



## Case report

# Central hemangioma of the maxilla: report of a case with an exophytic growth

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## KEYWORDS

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**Summary** A case of intraosseous hemangioma occurring in the maxilla of a 74 year-old-female is described. A radiolucent lesion in the maxilla pointed out by her dentist grew rapidly in a month. CT examination revealed an intraosseous mass lesion with scalloping margins and rapid enhancement with contrastive media. MRI indicated a well-demarcated and exophytic lesion with high T2-weight signals. The lesion, measuring 20×38×58 mm in size, was clinically suspected of malignancy and surgically resected together with the right half of the maxillary bone. Histopathologically, the lesion was a fibrous granulation tissue absorbing the maxillary bone and expanding into the oral mucosa. It was largely occupied with blood coagulae in the center and contained many dilated blood vessels with single-layered endothelial linings in the periphery including the bone. This is the first case report of central hemangioma with an exophytic growth.

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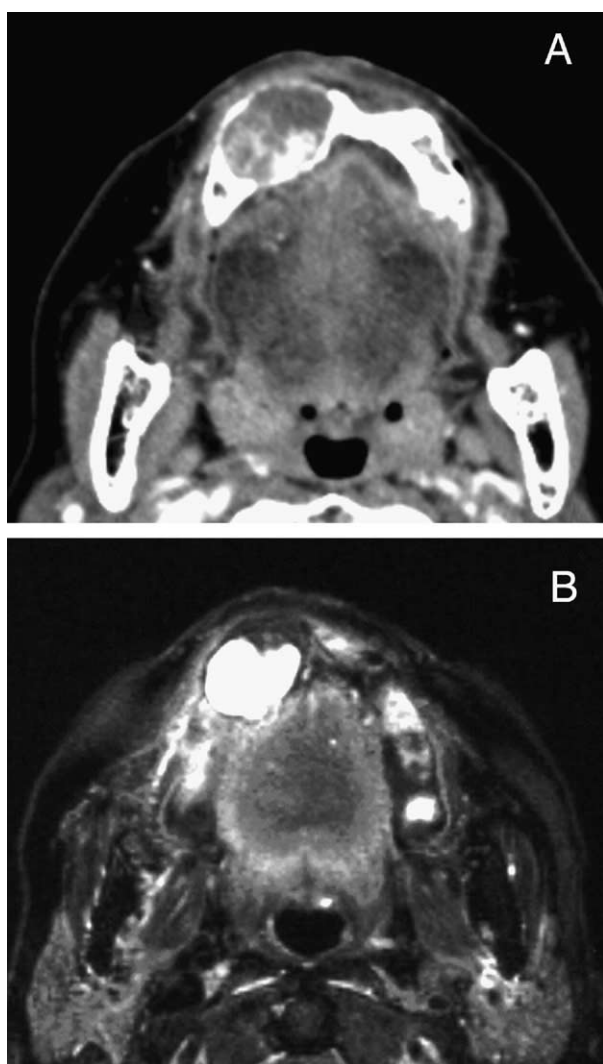
A 74-year-old Japanese woman visited her dentist complaining about unfitness of her maxillary partial

denture, which she had worn with periods of interruption during two years after extraction of right molar teeth due to severe periodontitis. On radiographic examination, a radiolucent lesion in the right premolar region of the maxilla was found, and she was referred to the Niigata University Dental Hospital. At the first visit, the patient had a conspicuous swelling around her nasal vestibule.

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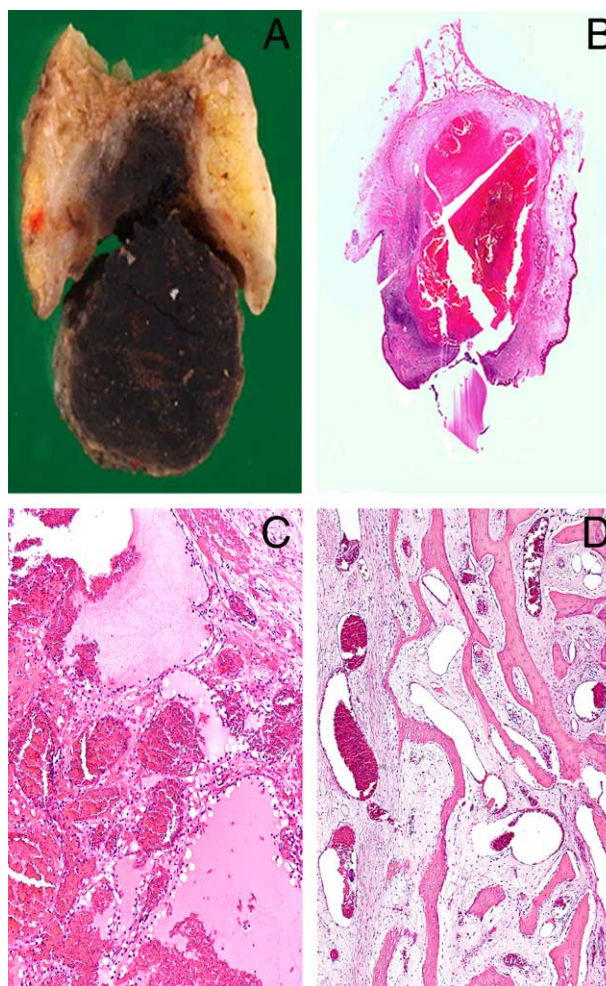
Intraorally, there was a diffuse and erythematous protrusion arising from the maxillary gingiva of the anterior to premolar region. The canine and lateral incisor teeth showed significant mobility and the right second premolar was removed due to the expansion of the lesion. CT examinations revealed an intraosseous mass lesion with scalloping margins, ill-defined margins in the vestibular side and a well-demarcated margin in the lingual side (Fig. 1A). Two biopsies were performed but their histopathological diagnoses were just granulation tissue or fibrin clots. One month after the first visit, the lesion started to grow rapidly into the oral cavity and formed a bleeding and dome-shaped protrusion at the molar region. Its surface was covered with



**Figure 1** Diagnostic images for central hemangioma in the maxilla of a 74-year-old female. (A) CT image showing an intraosseous mass lesion with scalloping margins irregularly enhanced with contrastive media. (B) MR image showing high T2-weighted signals in the intraosseous as well as exophytic parts.

necrotic tissues. MRI images demonstrated high T2-weighted signals in the intra-bony as well as exophytic parts (Fig. 1B). Based on clinical suspicion of malignant vascular tumor, hemi-maxillectomy was performed under general anesthesia.

On cut surface of the surgically removed specimen, a dark-brown colored round-shape coagulum was located in the maxillary bone. The coagulum extended to the base of the maxillary sinus and was



**Figure 2** Surgical specimens of central hemangioma: (A) cut surface of a polypoid mass protruded from the premolar region of the gingiva. Dark brown-colored and round-shaped blood coagulum located within the maxillary bone extending to the base of the right maxillary sinus; (B) cut surface of the maxilla at the canine region. HE, 1.5 $\times$ . A large blood coagulum involved the maxilla bone entirely, but the gingival mucosa was intact; (C) high power view of (B). HE, 50 $\times$ . The coagulum was surrounded by fibrous granulation tissue, in which there is a proliferation of thin walled cavernous vessels lined by a single-layered endothelial cells and (D). HE, 120 $\times$ . The medullary space of the surrounding bone was fibrous scattering cavernous blood vessels between bony trabeculae.

continuous with the core portion of the pedunculated tumescent mass on the gingiva (Fig. 2A). Histopathologically, the major part of the lesion was a large blood coagulum showing a minimal trace of blood vessel structures in it, and there was no evidence of malignancy and invasive growth (Fig. 2B). The blood coagulum was surrounded by fibrous granulation tissue with a prominent proliferation of thin-walled cavernous vessels lined by single-layered flat endothelial cells. The maxillary bone was in a large part absorbed by the lesion (Fig. 2C). There were occasionally cholesterol granulomas in the luminal surface of the granulation tissue. Vascular proliferation was seen not only around the coagulum but also within the medullary space of the surrounding bone (Fig. 2D). The patient's post-operative course was uneventful for one year.

## Discussion

Central hemangioma of the jaws is a rare lesion [1–4], representing only 0.8% of 120 intra-oral hemangiomas and constituting less than 0.2% of all bony neoplasms recorded in our surgical pathology services from 1965–2002. Its incidence among oral hemangiomas and jaw bone tumors has not been well documented in the literature. Its occurrence is twice as common in the mandible than in the maxilla among only 18 cases retrieved from the literature. The peak incidence is in the second decade of life differing from our patient and there is a sex predilection that favors female 2:1 over males [1–3]. It is generally accepted that intraosseous hemangiomas to be hamartomas [1,2] and vascular malformations [5] rather than true neoplasms.

Most of the reported central hemangioma cases were located within the bone sometimes presenting hard and non-tender swellings but generally

without symptoms [1–4]. In contrast, the present case showed the rapid exophytic expansion in a short interval, which was suggestive of malignancy, partly because the repeated biopsies did not work. Although it remained uncertain why the central hemangioma outgrew into the oral cavity from the clinical history, some traumatic or inflammatory stimulations due to the unfitted denture or her longstanding periodontitis might induce granulation reaction against hemorrhage or thrombus formation in the hemangioma. Repeated biopsies might enhance such granulation processes. Thus, we considered it worthwhile to document that central hemangiomas, especially those arising in the maxillary bone, have a potential to grow outside by destructing the cortical plate in this first case report.

Histopathologically, the surgical specimen showed cavernous hemangioma involving the medullary space of the maxillary bone. Among the 18 reported cases, five were histologically cavernous, one was capillary, and others were not described [3,4]. Central hemangiomas should be differentiated from arteriovenous malformations, in which expansive growth and bleeding occur usually after trauma or infection, resulting in acute painful swelling [5]. In the present case, however, vascular structures are all uniformly cavernous and there was no symptom of pain in the patient's history.

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