

Ameloblastic Carcinoma of Maxillary Sinus

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ABSTRACT

Ameloblastic carcinoma is a rare malignant lesion with characteristic histopathologic feature, while sometime this feature is vague, tumoral cells appear such as ameloblastoma but they show cytologic atypia. In radiographic appearance behave similar to typical ameloblastoma. It may present as a cystic lesion with benign clinical features or as a large, ulceration, significant bone destruction and loosened tooth. Lesion often found after an incisional biopsy unexpectedly. Because of their aggressive behavior (extension to surrounding soft tissue, extensive bone destruction, lymph node involvement and metastasis) lesion require wide excision is treatment of choice. Regional lymph node dissection, radiotherapy and chemotherapy have restricted value for treatment. Close periodic evaluation of patient is obligatory. We report a case of ameloblastic carcinoma of maxilla in 46-year-old male patient, including radiographic, clinical, pathologic features. Clinical and radiographic view mimic ameloblastoma, while in histologic sections revealed cytologic atypia associated by ameloblastic carcinoma. Patient scheduled for preoperative radiotherapy before radical surgery.

KEYWORDS: Ameloblastic Carcinoma, Ameloblastoma, Maxilla, Case Report

INTRODUCTION

Tumor of lower orofacial region may be benign or malignant. Most primary malignant lesion concludes with sarcoma, carcinoma of salivary glands, especially squamous cell carcinoma and melanomas. Breast, lung, abdominal organs and prostate cancers can metastasize to lower face structural anatomy.^{1,2} Benign tumor may have odontogenic or non-odontogenic source, ameloblastoma reveal 1% of all jaw tumor.^{3,4} Ameloblastomas are sub classified as a common odontogenic tumor with significant clinical view.⁵ Ameloblastoma is an odontogenic tumor that arises from dental embryonic remnant such as epithelium of cyst, dental lamina or enamel organ.⁶ In 1984 Slootweg And Muller suggested a classification system for malignant tumor with feature of ameloblastoma: type 1: primary intraosseous carcinoma ex odontogenic cyst type 2:a: malignant ameloblastoma b: ameloblastic carcinoma, arising de novo, ex ameloblastoma or ex odontogenic cyst type 3: PIOC arising de novo.⁶ Carcinoma show the presence of both histopathologic feature of ameloblastoma and carcinoma. Tumor may metastasize, histologic features of malignancy may be observed in both primary tumor and metastasize.⁴ Malignant ameloblastoma convey lesion that metastasizes, however both metastatic lesion and primary tumor have benign histologic feature.^{6,3} The incidence of carcinoma is greater than malignant ameloblastoma.⁶ Ameloblastic carcinoma is a rare odontogenic epithelium malignant tumor, we have reported 70 cases from 1984-2011.⁴ When revealed as an aggressive appearance, it may be diagnosed as a malignant tumor, but in cases without an aggressive

feature, it is difficult to distinguish ameloblastic carcinoma from ameloblastoma.⁷ In clinical view it may mimic ameloblastoma, hard expansile mass, with displaced and mobile tooth and normal overlying mucosa, however it may be seen as rapid growing mass, ulceration, bleeding, fistula, pain and mobility. Tumor metastasizes to lung in most cases.³ Despite area that resemble ameloblastoma, ameloblastic carcinoma shows changed patterns and cytologic view. The presence of sheets, island, or trabeculae of epithelium and rare presence of stellate reticulum-like lesion should alert the pathologist for possibility of ameloblastic carcinoma. Round to spindle shaped epithelial cells with no differentiation suggest malignant process.⁸ Other features such as high mitotic index, necrosis, neural and vascular invasion, cytological atypia calcification and hyperchromatism observed too.⁸ Presence of many clear cells suggest ameloblastic carcinoma (figure 1A,B).⁸ In radiographic view represented well-defined, maybe corticated even scalloped border radiolucency, usually have unilocular appearance, maybe multilocular (soap bubble and honey comb). It have predominancy in mandible in molar and premolar region. Sometime lesion destroy cortical border and invade to adjacent soft tissue. Effect in surrounding structure consist of tooth bodily displacement, root resorption similar to benign tumor, lamina dura and cortical boundary destruction, mandibular canal displacement even erosion. Sign of osseous destruction similar to ameloblastoma can be found in ameloblastic carcinoma, these lytic phenomena may be assessed by CT and MRI imaging (figure

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2A,B).¹⁰ Main differential diagnosis ameloblastoma, odontogenic keratocyst, odontogenic myxoma, central muco-epidermoid carcinoma (indistinguishable in radiography), carcinoma in dental cyst and central giant cell tumor.³ Wide local excision is treatment choice, regional lymph node dissection performed too.⁶ radiotherapy and chemotherapy have limited value, low sensitivity, due to low sensitivity prognosis is poor.^{3,4,6} Certain authors have suggested surgery plus radiotherapy, while other have doubt about these combination. There are few report on chemotherapy effect, but preoperative radiotherapy suggested to diminish tumor size.⁹ Tumor are prone to recur that justify a long follow up.¹⁰ This study reports a case of ameloblastic carcinoma. We present clinical, radiographic and histopathologic feature of ameloblastic carcinoma.

CASE REPORT

A 46 year old male was referred to Oral and Maxillofacial Surgery Center, Isfahan University, Faculty of Dentistry, Isfahan, Iran for evaluating a rapid growing swelling. Intraoral examination revealed lesion in right hard palate with smooth and regular contour, in alveolar mucosa of left maxilla and in right vestibule with papillomatosis shape. Patient had a history of two surgeries (Mucoepidermoid Carcinoma). No enlarged lymph node were palpable. Plain radiography and CT scans were performed. In coronal CT (soft tissue algorithm) a large expansible corticated radiolucent lesion in right side of maxilla was found. (Fig No.1).

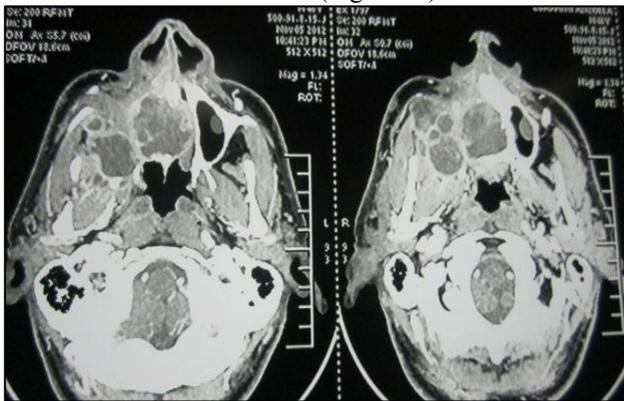


Fig No.1: CT Scan View

Lesion occupied a large portion of right maxillary sinus (radiographic report suggested sinus was filled with soft tissue), lesion involved right orbit (with destruction of orbital floor), right maxillary alveolus, right and left nasal cavity. Destruction of all maxillary sinus wall was seen. Lesion crossed the midline. Right OMC was closed, but left OMC was opened. Nasal septum was involved too. Lesion has extended posteriorly until nasopharynx and pterygoid plate. In axial CT (soft tissue level) a large corticated lucency in right maxillary side with crossing the midline was seen (Fig No.2). Facial asymmetry was observed too. Zygomatic bone was intact. Left maxillary sinus retention cyst was seen. Lesion had multilocular appearance in some areas. Based on these finding the diagnosis was recurrence of a tumor in right maxillary sinus, with aggressive behaviour. Diagnoses of ameloblastoma involving the right maxilla was made. Histopathologic view showed fragmentation of ameloblastic odontogenic epithelium occasionally arranged in palisades. The most central cells were arranged more loosely resembling the stellate reticulum. (Fig No 3,4). Metaplasia also was noted. Epithelial cell showed neoplastic proliferation. Certain part of sample resembled feature of ameloblastoma, however the cytology of some part confirmed ameloblastic carcinoma. Patient was scheduled for surgery, including right maxillectomy and radiotherapy. The patient was followed every three months. After one year there was no evidence of recurrence.

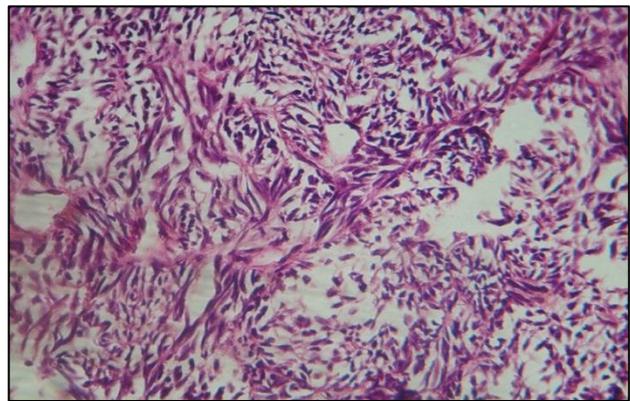


Fig No.3: Histopathological Picture



Fig No.2: CT Scan View

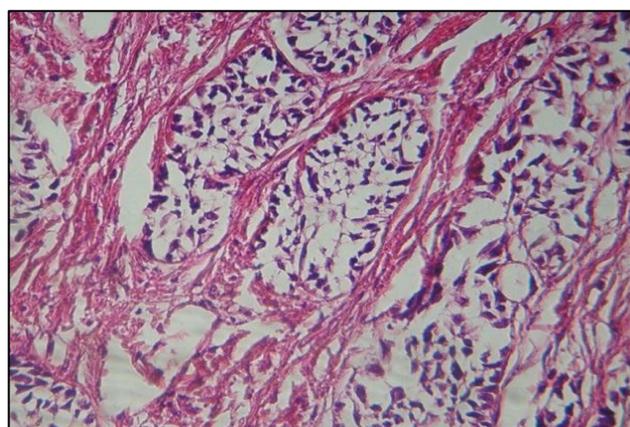


Fig No.4: Histopathological Picture

DISCUSSION

Ameloblastic carcinoma is a rare, malignant neoplasm with poor prognosis and predominancy in mandible. The most common symptom is a rapidly processing, painful swelling.^{10,11,12} Clinically, these lesions are more aggressive than typical ameloblastoma. Perforation of cortical plate, invasion to adjacent soft tissue, recurrent lesion, metastasis usually to cervical lymph node, are associated with ameloblastic carcinoma.¹⁰ In this case cervical lymph nodes were non palpable. The patient had no metastatic lesion at the time of diagnosis. In this case carcinoma occurred in right maxilla, extending across to left side. While one third originate in maxilla, most cases involve mandible.^{3,10,11,12} The male to female ratio is 5:3 with majority of cases occurring in patient aged 50-60.¹³ The present case involved a 46-year-old male. The patient in this case presented facial asymmetry, rapid growth, pain. The radiographic appearance of ameloblastic carcinoma was similar to ameloblastoma. In majority of cases a radiolucent intraosseous lesion is revealed^{9,10} as was showed in this case. In this case radiographic appearance was consistent with ameloblastoma. Differential diagnosis was carcinoma arising in the lining of an odontogenic cyst. The epithelium of squamous odontogenic tumor lacked any cytological evidence of malignant disease.⁶ Basal cell carcinoma, primary intra-alveolar epidermoid carcinoma had to be considered.⁸ The term of ameloblastic carcinoma may be applied for this case with histologic feature of malignancy such as pleomorphism along with indisputable feature of typical ameloblastoma. It accepted that maxillary ameloblastoma should be treated as radically as possible due to spongy architecture of maxilla.¹⁴ In this case preoperative radiotherapy was started to decrease the tumor size prior to radical surgery. A systematic evaluation of the chest by periodic imaging is recommended due to lung metastasis.¹⁵ We had no metastatic report in this case.

CONCLUSION

Ameloblastic carcinoma is a very rare malignant odontogenic tumor with characteristic histopathologic feature. Diagnoses at early stage and close periodic evaluation for metastasis and potential to pulmonary involvement are necessary

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