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Case report

Central neurofibroma of the mandible: Report of a case and review of the literature☆

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ABSTRACT

A case of solitary central neurofibroma of the mandible arising in a 70-year-old man is reported. The patient had a radiolucent lesion, measuring 16 mm in diameter, with central radiopaque freckles in the right incisor–canine region of the mandible, which was discovered incidentally during routine dental check-up. The lesion was persisted for 10 years without any symptoms or complication. It was clinically diagnosed as benign tumor and surgically removed under general anesthesia. Histologically, the removed tumor was relatively well demarcated and composed of a sparse proliferation of elongated spindle-shaped cells with wavy nuclei in loosely textured connective tissue stroma, which was associated with bone formation. Clinicopathological features of central neurofibroma of the jaw bone are reviewed from the literature. This is the first report of central neurofibroma arising in the anterior part of the mandible.

1. Introduction

Neurofibroma, a benign neurogenic tumor, is considered to be originated from neural crest-derived Schwann cells of the nerve sheath, perineural fibroblasts, or both [1–3]. It is reported more frequently in the soft part of the head and neck region than in other parts of the body [4]. Neurofibroma of the head and neck has been believed to arise along the 5th, 7th–9th, or 11th–12th cranial nerves [3,4], while its intraoral occurrence is uncommon [5]. Its occurrence within bone tissues of the mandible and maxilla is extremely rare [4,5]. Solitary intraosseous (central) neurofibroma in bones other than the jaw has been further rarely reported in the literature [6]. Within the jaw bone, they mostly involve the mandible but less frequently the maxilla [4,5].

This particular intraosseous tumor tends to persist for years especially in the absence of clinical symptoms without awareness by patients and is usually discovered incidentally [4,5,7–9]. Such a clinical course seems to reflect its characteristic histology with degenerative changes and sometimes calcification, which has been rarely reported in the literature [10,11]. This report describes such a rare long-standing case of central neurofibroma arising in an infrequent location of the incisor–canine area of the mandible, emphasizes its histopathological characteristics, and reviews the literature to characterize clinicopathological features of central neurofibroma of the jaw bone.

2. Case report

A 60-year-old male patient was referred to our hospital for dental caries treatment. On a panoramic X-ray, a painless well-defined radiolucent lesion with multiple radiopaque freckles was incidentally discovered in the right incisors–canine region of the mandible. The lesion, measuring about 15 mm in longest diameter, had a thin and smooth radiopaque rim. There was no relation of the lesion to tooth roots (Fig. 1A). The patient was not aware of the lesion

☆ AsianAOMS: Asian Association of Oral and Maxillofacial Surgeons; ASOMP: Asian Society of Oral and Maxillofacial Pathology; JSOP: Japanese Society of Oral Pathology; JSOMS: Japanese Society of Oral and Maxillofacial Surgeons; JSOM: Japanese Society of Oral Medicine; JAMI: Japanese Academy of Maxillofacial Implants.

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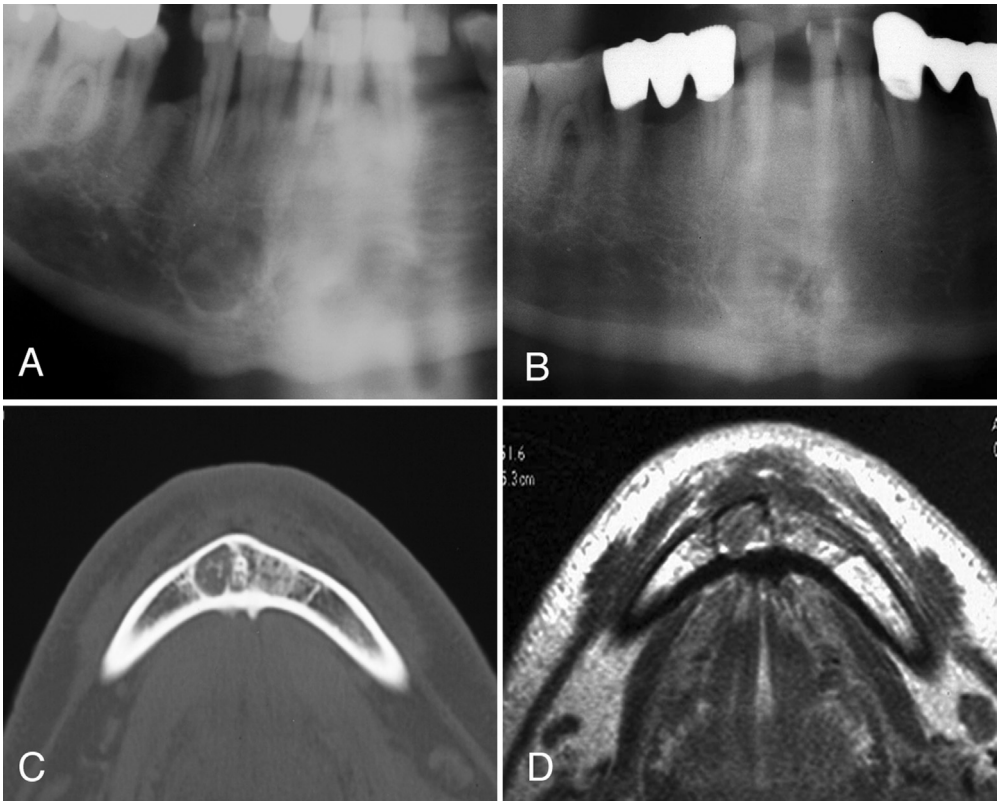

Fig. 1. Radiographic images of central neurofibroma: (A) panoramic X-ray; (B) panoramic X-ray, 10 years before; (C) CT scan, axial, bone window; (D) MRI, axial, T_1-weighted, contrast-enhanced. A well-demarcated radiolucent lesion was found in the right central incisor to the canine region of the mandible (A), which was similar in size and location to one 10 years prior (B). CT images disclosed a well-demarcated osteolytic lesion with multiple radiopacities (C). The lesion had intermediate T_1-weighted signal intensities, which equaled those of muscle tissues, on MR images (D).

and did not want any further treatment for it at that time. He did not want to receive further examination for the mandibular lesion. After ten years from the first visit, the patient, who was 70 years old by this time, revisited the hospital for treatment for the mandibular lesion because the mandibular lesion was again pointed out from his home dentist. A new panoramic X-ray showed no obvious difference from the initial findings (Fig. 1B). Axial computer tomography (CT) images in the bone window disclosed a well-demarcated osteolytic lesion with multiple scattered radiopaque materials. It was isolated from either the bone marrow or the lower cortical plate of the mandible, though there was slight thinning of the buccal cortical plate (Fig. 1C). Axial T1-weighted MR images showed intermediate signal intensities of the lesion, which were equal to those of muscle (Fig. 1D). It was clinically diagnosed as benign tumor, and was easily removed surgically under general anesthesia.

The surgical material was an elastic hard and globular-shaped mass covered by smooth-surfaced grayish white connective tissue, measuring 16 mm × 11 mm × 8 mm in size. It was sagittally cut into three slices. On the cut surface, there was a well-circumscribed, oval-shaped, and grayish white-colored fibrous tumor, measuring 10 mm in diameter, with scattered small hard materials in it. After brief decalcification, the tumor tissue was routinely processed for hematoxylin and eosin stained sections. Histopathologically, The lesion was a well demarcated fibrous nodule scattering small hard tissues with an impression-like space for a tooth root in the upper end. The peripheral area was more densely cellular, while the central area looked more spongy. There was no definite fibrous capsule, though the hard tissue were arranged in parallel to the peripheral boundary, which looked periosteal reaction, while those in the central area tended to be larger and more round-shaped (Fig. 2A). The tumor was composed of a sparse proliferation of elongated spindle-shaped cells with wavy nuclei in loosely textured fibrous connective tissue. Most of the hard tissues resembled bone trabeculae but showed different maturation degrees. The tumor stroma was rich in vascularity (Fig. 2B and C). In the central area, the tumor cells were loosely arranged showing a meshwork like pattern (Fig. 2C). Thin but definite peripheral nerve fibers are scattered in the center of the tumor. Mast cells were abundant but no other inflammatory cell infiltration was seen within the tumor. Most of the spindle-shaped tumor cells were immunohistochemically positive for S-100 protein (Fig. 2D).

3. Review of the literature

Our review of the English literature from 1944 to 2011 revealed a total of 39 cases of central neurofibroma of the jaw bone with complete clinical data. Our present case is the 40th one to be documented. The 40 cases are summarized in Table 1. Thirty-four cases (85%) occurred in the mandible [4–30] and 6 cases (15%) in the maxilla (31–36). Among the 34 mandibular cases, 13 cases were restricted to the molar region (38.2%), 5 cases in the molar–premolar region (14.7%), 3 cases in the molar–ramus region (8.8%), 2 cases in the premolar–ramus region (5.8%) and one case in the incisor–molar region (2.9%), 4 cases were restricted to the premolar region (11.7%), 4 cases within the ramus (11.7%), one case in the incisor–premolar region (2.9%), and one case in the incisor–canine region (the present case). Among the six maxillary cases, four cases were in the molar region [31–33], one case in the premolar region [34], one case in the canine–premolar region [35], and one case in the incisal region [36].

Regarding to gender, there were 19 male and 21 female patients with ratio of 1:1.1, which indicated no predilection between males and females. The mean age was 27.9 years ranging from 5 months