Ameloblastic Carcinoma of Mandible: A (Case Report)

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Abstract

Ameloblastic carcinoma is a rare malignant odontogenic tumor that is further classified into being primary or secondary arising from a preexisting benign ameloblastoma. It affects the mandible in two thirds of the patients. There is no standard treatment protocol for this lesion but radical surgical excision with or without radiotherapy is reported in the majority of cases. In this paper we present a case of a 60 year old female diagnosed with ameloblastic carcinoma of the mandible that was treated by radical resection of the mandible with selective neck dissection and postoperative radiotherapy.

Key words: Ameloblastic carcinoma, Mandible, Odontogenic tumorton

Introduction

Ameloblastic carcinoma, a term introduced by Shafer (Mubeen et al 2010, Ponnam et al 2012, Keerthi et al 2013), is rare and it is further classified into being primary or secondary arising from a preexisting benign ameloblastoma. (Gijiljames et al 2007, Perera et al 2013, Keerthi et al 2013) In an analysis of all the published cases of ameloblastic carcinoma in English language between the years 1984 and 2004 only 37 cases were reported (Akrish et al 2007), while less than 70 cases of Ameloblastic Carcinoma have been reported in the literature elsewhere. (Benlyazid et al 2007)

The world health organization (WHO) classification has distinguished two malignant variants of ameloblastoma, namely; metastasizing or malignant ameloblastoma which shows signs of metastasis, mostly to the lung followed by cervical lymph nodes and spine, while retaining the histological features of solid ameloblastoma, and ameloblastic carcinoma that shows histological features of malignant transformation like pleomorphism and mitosis. (Gillijames et al 2007, Pogrel and Montes 2009, Perera et al 2013)

It affects the mandible in two thirds of the patients while the maxilla is affected in the remaining one third. (Mubeen et al 2010, Keerthi et al 2013) There is no standard treatment protocol for this lesion but radical surgical excision with or without radiotherapy is reported in the majority of cases. (Arotiba et al 2005, Ponnam et al 2012, Perera et al 2013, Keerthi et al 2013)

In this paper we present a case of a patient diagnosed with ameloblastic carcinoma of mandible that was treated by radical composite resection of the mandible with selective neck dissection and postoperative radiotherapy.

Case Report

A 60-year-old female attended the consult clinic at Al-Yarmook teaching hospital complaining from a swelling involving the right side of the body of the mandible. The patient stated that the swelling was present and slowly growing for more than 2 years with a recent increase in its size within the last 2 months. On clinical examination the swelling was firm involving the molar and premolar region of the mandible extending to involve the submandibular region, the overlying skin was normal in color and texture except for a focal area where the skin was red in color and tethered to the mass at the submandibular region (Fig.1), the patient reported no numbness of the lower lip. CT scan showed a circumscribed mass of about 7cm×6cm×5cm involving
An incisional biopsy was taken from the mass via an intraoral vestibular incision. The specimen was sent for histopathological examination, and the diagnosis was Ameloblastic carcinoma. The treatment plan entailed resection of the mandible from the mandibular foramen to the mental foramen with the overlying soft tissues and skin together with selective neck dissection for the levels I through III. Reconstruction of the defect was by primary closure of the intraoral tissues and advancement of the neck flap to close the skin defect without reconstruction of the mandible, this decision was made after discussion with the anesthetist and the internist who both recommended a less extensive surgery since the patient was diabetic and hypertensive with mild cardiomegaly.

The operation was carried out under general anesthesia. The approach was through a transverse cervical incision extending from the mastoid region to the mental region, extending to the midline through the lower lip to allow a lip split. Selective neck dissection was completed first clearing level III and level II. Level I was tethered with the lesion which was removed en bloc with the mandible from the lower canine region to the mandibular foramen (Fig.3), the overlying tethered skin was removed with the specimen. Reconstruction was carried out by primary suturing of the intraoral tissues and advancement of the neck flap to close the skin defect. Postoperatively the patient was admitted to the intensive care unit for two days wherein her medical status was monitored carefully. She remained hospitalized for...
7 days with an uneventful recovery. The patient was followed up weekly during the first 3 months postoperatively and monthly thereafter. The specimen was submitted for histopathological examination where the diagnosis of Ameloblastic carcinoma was confirmed. After consultation with the oncologist postoperative radiotherapy was decided and the patient started receiving radiation therapy after about 6 weeks postoperatively. The patient was followed up on a regular monthly basis, and after 15 months follow up period, no recurrence at the primary site or cervical lymph nodes metastasis was detected based on clinical examination and ultrasonography of the neck, the patient only complained from slight difficulty in mastication ipsilaterally and she has no esthetic concerns (Fig.4).

Histopathological notes
The sections showed islands and follicles of malignant epithelial cells, with central keratinization and focal palisading columnar cells with reverse polarity. Clear cells were also seen along with significant mitotic activity and focal necrosis. The margins of bone resection were free of malignant cells, the lymph nodes of level I was part of the mass while lymph node levels II and III were free of malignant cells. Serial sections of the overlying epithelium as well as epidermis showed no dysplastic changes (Fig.5).

Discussion
In the 2005 WHO classification of odontogenic tumors, ameloblastic carcinoma is divided into a primary type and secondary (dedifferentiated) type of which intraosseous and peripheral types are distinguished. It is agreed now that ameloblastic carcinoma is the term used for tumors with histological evidence of malignancy in the primary, recurrent, or metastatic tumor regardless of whether there is metastasis or not (Arotiba et al 2005). Its pathogenesis is not clear, although repeated trauma caused by surgery has been suggested as a cause for the malignant transformation (Arotiba et al 2005), it is thought to arise from remnants of dental lamina, enamel organ or epithelial lining of odontogenic cysts (Avon et al 2003). Despite the paucity of the reported cases but certain patterns could be demonstrated; the mean age of occurrence was found to be 52 years, with male predilection, also the posterior mandible was the favored site (Corio et al 1987, Akrish et al 2007). The history and the clinical presentation of our case indicate that the lesion arose from a preexisting ameloblastoma. The treatment provided for our pa-
tient included resection of the mandible with neck dissection and postoperative radiotherapy; it is similar to that reported in the majority of reports, which is regarded as the treatment of choice (Bruce and Jackson 1991). Other modalities of treatment like primary external radiotherapy (Philip et al 2005), carbon ion therapy (Jensen et al 2011) and the use of Gamma knife stereotactic radiosurgery (Perera et al 2013) have been reported. The recurrence of the reported cases ranges between 15%-25% after wide surgical excision and more than 90% after local curettage. The reported survival rate ranges from 3 months to 5 years. (Ponnam et al 2012, Keerthi et al 2013).

Although the histological diagnosis is not an easy task for the pathologist (George and Bishen 2011), the main histological features that suggest the diagnosis of ameloblastic carcinoma include; cellular features of ameloblastoma namely; follicular pattern, peripheral palisading, reverse polarity, cellular atypia, increased mitotic figures, necrosis, and the presence of clear cells (Dhir et al 2003, Mubeen et al 2007, Angiero et al 2008). This histological picture, which is seen in the present case, is consistent with Ameloblastic Carcinoma rather than intraosseous squamous cell carcinoma as the sections reveal a follicular pattern resembling odontogenic tissue with no evidence of dysplastic surface epithelium.

In conclusion, ameloblastic carcinoma is a rare tumor of unclear pathogenesis, but some hypotheses have been set forth. There is no standard treatment protocol for this lesion but radical surgical excision with or without radiotherapy is reported in the majority of cases. The histopathological diagnosis of this tumor is far from being straightforward and certain features must be present to make the proper diagnosis.

References


