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CASE REPORT

Multiple osseous dysplasia arising from impacted teeth: report of a case associated with odontogenic lesions

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We report a case of osseous dysplasia (OD) showing extremely rare clinical features. A 37-year-old Japanese woman was referred to our clinic complaining of a left alveolar bony swelling at an impacted canine. Radiographic examinations revealed a mixed radiopaque lesion involving the impacted left canine and also revealed an impacted left wisdom tooth with a cystic change and a clear radiopaque lesion suspected to be odontoma. All the extracted upper teeth and odontoma showed hypercementosis and the canine was fully involved in it. Histopathologically, they showed the same features and were diagnosed as OD. The findings of multiple OD from incompletely erupted teeth and odontoma in our case may well show that OD can arise from any tooth with periodontal ligaments and that focal OD can show expansive growth like the ossifying fibroma.
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Keywords: cemento-osseous dysplasia; dentigerous cyst; impacted tooth; odontoma; osseous dysplasia; ossifying fibroma

A 37-year-old Japanese female was referred to our department in October 2004 with swelling of the alveolar bone at the upper left canine. She had noticed the painless bony swelling at the upper left canine area several years before and she was referred to a dental clinic. An initial examination revealed the remains of an erupted deciduous canine and a periapically located mixed radiopaque lesion. The deciduous canine was removed, but a continuous discharge of pus was observed after this treatment. She was recommended to undergo treatments at our clinic 1 week after the extraction. The interview for the patient did not reveal any familial history of systemic lesions or related jaw lesions.

Clinical examination revealed a painless rounded bony swelling at the upper alveolar bone of the canine area and a pus discharge from the extracted cavity of the deciduous canine. The upper left second molar had partly erupted, but any abnormal findings were only observed in the alveolar bone. Radiographic examinations revealed the presence of an impacted upper left impacted canine surrounded by an ill-defined mixed radiopaque lesion (Fig. 1a). This examination also revealed the presence of deeply located upper and lower left impacted wisdom teeth. A radiolucent lesion was present around the upper wisdom tooth crown and a radiopaque mass was located in it. The lower wisdom tooth also had a radiolucent coronal lesion. Computerized tomography (CT) examination revealed an alveolar bone expansion near the mixed radiopaque mass, which fully enveloped the canine (Fig. 1b). This mass was surrounded by an irregular radiolucent line. The migrated upper left wisdom tooth was partly present in the maxillary sinus and the tooth crown was enveloped by a cystic lesion. The clear radiopaque mass was located within this cystic cavity. The dental root of the upper left second molar had an irregular margin compared with other erupted teeth (Fig. 1c).

A biopsy was performed from the swelling mass of alveolar bone at the impacted canine area. Histologically, the biopsy specimen was composed of cellular fibrous stroma admixed with bony or cementum-like materials. From these findings, this lesion was considered to be a fibro-osseous lesion: either an ossifying fibroma or an osseous dysplasia (OD).

Under a diagnosis of fibro-osseous lesion, the impacted canine and multiple odontogenic lesions, including the odontoma and the dentigerous cyst in the upper wisdom teeth, were excised. According to the patient's demands, both incompletely erupted teeth, the upper left second molar and lower left wisdom tooth, were also removed. During the operation, partial destruction of the alveolar cortex by the tumor was observed. We

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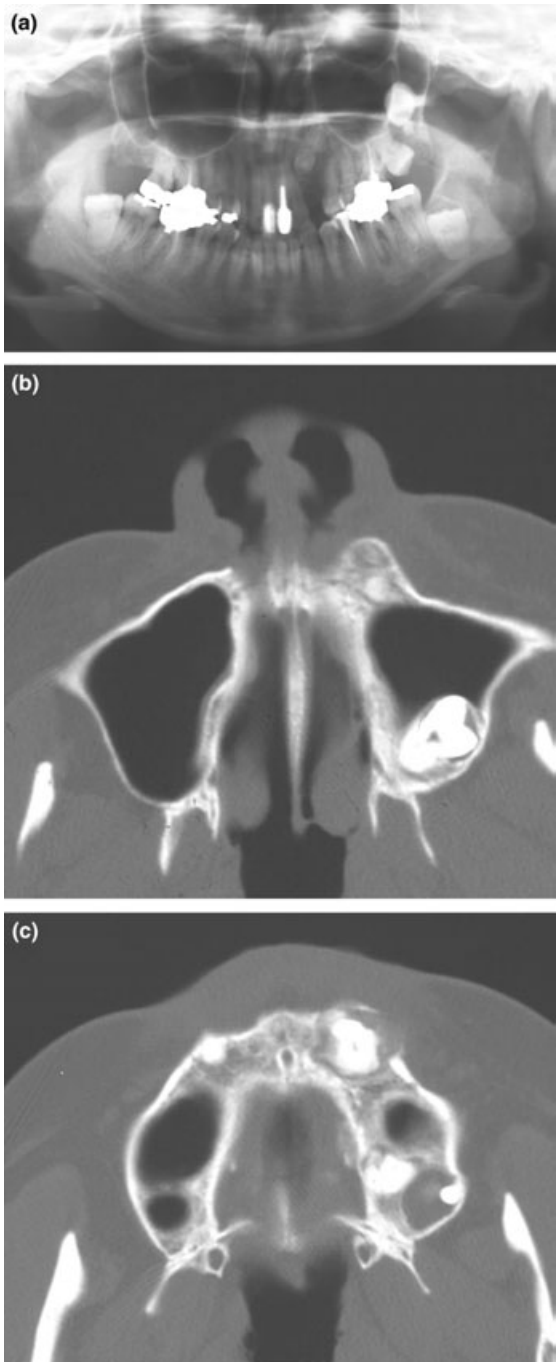

Figure 1 Radiograph and CT scanned image. (a) Panoramic radiograph. The panoramic radiograph shows the presence of the ill-defined mixed radiopaque lesion, which fully envelops the impacted upper left canine and the divergence of the dental root of the left lateral incisor and the first premolar. The radiograph also shows the impacted upper left wisdom tooth that has migrated into the maxillary sinus and the radiolucent lesion including the tooth-like radiopaque lesion. The upper left second molar erupted incompletely and the impacted lower left wisdom tooth with the enlarged dental follicle are seen, but the clear periapical changes are not evident in this radiograph. (b) CT of the migrated upper wisdom tooth. The upper left wisdom tooth migrated into the maxillary sinus and the disappearance of the periodontal space is also seen. The tooth crown was enveloped by the cyst. The alveolar bone expansion near the mixed radiopaque lesion around the root apex of the upper left canine caused the protrusion of the left alar base. (c) CT of the odontoma and dental root of the second molar. This slice shows the ill-defined, rounded mixed radiopaque lesions enveloping the canine and causing with bony expansion of the alveolar bone. The dentigerous cyst is a clear radiopaque mass suspected to be odontoma. The dental root of the upper left second molar shows an irregular surface and the periodontal space is not observed.

extirpated the tumor by reducing the surrounding bone and by separating the tumor itself using surgical bars. The tumor had partly adhered with the surrounding bone and no clear enveloping soft tissue was observed.

The impacted wisdom tooth with the cystic lesion and a tooth-like mass were removed by reduction of the surrounding alveolar bone and the incompletely erupted second molar was also removed. The second molar and wisdom tooth were not easily removed because of adhesion to the alveolar bone. After removal of the lesions in the upper jaw, the lower left wisdom tooth and the enlarged dental follicle were removed. The removal of this tooth was also somewhat difficult and required tooth separation because of adhesion of the dental root (Fig. 2). The excised specimens of the cystic lesions from the lower and upper wisdom teeth and the tumoral lesion around the tooth crown of the canine were examined histologically. Also, the odontoma-like lesion and all the removed teeth except the lower wisdom tooth were also examined microscopically to analyze histological changes of the dental roots.

Histological examination revealed that the lesions around the upper left canine, the root of the second upper molar and the root of the upper wisdom tooth were composed of the same components as the biopsy specimen (Fig. 3a). The roots of these teeth showed hypercementosis and the continuity between the fibro-osseous lesion and the root surface was evident (Fig. 3b). The cystic lesion of the upper wisdom tooth was diagnosed as a follicular cyst (Fig. 3c). The lesion adhered to the cyst wall is represented by an irregular dental structure surrounded by the fibro-osseous lesion (Fig. 3d). The cystic lesion of the lower wisdom tooth histologically looked like a dental follicle. Though they showed rather extensive nature, we made a final diagnosis as multiple focal OD arising from incompletely erupted teeth because of its unilateral occurrence.

The post-operative course was uneventful and a radiographic examination 6 months after the operation did not reveal any new developed radiopaque lesions in the jaws, including the excised area.

Comments

Osseous dyplasia is the most common fibro-osseous lesion in jaws. This lesion is defined as idiopathic processes located in the periapical region of the tooth-bearing jaw area, histologically characterized by a replacement of normal bone by fibrous tissue and metaplastic bone (1). This entity 'osseous dysplasia' has been called as 'cemento-osseous dysplasia (COD)' (2), because of their histological finding with woven lamellar bone and masses of cementum-like materials.

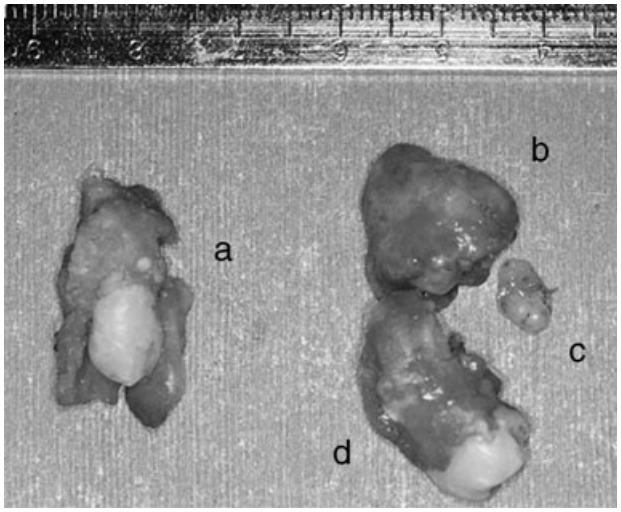

Figure 2 Extirpated specimen. (a) The rounded bony hard lesion and upper left canine. The lesions originated from dental root and enveloped the tooth crown of the canine. The lesion does not have clear soft-tissue capsulations. To show the relationship of the tooth crown, the tumor was partly removed. (b) The dentigerous cyst and the upper wisdom tooth. The dental root of this tooth shows a rather irregular surface but does not show clear morphological changes. (c) Odontoma. (d) The upper left second molar. The bony hard lesion adhered to the dental root.

However, in the new histological classification by WHO in 2005 (1), almost of the term of 'cemento-' or 'cemental' used for the names of categories included in this entity are deleted and now this entity is also called simply as OD. In this new classification (3), this change is also performed in other entity of bone-related lesion 'ossifying fibroma', which has been called as cemento-ossifying fibroma in the previous classification (2). These steps may be taken to finalize the discussion, whether cementum is produced in these lesions or not.

In the past, quite numerous terms had been applied for this entity (4), till the permeation of the features of COD described in the WHO classification in 1992 (2). This classification of COD was helpful for pooling these numerous different terms under one heading and had only three categories [periapical cemental dysplasia, florid COD (FICOD) and others]; however, there are no characteristic histological appearances among them and varied clinical features, including equivocal radiographic appearances, also cause confusion in their diagnosis. In other word, these terms were not sufficient to comprehend the varied clinical features of COD with the current definitions. Some reports recommended using the term 'Focal COD' for localized, self-limited COD instead of periapical cemental dysplasia (5–7) and this term could include most of the cemento-osseous lesions in jaws. On the other hand, FICOD has been used for the dysplastic lesion, which shows rather severe sclerotic changes affecting a wide area of the jaws, such as multiquadrant occurrences. In the 1992 WHO classification (2), FICOD was used synonymously with gigantiform cementoma. However, the term 'FlCOD' included so many cases showing different features that it may now become difficult to imagine a typical case of FlCOD. Another interpretation of FlCOD that denoted an extensive or multiple involvement of the jaw by focal CODs (5) also caused confusion. Focal CODs often appear in multiple locations and often in 'multiquadrants' without showing an aggressive nature. Therefore it was difficult to use the term 'multiquadrants' only for distinguishing FlCOD from multiple focal COD and these classifications were likely to be used according to personal preferences.

In new classification by WHO (1), this entity has four categories according to clinical features; periapical OD, focal OD, florid OD and familial gigantiform cementoma. Former two types are limited lesion and periapical OD occurs in the anterior mandible and focal OD in posterior jaw quadrant. Latter two types are more extensive and occur bilaterally in the mandible or even involving all four jaw quadrants. The florid OD mainly occurred in middle-aged woman and the familial gigantiform cementoma occurs in young age with considerable jaw expansion. This classification must contribute to solve the previous confused definitions of each type of ODs, but it may be still impossible for these four categories to comprehend the various clinical features of ODs. Nevertheless, some of these ODs seem to represent the same pathological process (8) and focal OD must include some cases that will develop into florid OD. Therefore, each term may only show one stage of the same disease, so OD should always be treated as a lesion to reflect its progressive nature.

Our present case has some unique clinical findings that have never been reported in the past:

1 Multiple occurrences of cemental hyperplasia invading to the root surface of incompletely erupted teeth, including odontoma in a single quadrant of the jaw.
2 Tumoral growth in the lesion arising from the impacted canine, with remarkable expansion of the maxillary bone, involvement of the crown of the teeth and divergence of adjacent teeth.
3 OD association of dentigerous cyst.

The close proximity to the periodontal ligament and some histopathological similarities to this tissue have led to a presumption that OD and ossifying fibroma are of periodontal origin (1, 3), but there is no scientific evidence to support this hypothesis (5). Kawai et al. (9), who analyzed the radiographic appearance of OD, showed that most lesions have normal periodontal space between the dental root and lesion, and they developed a hypothesis of pathogenesis that the cases showing active hypercementosis may have originated from periodontal ligament and other cases having periodontal space may have originated from medullary bone.

The most considerable reason for the continuing uncertainty of the origin may be the self-limited and asymptomatic nature of this lesion, which does not require aggressive treatments such as the removal of the tooth, which would provide a detailed histological relationship between the periodontal ligament and OD from the pre-surgical examination. In fact there are no reports showing histological relationships between

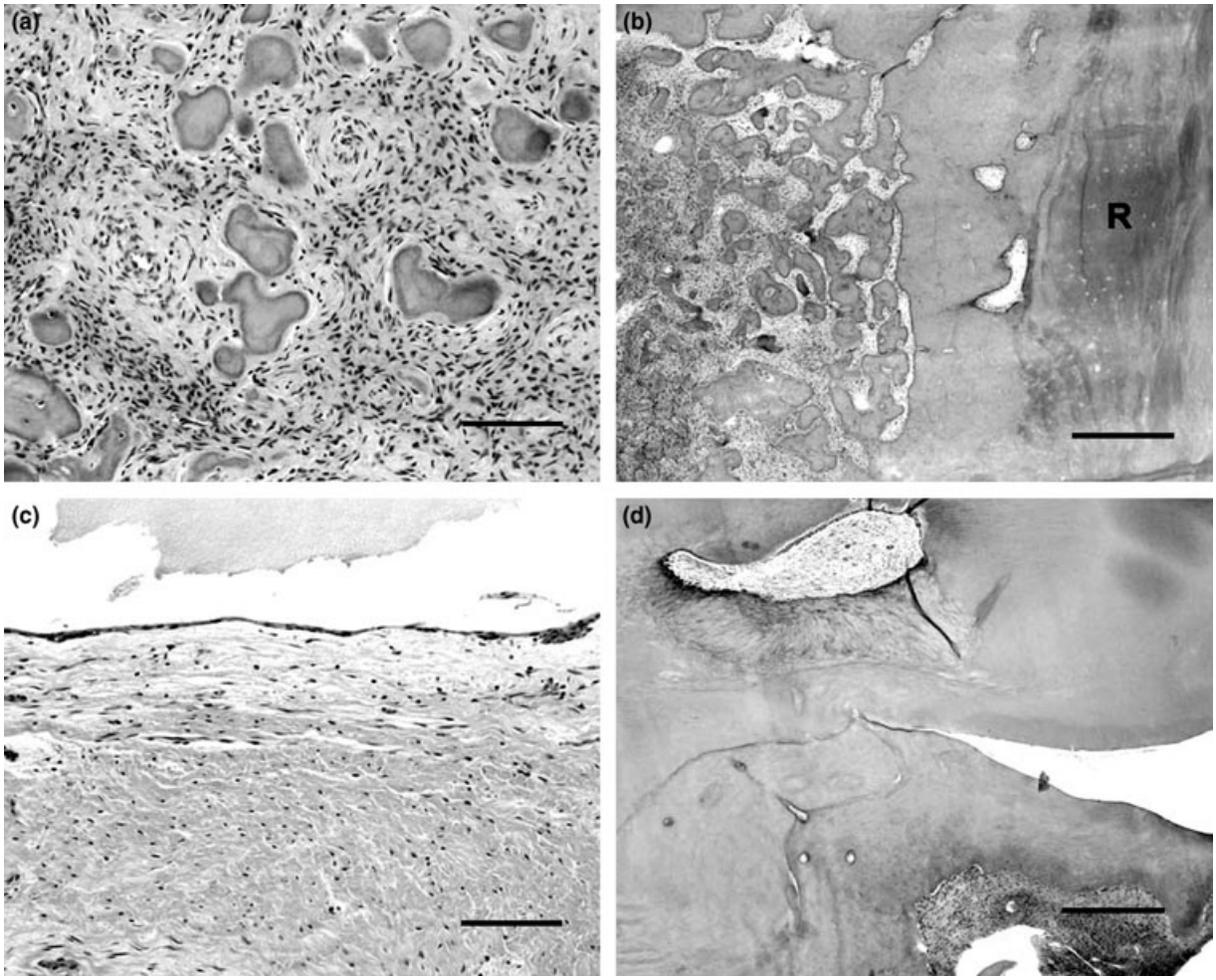

Figure 3 Histopathological findings. (a) The lesion around the tooth is composed of cellular fibrous stroma admixed with bony or cementum-like materials. (b) The root of the tooth shows hypercementosis and is connected to the surrounding hard tissues. (c) The cystic lesion of the upper wisdom tooth is composed of fibrous connective tissue lined by odontogenic epithelium. (d) The irregular dental structure that adhered to the cyst wall is surrounded by cementum-like material and cellular fibrous tissue. (H&E; a,c; bar 50 μm; b,d; bar 200 μm; R, root surface of the tooth).

multiple OD and vital teeth. Fortunately, we had a chance to observe these relationships in several materials, which displayed active cemental hyperplasia invading toward the dental root and a lesion showing gradual changes to OD. These histological features support the hypothesis that OD originates from periodontal ligaments and may indicate the possibility of the contribution of some systemic factors and also suggests that it is not just a reactive lesion.

The lesion arising from the impacted canine root in our present case showed extremely different clinical features from other lesions, i.e. full envelopment of the tooth, expansion of the alveolar bone and divergence of adjacent teeth. Because there were no clear radiographic findings showing the presence of other ODs in the jaws, this lesion was thought to be a solitary lesion with a neoplastic nature and these clinical features permitted us to diagnose it as an ossifying fibroma.

An osseous lesion being associated with an impacted tooth is extremely rare and only one case of ossifying fibroma arising from an impacted tooth has been reported to our knowledge (10). In that case, the lesion had enveloped the migrated, impacted wisdom tooth, similar to our case. The difficulty of histologically differentiating between ossifying fibroma and OD has been noted (1, 5, 6). If the canine lesion in our present case had appeared alone, ossifying fibroma might have been an adequate diagnosis according to the clinical features, such as its neoplastic natures. However, because the histology of the canine lesion matched that of the other excised materials in our case and because there are no reported cases of ossifying fibroma associated with other ODs in the existing literature, we could not diagnose this lesion as an ossifying fibroma and finally concluded that it was only a different clinical manifestation of other lesions.

The production of solitary bone cysts is a well-known phenomenon associated with florid OD. The pathogenesis of this lesion is unclear, but it is thought to be different from that of the ordinary solitary bone cyst. Recently Wakasa et al. (11) reported a case of this cyst developing during the follow-up period after surgery for OD and they suggested that the disorderly production of trabeculae in florid OD might result in the obstruction

of lymphatic drainage, including cystic degeneration. In our present case, a dentigerous cyst was observed in the migrated upper wisdom tooth and an odontoma was located beside the cyst. Teeth that have not erupted because of odontoma often show cystic changes in the dental follicle that are diagnosed histologically as dentigerous cysts (12). These phenomena are commonly seen in mixed dentition. Both odontogenic lesions in our case had occurred in this period and the dysplastic changes in the periodontal ligaments may have begun when the patient was middle-aged. These facts caused us to consider that these osseous dysplastic changes only occur in the jaws involve previous odontogenic lesion. The facts also indicate that OD can occur at any time and involves the periodontal ligaments, but it is clear that a case of OD associated with odontogenic lesions is extremely rare.

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