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Central giant cell lesion of the mandible in a 2-year old girl

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ABSTRACT

Central giant cell lesions are rare, benign, osteolytic, pseudocystic, solitary, localized lesions that are common in the skeletal structure, but less so in the maxillofacial region. Furthermore, to perform panoramic radiography and cone-beam computed tomography, it is necessary to prepare patients properly and to position their heads carefully. However, this can be difficult in pediatric patients, who may be anxious. In this report, we describe the case of a central giant cell lesion of the mandible in a 2-year-old girl that was evaluated with multidetector computed tomography. ***(Imaging Sci Dent 2017; 47: 209-13)***

KEY WORDS: Granuloma, Giant Cell; Multidetector Computed Tomography; Mandible; Child

Central giant cell lesions of the jaw are relatively uncommon lesions that cause thinning and expansion of the cortical bone.[1,2] Either jaw can be affected, but the mandible is affected more often. Although these lesions are benign, they can be clinically classified as non-aggressive or locally aggressive, and they occasionally recur after treatment with curettage or surgical excision.[3]

Histopathologically, these lesions are characterized by a granulomatous appearance, with the presence of increased amounts of multinucleated giant cells (osteoclast-like cells) and surrounding mononuclear cells.[4] In 1953, Jaffe categorized most giant cell lesions in the jaw as reactive processes, but not as neoplasms, such as giant cell tumors, and named them central giant cell reparative granulomas.[5,6] Thereafter, the word reparative was deleted because it was realized that many of these lesions are more destructive than reparative.

The recent World Health Organization classification in 2013 defines a giant cell-rich tumor with histopathologic characteristics resembling with those of a central giant cell granuloma as a giant cell lesion of the small bones.[7] The histopathologic features are fibrous tissue with hemorrhage, hemosiderin deposits, irregularly distributed giant cells, and reactive bone formation. Despite the similarity of the histopathologic findings, it is considered that this newly defined group does not include central giant cell granuloma of the jaw. Recent advances and insights into the molecular pathogenesis of giant cell lesions may clarify our understanding of these diseases in the near future.[8]

Most cases of this disease are observed in children or young adults. Waldron and Shafer[9] reported that 74% of patients were under 30 years old among their 38 cases. However, only 16% of their cases were younger than 10 years old.

For panoramic radiography and cone-beam computed tomography (CBCT), it is necessary to prepare patients properly and to position their heads carefully. However, this can be difficult in pediatric patients, who may be anxious. This report describes the case of a giant cell lesion of the small bones of the mandible in a 2-year-old child that was evaluated with multidetector computed tomography (MDCT), and discusses the usefulness of MDCT for pediatric patients.

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Case Report

A 2-year-old girl presented with the premature exfoliation of a left central deciduous incisor of the mandible. A clinical photograph showed an extracted tooth wound in the central area of the left side of the mandible (Fig. 1). A periapical radiograph showed root resorption of the left mandibular lateral deciduous incisor of the mandible (Fig. 2).

Computed tomographic imaging was performed with a 16 MDCT scanner (Aquilion TSX-101A, Toshiba Medical Systems, Otawara, Japan) using the maxillofacial protocol at our hospital: tube voltage, 120 kVp; tube current, 150 mA; field of view, 240 × 240 mm; and rotation time, 0.50 s. The protocol consisted of axial acquisition (0.50 mm) with axial, coronal, and sagittal multiplanar reformation (MPR) images.

Computed tomography using the axial soft-tissue algo-

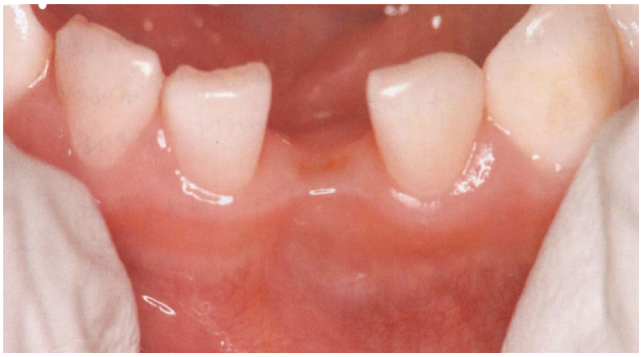

Fig. 1. A clinical intraoral photograph shows an extracted tooth wound in the central area.

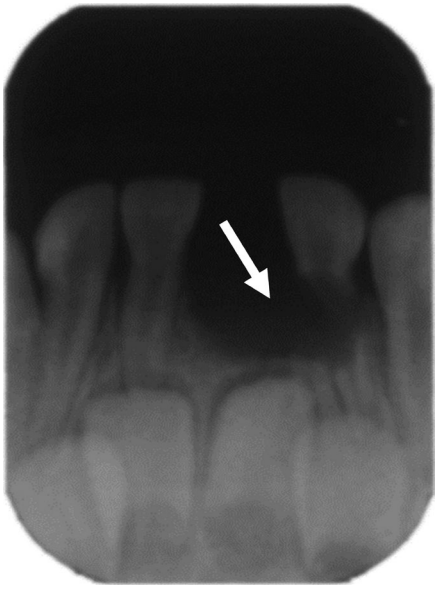

Fig. 2. A periapical radiograph shows root resorption of the left mandibular lateral deciduous incisor of the mandible.

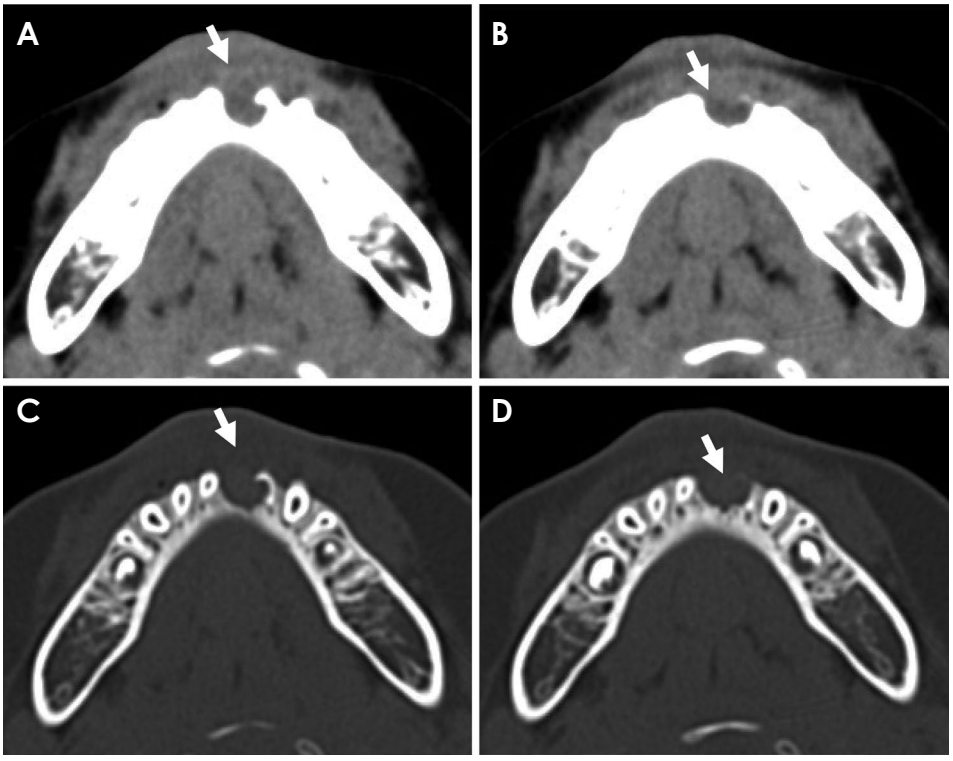

Fig. 3. Axial soft-tissue algorithm CT images (A and B) show a mass lesion along the central area (arrow). Bone-tissue algorithm CT images (C and D) show an expansile lesion with an irregular and fairly well-defined area in the central area (arrow). CT, computed tomography.

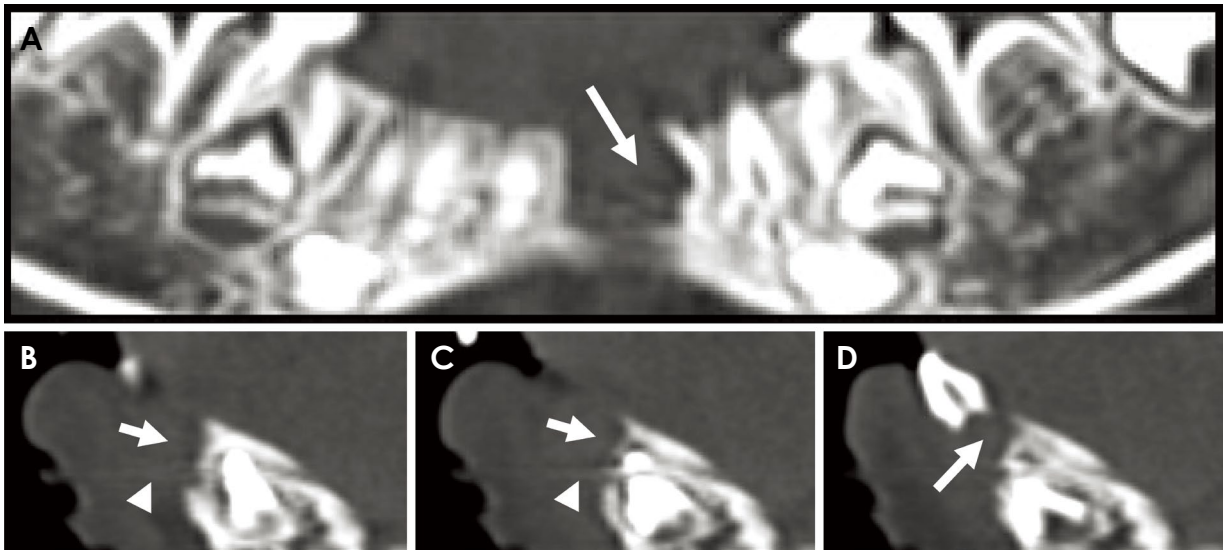

Fig. 4. Multiplanar panoramic (A) and cross-sectional (B-D) reformation images more clearly demonstrate the root resorption of the left mandibular lateral deciduous incisor of the mandible and an expansile lesion in the central area (arrow).

rithm showed a mass lesion along the central area (Fig. 3A and C). Computed tomography using the bone-tissue algorithm showed an expansile lesion with an irregular and fairly well defined area in the central part of the left side of the mandible (Fig. 3B and D).

MPR images more clearly demonstrated the root resorption of the left mandibular lateral deciduous incisor of the mandible and a well-defined unilocular lesion in the central area of the left side of the mandible (Fig. 4).

The lesion was removed via enucleation and curettage with a local osteotomy. The histopathological examination revealed a solid proliferation of oval to spindle-shaped fibroblasts with multinucleated giant cells. The fibroblasts were arranged in a whorled or fascicular pattern. Giant cells with fewer than 20 nuclei were seen around the hemorrhage (Fig. 5A and B). The multinucleated giant cells and mononuclear cells were positive for CD68 (Fig. 5C), receptor activator of nuclear factor kappa B (RANK), and cathepsin K by immunohistochemistry, but were negative for p63. The nuclear shapes of the mononuclear cells were not similar to those of the multinucleated giant cells. The cellular and nuclear atypia of these cells was mild. Therefore, the histopathological diagnosis in this case was a giant cell lesion of the small bones. At a 5-year follow-up, there was no evidence of local recurrence.

Discussion

Giant cell lesions of the bone are common in the long bones, but rare in the facial bone, which accounts for only 2%-12% of all giant cell tumors of the body.[1,2] To the best of our knowledge, few case reports have described giant cell lesions of the mandible.[1,2] Furthermore, in recent years, giant cell reparative granuloma has been classified as an osteoclastic giant cell-rich tumor (namely, giant cell lesion of the small bones), which comprises very rare tumor-like lesions consisting of fibrous tissue with hemorrhage, hemosiderin deposits, irregularly distributed giant cells, and reactive bone formation.[7] This report presented the case of a giant cell lesion of the small bones of the mandible in a 2-year-old child that was evaluated with MDCT.

In this case, the histopathological examination revealed solid proliferation of oval to spindle-shaped fibroblasts with multinucleated giant cells. The fibroblasts were arranged in a whorled or fascicular pattern. Giant cells with fewer than 20 nuclei were seen around the hemorrhage. The multinucleated giant cells and mononuclear cells were positive for CD68,[10] RANK,[11] and cathepsin K[12] by immunohistochemistry, but were negative for p63.[13-15] The nuclear shapes of the mononuclear cells were not similar to those of the multinucleated giant cells. The cellular and nuclear atypia of these cells was mild. Therefore, the histopathological diagnosis in this case was giant cell lesion of the small bones. The incidence of giant cell tumors is 2%-12% in the head and neck region; of these cases, 90% affect the posterior mandible, and they usually occur in young adults under 20 years of age.[1] Recently, Jadu et al.[16] reported the case of a 31-year-old man with a central giant cell granuloma of the mandibular condyle,

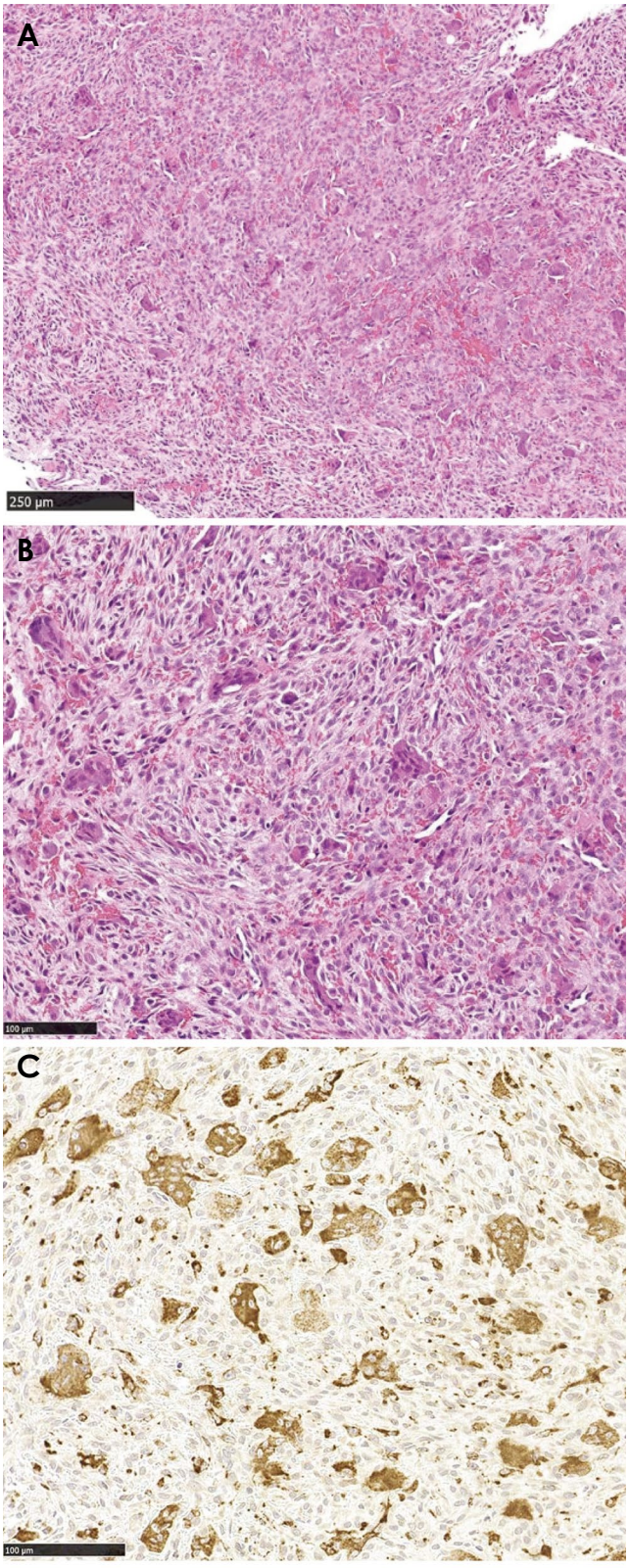

Fig. 5. A and B. The multinucleated osteoclast-like giant cells are embedded in a fibrous stroma comprising oval to spindle-shaped cells around the region of hemorrhage. C. Positive immunohistochemical staining for CD68.

and Mohan et al.[17] presented a case of a 4-year-old boy with a central giant cell granuloma in the right mandibular region. Therefore, a giant cell lesion in the central area of the mandible in a 2-year-old, as described in this report, is a very rare occurrence.

Giant cell tumors are often benign, but they can be locally aggressive and are able to spread by metastasis.[18-20] The characteristic imaging findings of this tumor still have not received sufficient consideration in the literature. In this study, periapical radiography showed root resorption of the left mandibular lateral deciduous incisor of the mandible and a radiolucency in the central area. Kulkarni et al.[1] reported a case of an extensive giant cell tumor of the mandible. Recently, MDCT with MPR and 3-dimensional images has become a standard part of oral and maxillofacial imaging, as this imaging technique is highly sensitive for oral and maxillofacial lesions.[21,22] In this study, MDCT with MPR images more clearly demonstrated the root resorption of the left mandibular lateral deciduous incisor of the mandible and an expansile lesion in the central area.

Furthermore, regarding the radiation dose administered during CT, Mah et al.[23] showed that the effective dose for imaging the maxillofacial volume with CBCT was significantly lower than that required for CT imaging methods. However, CBCT is difficult in pediatric patients, because it is necessary to prepare patients properly for CBCT and to position their heads carefully in the focal trough. We considered MDCT to be an effective tool for assessing the maxillofacial lesion in this infant patient because of its ability to acquire images rapidly. In such circumstances, MDCT may be justified for the diagnosis and treatment of the patient. In our case, imaging was performed with a 16-MDCT scanner using the maxillofacial protocol at our hospital: tube voltage, 120 kVp; tube current, 150 mA; field of view, 240 × 240 mm; and rotation time, 0.50 s. Nonetheless, we should consider lower-dose protocols for pediatric patients in the future.

In conclusion, we presented the case of a giant cell lesion of the small bones of the mandible in a 2-year-old child that was evaluated with MDCT. MDCT can be an effective tool for assessing maxillofacial lesions in infants or young children because of its ability to acquire images rapidly.

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