex odontoma in the mandibular jaw and coronoid hypoplasia; so far, no cases have been reported with such an association.

The developmental stage of odontomas can be determined based on the evaluation of degree of calcification of the lesion by radiographic investigation.²¹ The initial phase is recognised by the radiolucent appearance of the odontoma, which is due to the absence of dental tissue calcification.^{22 23} The intermediate phase is recognised by its partial calcification. The terminal phase is recognised by a classical radiopaque appearance and exhibits predominant calcification, with a surrounding radiolucent halo around the radiopaque area.² The radiological interpretation of the present case showed a radiopaque mass surrounded by a radiolucent halo, and was suggestive of a terminal phase of odontoma development.

The present case was treated with surgical removal of the lesions along with the fibrous capsules. The histopathological examination of the biopsied tissue was helpful in achieving the diagnosis of complex odontoma.

In conclusion, coronoid hypoplasia is a rare developmental abnormality of the oral and maxillofacial region. The lesion should be considered in the list of developmental pathology affecting the jaw bone. Complex odontomas are common odontogenic tumours, but bilateral occurrence of this lesion is considered an unusual occurrence. The diagnosis of bilateral coronoid hypoplasia and complex odontoma was achieved by routine radiological investigation.

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Learning points

- ▶ Odontomas and condylar hypoplasia patients are usually asymptomatic.
- ▶ Consider coronoid hypoplasia in developmental pathology affecting the jaw bone.
- ▶ Careful observation of panoramic radiographs is important in the diagnosis of coronoid hypoplasia.
- ▶ Always consider radiographic investigation when attending to patients with missing teeth, especially in those with multiple missing teeth; further recommendation for panoramic radiographs should be included for such patients (multiple missing teeth).
- ▶ Complete surgical excision of odontomas is usually accompanied with a better prognosis.

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