CASE REPORT

Giant ameloblastoma of jaw successfully treated by radiotherapy

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Summary Ameloblastomas are uncommon tumors, which were thought to be radioreistant. However there is lack of well-documented evidence in the literature concerning their relative radio responsiveness or radio resistance nature. The present article reports a case of ameloblastoma of jaw, successfully treated by radiotherapy.

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KEYWORDS Ameloblastoma; Adamantinoma; Jaw; Megavoltage radiotherapy

Introduction

Ameloblastoma (Adamantinoma) are generally considered to be radio resistant tumors. However, there is lack of well-documented evidence in the literature concerning their relative radio responsiveness or radio resistance. Many of the old reports mean little because radiotherapy was changed dramatically with the introduction of megavoltage irradiation in the late 1950s, allowing for better distribution of the radiation to the tumor than had previously been possible. The present article reports a case of ameloblastoma of jaw, successfully treated by radiotherapy. An attempt has been made to answer the question of the value of modern therapeutic irradiation in the treatment of ameloblastoma.

Case report

A 64 years old male referred to our head and neck cancer clinic in October 2003 with chief complaints of gradually increasing painless swelling in the right side of face for the past 8 years. Physical examination revealed a huge firm to hard growth on right side of the upper and lower alveolus. A firm to hard growth was present in the right infratemporal fossa. No significant lymphadenopathy was found in the cervical and supra clavicular area. All the baseline investigations including complete hemogram,
liver function test, renal function test, X-ray chest and USG abdomen were well within normal limits. A contrast enhanced CT scan face revealed a big soft tissue mass on right side of the neck and face with distorted normal anatomy (Fig. 1). The mass was invading the oral cavity, buccal mucosa and pressing over the oropharynx. Mass was also invading the right infratemporal fossa. There was a central area of necrosis with patchy contrast enhancement seen. Histopathological examination revealed dense cluster of pleomorphic tumor cells with big hyperchromatic nuclei (Fig. 2). There were clusters of tumor cells with large oval to roundish nuclei with ill-defined pink cytoplasm and dispersed chromatin confirming the diagnosis of ameloblastoma.

**Figure 1**  A contrast enhanced CT scan face showing a big soft tissue mass on right side of the face and neck. The mass was invading the oral cavity, buccal mucosa and pressing over the oropharynx. Mass was also invading the right infratemporal fossa. There was a central area of necrosis with patchy contrast enhancement seen.

**Figure 2**  (H&E ×1000—in microscope) smear shows a dense cluster of pleomorphic tumor cells with big hyperchromatic nuclei. There were clusters of tumor cells with large oval to roundish nuclei with ill-defined pink cytoplasm with dispersed chromatin.
Due to the advance and inoperable nature of tumor, patient was referred to us from department of surgical oncology for the favor of radiation therapy. Patient was subjected to radiation therapy after metastatic work up. He was given 60 Gy in 30 fractions using 2 Gy fraction, 5 days a week by parallel and opposed lateral fields with 2:1 weightage on the affected side by Theratron 780C machine shielding the eye and brain. The radiotherapy was well tolerated. The soft tissue swelling showed dramatic response and started resolving slowly which was assessed by serial clinical and radiological examination. At present he is having a facial asymmetry of right side with near to complete resolution of the disease in the latest CECT (June 2005) and a KPS of 100 (Fig. 3). He has completed 2 years of radiation therapy with no further disease activity.

Discussion

Term ‘Adamantinoma’ was introduced by Malassez\(^1\) in 1885 to denote the odontogenic tumor that had been recognized by Cusack\(^2\) in 1827 and described later by Falksson.\(^3\) Ivey and Churchill in 1930 changed the name to ‘Ameloblastoma’.\(^4\) The ameloblastoma is an odontogenic epithelial tumor histologically similar to the embryonic tissue that produces enamel during tooth formation.\(^5\) The biological behavior of these tumors has been well established, in that they are slow growing, locally invasive tumors with a high rate of recurrence if not removed adequately.\(^6\) Despite the extensive literature, the principles of treatment are still not well established, although most authors reviewing surgical treatments of ameloblastoma see m to regard ameloblastomas as radioresistant, and hence find no place for irradiation in the treatment armamentarium.\(^5,7,8\) These results were obtained in the premegavoltage days of radiation therapy, Sehdev et al.\(^8\) in their review of the Memorial Sloan-Kettering experience, mentioned 11 patients treated during the period of 1921–1951 prior to the introduction of mega voltage external irradiation. Three patients had persistent disease and six had an initial response followed by later recurrence. Gardner and Pecak\(^6\) in 1980...
studied the role of radiation therapy in ameloblastoma and concluded, “There appears to be general agreement in the literature that the ameloblastoma is radioresistant and that consequently radiotherapy should not be used in its treatment”. However, there is very little evidence reported, especially recently to substantiate this belief. In 1982, Reynolds and Pacyniak9 wrote an important, well-reasoned paper on the effect of irradiation on ameloblastoma in which they discussed the basic principles of radiotherapy. They also analyzed, using modern radiotherapeutic principles. These authors concluded that clinicians still do not know if ameloblastomas are radioresistant or radioresponsive but indicated that they believed radiotherapy had a possible role in the treatment of those patients who were poor surgical risk. They also were unwilling to accept extensive surgical procedures. A second important paper is that of Atkinson et al in 1984.10 They published a series of 10 cases of ameloblastoma treated by megavoltage irradiation, seven with primary irradiation only and three with both irradiation and surgery. They concluded, on the basis of their experience and an evaluation of literature, that ameloblastomas are radioresponsive. Since then, there are few reports published in the literature supporting the role of radiation therapy as a useful modality of treatment in case of ameloblastomas.11–15

We, here present a case of advanced ameloblastoma responding well to radiation therapy supporting the view of radioresponsive nature of this tumor.

There are a number of reasons why our present knowledge is inadequate concerning the relative radiosensitivity or radioresponsiveness of ameloblastomas. But, it is apparent from this case and from the literature, that the ameloblastoma is not an inherently radioresistant tumor. We believe that properly applied megavoltage radiation techniques have a useful role in the management of these tumors, particularly in those cases where a full surgical excision would be technically difficult because of bulk and local invasion or where other medical factors including age, would make radical surgery inappropriate.

Conclusion

This case further highlights the role radiotherapy in advanced, inoperable ameloblastoma of jaw as a definitive therapeutic option. It further warrants that advanced, inoperable ameloblastoma be subjected to definitive megavoltage radiation therapy to define the role of radiotherapy in this tumor.

References