A case of desmoplastic ameloblastoma occupying maxillary sinus

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Abstract

We report a rare case of desmoplastic ameloblastoma lesion that filled the entire maxillary sinus. The patient visited our hospital with a chief complaint of swelling around the upper left premolars. A panoramic X-ray captured an image of a mixture of ill-defined radiolucency and radiopacity from the swollen area to the maxillary sinus. Computed tomography (CT) and Magnetic resonance imaging (MRI) showed that the lesion occupied almost the entire left maxillary sinus and had entered the nasal cavity. A pathologic diagnosis of ameloblastoma was made after biopsy, and the tumor was removed and the marginal bone curetted under general anesthesia. A CT scan at 4 months postoperatively indicated the presence of residual and recurrent tumor in the area of the upper left lateral incisor, and removal and curettage were performed again. Recurrence may be detected relatively easily based on radiographic characteristics, and therefore follow-up with an X-ray examination such as a CT scan is important.

Keywords: Desmoplastic ameloblastoma, maxillary sinus

Introduction

Several cases of desmoplastic ameloblastoma (DA) of the maxilla that extended into the maxillary sinus have been described,[1-5] but there has been no report of DA occupying the entire sinus. Here, we report a rare case of a DA lesion that filled the entire maxillary sinus.

Case Report

The patient was a 29-year-old Japanese female who visited the Department of Oral and Maxillofacial Surgery at Mie University Hospital with a chief complaint of swelling around the upper left premolars. The swelling had persisted for 1 month without subjective symptoms such as pain before she was referred to our hospital by a local dentist.

At the initial examination, there was no buccal numbness, tenderness, nasal congestion, or eye manifestation. Palpation of the oral cavity indicated the presence of a bone-like and hard, but painless, mass [Figure 1]. A panoramic X-ray captured an image of a mixture of ill-defined radiolucency and radiopacity that extended from the swollen area to the maxillary sinus, and revealed root divergence between the upper left first premolar and second premolar, but the absence of periapical resorption [Figure 2]. A Computed tomography scan showed that a lesion containing bone-like tissues occupied almost the entire maxillary sinus. The lesion extended from the alveolar process of the premolars to the anterior region of the outer wall of the maxillary sinus, the palatine process, the lateral wall of the nasal cavity, and close to the floor of the orbit. The bone wall was relatively well preserved, but a part of the lesion had entered the nasal cavity [Figure 3]. The lesion showed heterogeneous isointensity on T1-weighted MRI and hypointensity on T2-weighted Magnetic resonance imaging [Figure 4].

The tumor was pathologically diagnosed as ameloblastoma based on biopsy results. Total resection of the tumor and partial resection of the maxilla was scheduled. However, based on the patient’s request and her age, removal of the tumor and curettage of the bone around the tumor, and extraction of #11, #12, #13, and #14 were performed under general anesthesia. The tumor had adhered to the bone and occupied almost the entire maxillary sinus, but did not reach the floor of the orbit or the posterior wall of the maxillary sinus. No capsule formation was seen in the tumor tissues. Histopathological findings from isolated tissues showed that the tumor parenchyma had a fiber architecture, which was similar to the enamel organ, with small alveolar or trabecular structures, and spread in the abundant interstitium consisting of scar-like fiber tissues with a few cellular components. Lamellar bone tissues were also observed. DA was diagnosed based on these findings [Figure 5]. The patient was followed up in the outpatient department after discharge, but a CT scan performed at 4 months postoperatively revealed the presence of residual and recurrent tumor in the area of the upper left lateral incisor. Therefore, removal and curettage...
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Two surgical procedures were performed again and the upper left lateral incisor was extracted. The subsequent course has been benign and the patient has been followed up in the outpatient department as of 12 months postoperatively.

Discussion

Ameloblastoma is the most common odontogenic epithelial tumor, accounting for 11–18% of all odontogenic tumors, and DA accounts for 4–12% of ameloblastomas. DA occurs in men and women aged 20–80 years old. The most common site is from the anterior teeth to the premolars in both the maxilla and mandible, unlike other types of ameloblastoma, which typically occur in the mandibular molars. The size is relatively small and the longest diameter is generally 2 cm or less.

The majority of ameloblastomas show radiolucency on X-ray, while approximately 50% of DA cases have a mixture of radiolucency and radiopacity. This suggests that DA infiltrates trabeculae and is sometimes associated with bone.
formation. DA is a histologically solid tumor without capsule formation, and is characterized by abundant collagen fiber formation in the interstitium. Yoshimura et al. reported that DA lesions were generally smaller than other ameloblastoma lesions because DA growth might be inhibited by collagen fibers. However, the DA lesion in our case occupied almost the entire maxillary sinus, which suggests that collagen fibers are unlikely to inhibit DA growth.

There are several reports of resection and curettage for treatment of DA, but the risk of recurrence is higher in these procedures compared to total resection with recurrence in 6 of 19 cases treated with removal and curettage. DA with extensive spreading to the maxillary sinus is relatively common, but a case with the lesion occupying almost the entire sinus, as in our case, is rare. DA generally grows slowly, but once it enters the maxillary sinus the growth may be accelerated because there are no tissues to interfere with growth. The prognosis is good with rare recurrence when extended resection is applied in cases of DA that spread to the maxillary sinus, but recurrence has been reported after removal and curettage as in our case.

Extended resection is the first line therapy for a case in which the tumor has entered and occupied the maxillary sinus. If removal and curettage are requested by the patient or performed for other reasons, wide removal of the lateral wall of the nasal cavity is required to allow detection of recurrence and inspection of the inside of the maxillary sinus from the opening of the nasal cavity using a fiberscope. Therapy should be selected based on the quality of life of patients, and the risk of recurrence following removal and curettage should be disclosed to the patient. Recurrence may be detected relatively easily based on radiographic characteristics, and therefore follow-up with an X-ray examination such as a CT scan is important.

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