Ameloblastic carcinoma- a case report
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Abstract
The ameloblastoma is the prototype of a benign neoplasm with odontogenic origin. It will exhibit cell replication and growth throughout its existence, but it will not metastasize. The rare cases of ameloblastoma that have metastasized are termed malignant ameloblastoma. Alternatively ameloblastic carcinoma represents a malignant tumour that bears a histologic resemblance to an ameloblastoma. Here we present a 55 year old male with a longstanding ameloblastoma of the mandible which turns into carcinoma.

Key Words: ameloblastoma; malignant; carcinoma

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Introduction
Ameloblastoma are locally invasive benign tumours of odontogenic origin with a high chance of local recurrence. It will therefore become progressively destructive if left untreated. The invasive ameloblastoma that occurs centrally in either jaw and will usually present as an asymptomatic expansion. Malignant ameloblastoma distinguishes from ameloblastoma by the presence of metastases. It shows the histopathology features of ameloblastoma, both in the primary tumour and in the metastatic deposits. The ameloblastic carcinoma has cytologic features of malignancy not only in a primary tumour but also in a recurrence or in any metastatic deposits. These lesions are locally aggressive, but metastasis does not necessarily occur(1).

In general these tumours are non-capsulated, infiltrating neoplasm, although sometimes there may be areas that appear well demarcated. According to W.H.O two sub types of ameloblastic carcinoma are recognized as primary type and secondary type. The secondary type of ameloblastic carcinoma has arisen from a pre-existing benign ameloblastoma. Here we are discussing a case report of untreated ameloblastoma which turned into ameloblastic carcinoma.

Case Report
The patient reported to the department of oral and maxillofacial surgery, for the first time in March 2007 at the age of 55 years. On examination, the presentation was that of a firm bony expansible mass from the premolar area to opposite premolar area, more prominent in lingual aspect of anterior mandible. The duration of swelling was six months and intermittent pain with frequent pus discharge. The panoramic radiograph revealed a well-defined radiolucency extending from 34 to 44 regions(figure 1). The biopsy was taken elsewhere and the report showed the lesion to be an ameloblastoma of the mandible. The report also showed focal squamous metaplasia. We planned for segmental resection and reconstruction with rib graft. But the patient did not turn up. The patient again reported in January 2009. This time clinical presentation was extra-oral swelling extending from right parasymphyseal region to left second premolar region with multiple draining sinuses. Mobility of segments was also noticed. The panoramic radiograph showed well defined radiolucency lesion with scalloped margin extending bilaterally from 36 to 46 region. Pathological fracture of mandible was also noticed. This time, hemimandibulectomy was performed along with the excision of perforated soft tissue. During this procedure, bilaterally enlarged submandibular lymph nodes, firm in consistency and 1cm in diameter were removed. The mandibular reconstruction was done with reconstruction plate.

Figure 1 Orthopantomogram

Histopathology report of specimen showed as dysplastic stratified squamous epithelium infiltrating into the underlying connective tissue stoma. Epithelium shows hyperchromatism, vesicular nucleus and abnormal mitotic figures. Vascularity and chronic inflammatory cell infiltrate were moderate. Lymph node showed no lesional tissue. Peripheral bony margins were devoid of any lesion tissues and suggestive of moderately differentiated squamous cell carcinoma. Patient was advised for radiation and long term periodical follow up.
Discusson

Naagai et al(2) divide the malignant tumours with features of ameloblastoma into two groups: 1) malignant ameloblastoma 2) ameloblastic carcinoma. Malignant ameloblastoma is an ameloblastoma with a benign and typical histological pattern which metastasis. Ameloblastic carcinoma is any ameloblastoma in which there is primary or recurrent evidence of malignancy regardless of metastasis(2).

Ameloblastic carcinoma is extremely rare, only 2% of cases were reported. In this case initially some confusion was occurred as to the cellular origin of the tumour. This created some diagnostic difficulty. However, thorough examination of first and second specimens by eminent pathologists, given as the tumour was most emphatically a plexiform histologic type of ameloblastoma which turned into carcinoma. Histological feature of malignancy is rare in ameloblastoma and in this case is quite unusual because such features were not evident at initial diagnosis.

According to Philipson & Reichart, the mean age of ameloblastic carcinoma was 34.4 years, with a range of 5-74 years(3). The male: female ratio was 1:2:1. The survival rate of ameloblastoma if metastasis occurs ranges from 3 months to 5 years(4). Most malignant ameloblastoma were reported as plexiform histopathologic type (3, 5) In this case, it was reported as plexiform type of ameloblastoma. According to the literature, the interval between initial diagnosis and metastasis is with an average of 10-12 years (5, 6).

According to Houston, during embryogenesis the odontogenic epithelium becomes entrapped in lymphoid tissue. When this epithelium undergoes benign neoplastic changes, an ameloblastoma could develop within a lymph node(7). This theory could explain occurrence of ameloblastoma in cervical lymph nodes. Lymphatic spread of ameloblastoma is well accepted. In ameloblastoma, it had been proposed that the metastasis originated from aspiration of tumour cells rather than conventional hematogenous or lymphatic spread. As with many other cancers ameloblastoma is also metastasis with lungs. Chest radiograph is advisable for distant metastasis. Neck examination is also considered along with the radiographic examination.

Chemotherapy and radiation may play a role in inoperable cases. In regional lymph node involvement resection along with neck dissection is the treatment of choice.

Conclusion

In conclusion, a review of 1036 cases of ameloblastoma by Small et al(8), were reported that ameloblastic carcinoma originate from histologically normal ameloblastoma. In this case also ameloblastic carcinoma originated from apparently normal ameloblastoma.

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