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Case Report

Odontogenic myxoma containing osteocement-like tissue: Report of a case with an unusual histopathological feature

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ABSTRACT

An odontogenic myxoma occurring in the mandible of a 54-year-old Japanese male is described; this contained unusual osteocement-like spherules, which are rarely reported. The radiological examination revealed a multilocular lesion with mixed radiolucent–radiopaque content. We performed a segmental mandibulectomy, and reconstructed the mandible using a titanium plate and iliac bone. Histopathologically, the tumor was composed mainly of myxomatous stroma in which a few stellate and spindle-shaped cells were suspended. A few odontogenic epithelial nests were also observed. In addition, there were osteocement-like spherules, showing concentric lamination. The presence of these osteocement-like structures has only been reported in three previous cases.

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1. Introduction

Odontogenic myxoma of the jaw was first described by Thoma and Goldman in 1947 [1]. It is an intraosseous neoplasm characterized by stellate and spindle-shaped cells embedded in abundant myxoid or mucoid extracellular matrix [2]. Simon et al. [3] reported the incidence of this rare odontogenic tumor at approximately 0.07 new cases per million people per year. Two-thirds of odontogenic myxomas are located in the mandible, and are most common in the molar region [4]. Here, we present an unusual case of odontogenic myxoma containing osteocement-like tissue.

2. Case report

A 54-year-old Japanese male presented with a chief complaint of a loose left mandibular second molar, which was noted 3 months before admission. His medical history was non-contributory. The physical examination showed a well-developed, well-nourished

man in no apparent distress. No facial asymmetry or discoloration was noted.

The oral examination revealed a small painless bony swelling in the oral vestibule of the left mandible from the second premolar to the third molar (Fig. 1A). The overlying mucosa was normal and the second molar tooth was mobile. Sensory function of the mandibular branch of the trigeminal nerve was normal bilaterally.

Panoramic radiographs revealed a multilocular mandibular lesion with mixed radiolucent–radiopaque content. The lesion had poorly defined borders, and was causing resorption of bone in the inferior cortex (Fig. 1B). Computed tomography revealed a mixed radiolucent–radiopaque lesion in the left mandible and thin expanded cortex and perforation of the mandible; the margin was ill defined (Fig. 2).

Based on the clinical and radiological features of the lesion, myxoma, ameloblastoma, and odontogenic tumor were suspected. A segmental mandibulectomy was performed using an extra-oral approach, from the distal aspect of the left mandibular canine to the mandibular angle, with clear margins of approximately 1.0 cm on each side of the lesion. The mandible was reconstructed with iliac bone and a titanium plate.

The tumor was located within the mandible, with an ill-defined margin, and composed mostly of myxoid matrix. Numerous calcified products were present, with a globular or trabecular pattern (Fig. 3). Microscopically, the tumor was composed mainly of myxomatous stroma in which a few stellate and spindle-shaped cells were suspended (Fig. 4). A few odontogenic epithelial nests were

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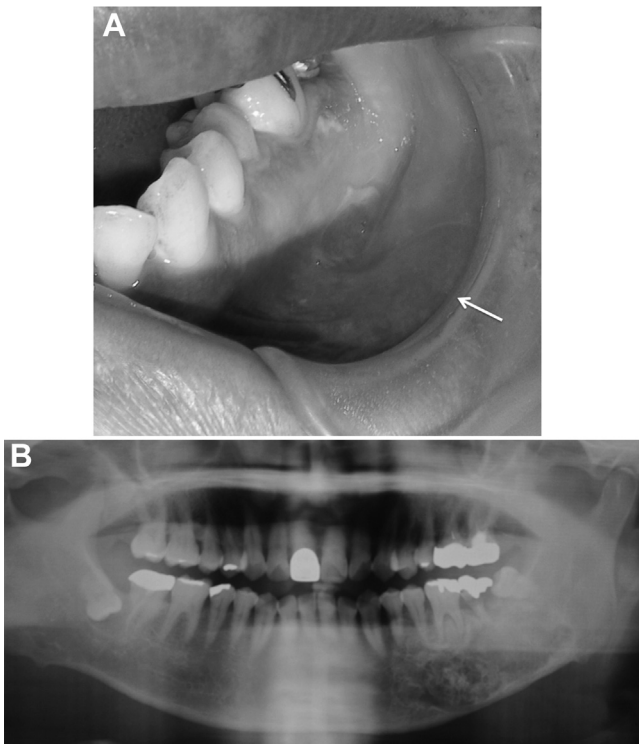


Fig. 1. (A) Clinical view of a small painless bony swelling in the oral vestibule of the left mandibular posterior region (arrow). (B) Panoramic radiographs revealed a multilocular lesion with mixed radiolucent–radiopaque content.

also observed (Fig. 5A). In addition, there were osteocement-like spherules with concentric lamination (Fig. 5B). Based on the histological findings, the tumor was diagnosed as an odontogenic myxoma. Follow-up at 10 months revealed no recurrence of the tumor.

3. Discussion

An odontogenic myxoma is a rare tumor that can occur at any age [2,3,5–7]. The majority of cases are diagnosed in the 2nd–4th decades of life [2–5]. In general, odontogenic myxomas are very rare in children younger than 10 years and in adults older than 50 years [3,8]. Odontogenic myxomas are generally

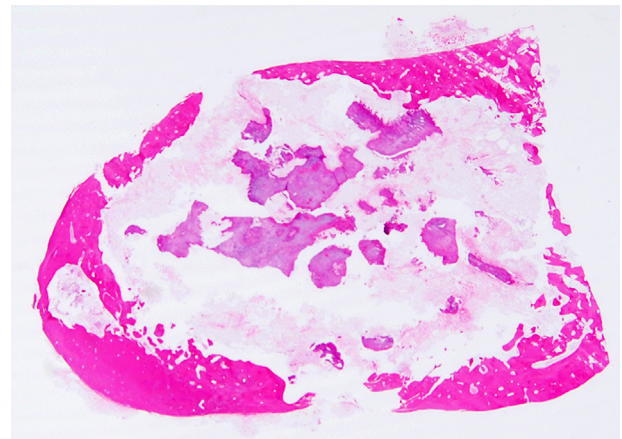


Fig. 3. Microscopic findings The tumor is located within the mandible, showing ill-defined margin, and mostly composed of myxoid matrix (Hematoxylin-eosin) (loupe view).

slow growing tumors with the potential to attain considerable size without producing noticeable signs or symptoms [7,9]. Some rapidly growing odontogenic myxomas have been reported [4,10]. Two-thirds of odontogenic myxomas are located in the mandible [11–13]. In both jaws, the molar region is most often affected [2–5].

Radiologically, an odontogenic myxoma may appear as a unilocular or multilocular radiolucency, with honeycomb or soap-bubble patterns, cortical expansion, and tooth displacement [2–4]. They are commonly associated with unerupted teeth [14]. Most odontogenic myxomas are reported to be multilocular with coarse or angular trabeculations [9], although some studies reported an equal distribution between unilocular and multilocular tumors [15,16]. Our patient had a multilocular mandibular lesion with a mixed radiolucent radiopaque content. The radiographic differential diagnosis of multilocular odontogenic myxomas should include ameloblastoma, intraosseous hemangioma, aneurismal bone cyst, central giant cell granuloma, cherubism, metastatic neoplasm to the jaws [15], glandular odontogenic cyst, and keratocystic odontogenic tumor [17]. The radiographic differential diagnosis of unilocular odontogenic myxomas should include simple cysts [15].

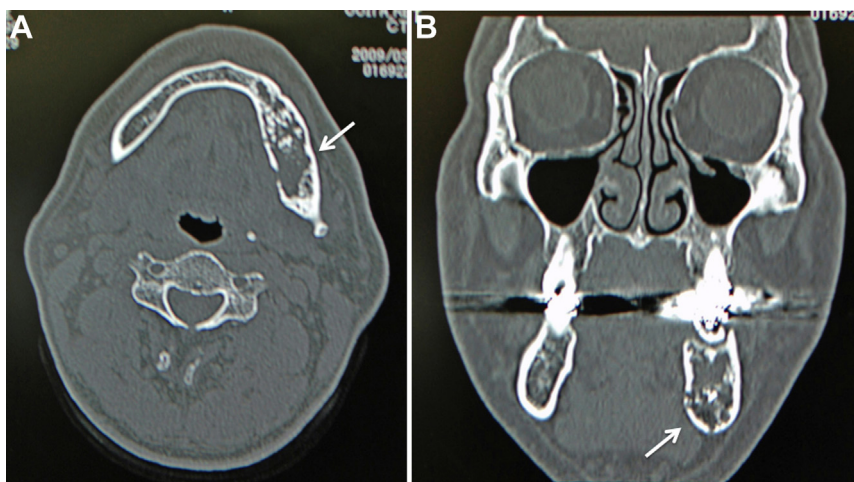


Fig. 2. Computed tomography view. (A) Computerized tomography revealed a mixed radiolucent–radiopaque lesion in the left mandible and expansion and perforation of the mandible (arrow) (horizontal view). (B) Computerized tomography revealed a mixed radiolucent–radiopaque lesion in the left mandible and expansion and perforation of the mandible (arrow) (coronal view).

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