

ABSTRACT

Ameloblastoma is the most common clinically significant epithelial odontogenic tumor, and is considered a benign but locally aggressive tumor of the craniofacial region. We present the case history of a 46-year-old male patient who had a nasopharyngeal carcinoma (NPC) treated by irradiation, which was complicated by the occurrence of an ameloblastoma. Following a review of the English language literature, this case history was identified as being the first report, to our knowledge, of the occurrence of ameloblastoma in a patient who had been irradiated for an NPC. This case report highlights the importance of regular dental care for a patient who had an NPC and was treated with radiation therapy.

KEY WORDS: ameloblastoma, radiotherapy, NPC, dental care

Occurrence of ameloblastoma in a patient with nasopharyngeal carcinoma after treatment by irradiation

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Spec Care Dentist 30(1): 27-30, 2010

Introduction

Nasopharyngeal carcinoma (NPC) is a rare head and neck malignancy in Caucasians, but it is particularly common in the southern Chinese population of Guangdong and in Hong Kong, Singapore, and Taiwan.¹ The standard treatment for NPC is radiotherapy (RT), but concurrent adjuvant chemotherapy improves survival rates.² Oral complications of radiation therapy (RT) for NPC are very common. The most common acute complications are mucositis, infection, xerostomia, and taste changes, whereas chronic complications due to prolonged effects of RT comprise hyposalivation, infection, taste changes, dysphagia, and trismus.³ Patients treated for NPC may have a high long-term risk of developing radiation- or chemotherapy-induced malignancies (second malignant tumor) after successful treatment for NPC.⁴

Ameloblastoma is the most common form of benign odontogenic tumor, predominantly originating in the mandible in 80% of cases and occurring in the molar-ramus area. It is a true neoplasm of enamel organ-type tissue that does not undergo differentiation to enamel formation.⁵ There are three clinical types of ameloblastoma: solid (multicystic), unicystic, and peripheral. The solid or multicystic ameloblastoma has a tendency to be locally invasive and has a high incidence of recurrence if not adequately removed. The unicystic type has a fibrous connective-tissue capsule sharing considerable similarities with odontogenic cysts both clinically and radiographically and, therefore, has a lower rate of recurrence. Peripheral

ameloblastomas occur solely in the soft tissues covering the tooth-bearing area.⁵

This paper reports a patient who had an NPC that was treated by irradiation and complicated by the development of an ameloblastoma during a period of loss from follow-up. Following a review of the English language literature, this case report was identified as being the first report, to our knowledge, of the occurrence of ameloblastoma in a patient who had a history of NPC treated by irradiation.

Case description

A 46-year-old Chinese male was diagnosed with NPC [Stage IIA according to the guidelines of the American Joint

Committee on Cancer (AJCC)⁶]; a radiation therapy (RT) regimen (once a day, five days per week, with a radiation dose of 225 cGy each time) was completed in a regional hospital in central Taiwan in March 2002. Due to migration to the city of Kaohsiung in southern Taiwan, the patient visited the Ear, Nose, and Throat (ENT) Department of Kaohsiung Medical University for his regular post-NPC follow-up. The patient was referred to the Department of Oral Pathology for dental care after RT for the NPC.⁷ The patient's oral habits included smoking cigarettes, drinking alcohol, and chewing betel-quid; he was tested positive for an Epstein-Barr virus serum test. An evaluation of a panoramic radiograph made at the initial dental appointment revealed normal bony findings except for the bilateral impacted mandibular third molars and bilateral malposed maxillary third molars (Figure 1). The patient was then lost for follow-up for about 2 years until July 2004. He returned with complain of discomfort over the right mandibular second bicuspid area. An intraoral examination showed a normal appearance over the vestibular area from the right mandibular canine to the second bicuspid. No obvious symptoms/signs were observed other than mild pain on percussion of the right mandibular second bicuspid. Neither dental caries nor any restoration was noted on this tooth, but there was no response to an electric pulp test, while adjacent teeth responded positively. Due to the negative vitality test, emergency endodontic treatment of this tooth was done and a panoramic radiograph was made (Figure 2). Compared with the first panoramic radiograph made in 2002, evaluation of this radiograph revealed a multilocular radiolucency around the right mandibular second bicuspid, and mesial root resorption of the right mandibular first molar. The patient was subsequently referred to the Department of Oral and Maxillofacial Surgery for further management. The initial clinical diagnosis by the oral surgeon was that the lesion was probably an ameloblastoma. Due to the history of RT, conservative enucleation was carried out



Figure 1. The first panoramic radiograph taken at the initial dental appointment revealed no bony lesion.



Figure 2. The second panoramic radiograph revealed a multilocular radiolucency around the right mandibular second bicuspid; mesial root resorption of the right mandibular first molar was suspected.

and tissue samples were sent for a histopathologic examination. The microscopic diagnosis revealed an ameloblastoma of a follicular type with acanthomatous changes. The patient again did not return for follow-up for three and a half years. In January 2008, he returned to our dental clinic with a mild swelling of the right mandibular posterior area. An intraoral examination revealed a buccal swelling corresponding to the right mandibular second bicuspid and the first molar. Upon panoramic

radiographic examination, a well-defined multilocular irregularly shaped radiolucency without a corticated margin was identified, extending to the distal aspect of the right mandibular first bicuspid (Figure 3a–3c). Therefore, a recurrence of the ameloblastoma was strongly suspected and the patient was again referred to the Department of Oral and Maxillofacial Surgery for surgical treatment. Due to the irradiation history, conservative enucleation was done again. A histological diagnosis of

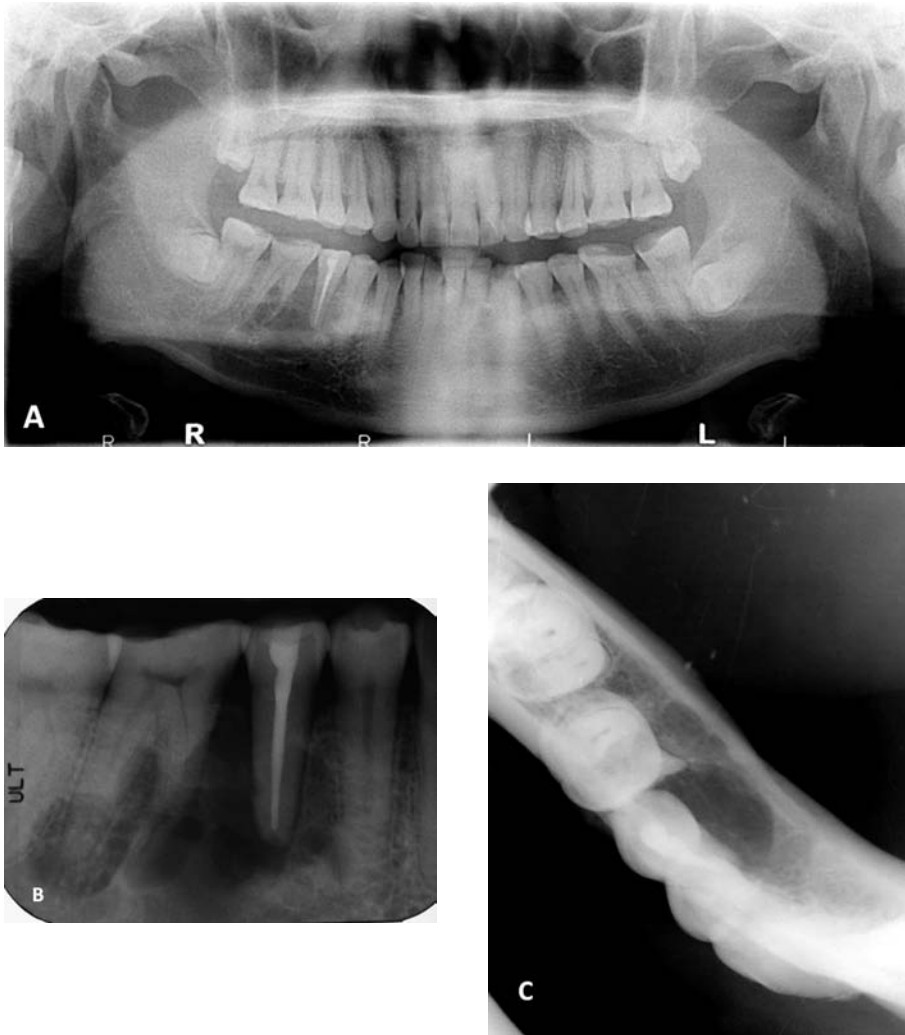


Figure 3. The third panoramic radiograph (a) as well as periapical (b) and occlusal (c) radiographies revealed a well-defined multilocular irregularly shaped radiolucency without a corticated margin, extending from the distal aspect of the right mandibular first bicuspid to the distal aspect of the right mandibular first molar.

ameloblastoma was again reported. The patient then returned for regular follow-up in the dental department. The patient was also referred to the ENT Department, where doctors found no recurrence of the NPC.

Discussion

As reported in our previous study,⁷ patients who have had irradiation therapy for NPC should develop a dental care regimen. Unfortunately, the patient in this report failed to attend for scheduled dental care after the initial visit;

otherwise, the ameloblastoma could have been detected earlier. Seldom painful unless secondarily infected, an early ameloblastoma in the jawbone may often produce no clinical signs or symptoms. An early diagnosis could only have been made by careful evaluation of a radiograph, particularly a panoramic radiograph. The occurrence of ameloblastoma in a patient who has been irradiated for an NPC may be rare; however, this particular case history emphasizes the importance of having regular dental follow-up appointments that should include a radiographic exam-

ination. Dental problems as well as any other pathologies after RT, although rare, might have been detected earlier.

To our knowledge, this is the first reported case history of an ameloblastoma in a patient who has been irradiated for an NPC, although second primary tumors may be radiation-induced in patients who have long-term survival after RT for NPC.⁴ In our patient, the ameloblastoma was noted very soon after RT. Ameloblastomas, despite being locally aggressive, are a benign odontogenic tumor. Therefore, we believe that the presence of the ameloblastoma in our patient was coincidental; and there was no direct relationship to the RT. Nevertheless, dental care providers for patients undergoing RT should be clinically aware of the possible occurrence of secondary tumors.

The optimal treatment for ameloblastoma includes enucleation and a more radical resection. Radiation treatment or RT combined with chemotherapy has also been shown to be able to control and reduce the size of ameloblastomas in patients with advanced or unresectable lesions.⁸⁻¹⁰ Our patient had undergone irradiation for NPC before the ameloblastoma was diagnosed; no clinical symptoms/signs in the oral cavity were reported during this period. Therefore, it was logical to consider whether irradiation might have exerted an inhibitory effect to localize or limit the growth of ameloblastoma temporarily or delay its development. This may explain why the ameloblastoma lesion in our patient developed very quickly into a noticeable lesion within about 2 years after irradiation was terminated.

Ameloblastoma is a benign but locally invasive neoplasm and frequently recurs after conservative surgery.¹¹ In a systematic review, Lau and Samman¹² reported recurrence rates of 3.6% after resection, 30.5% for enucleation alone, 16.0% for enucleation followed by application of Carnoy's solution, and 18% for marsupialization with or without other treatment in a second phase. In our patient, recurrence occurred almost 4 years after enucleation of the lesion. Due to the

irradiation history, instead of a radical mandibular resection, conservative treatment was carried out for this patient. Considering the high rate of recurrence of ameloblastoma, long-term close follow-up for such a patient is essential.

Conclusion

This case history reports ameloblastoma in a patient who had been irradiated for an NPC and emphasizes that regular dental care and routine panoramic examination is essential for such a compromised patient. Early diagnosis and periodic follow-up is necessary due to the high recurrence rate of ameloblastoma.

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