CASE REPORT

Management of Large Mandibular Ameloblastoma - A Case Report

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Abstract

Ameloblastoma is a benign epithelial odontogenic tumour but is often aggressive and destructive, with capacity to attain great size, erode bone and invade adjacent structures. They are the commonest neoplasms affecting jaws, with a high rate of recurrence if not removed adequately. Its incidence, combined with its clinical behaviour, makes ameloblastoma the most significant odontogenic neoplasm of concern of oral and maxillofacial surgeons. The challenge in the management of this tumour is not only to excise the tumour completely in order to prevent recurrence but also to provide the best reconstruction method with the intention of giving reasonable cosmetic and functional outcome to the patient.


Keywords- Ameloblastoma, Odontogenic tumor, Hemimandibulectomy, Resection

Introduction

Ameloblastoma, a true neoplasm of odontogenic epithelial origin, derives its name from the English word “amel” which means enamel and the greek word “blastos” which means germ.1,2 Although it is considered a benign tumour, its clinical behaviour identifies it as a tumour of intermediate malignant potential which lies in the grey zone between benign and malignant neoplasm.3 It is characterized by slow but persistent growth and local infiltration, with a high rate of recurrence if not removed adequately.4

Until relatively recently, controversy existed regarding the management of benign ameloblastoma. However the elucidation of the biologic properties and aggressive clinical behaviour of the various histopathologic varients ofameloblastoma has resulted in a high rate of recurrence (50%-90%) when conservatively treated.5 Aggressive resection of this benign tumour is, therefore, considered the only predictable curative form of therapy. Here we report a case, of a large mandibular ameloblastoma extending to posterior maxilla and a discussion on its surgical management.
Case Report

A 40-year old male presented with the complaint of a huge painless swelling on the left side of the face since last eighteen months which progressively increased in size. The patient visited the clinician eight months after a noticeable swelling developed and was prescribed antibiotics, suspecting dento-alveolar infection. Then after, the patient was irregular in dental visit and hence unable to continue the treatment resulting in the gradual increase in size of the lesion till it reached the present size. The swelling was associated with paraesthesia of his left side of face mainly over the jaw and he also had difficulty in tongue movement and difficulty in chewing due to the lingual extension of the mass intraorally. The patient also gives history of extraction of an un-restorable and carious left mandibular second molar two years back.

His general physical status was normal. Extraorally, a large firm swelling measuring around 10×12 cm in size was seen on the left lower portion of the face which was bony hard and tender on palpation, extending, anteriorly from the left angle of mouth to the anterior border of sternocleidomastoid muscle posteriorly (Figure 1, 2). The skin over the swelling was normal in appearance. There was no enlarged lymph node over the neck region. Mouth opening was reduced to two finger breadth and introral examination revealed an exophytic lesion on the left side of the oral cavity that filled the entire oral cavity protruding over the tongue depressing the later resulting in restricted tongue movement (Figure 3).

Panoramic view of the patient showed a mixed radiolucent, radiopaque area in the left side of the mandible extending from the distal aspect of left mandibular first molar to left mandibular body, angle and ramus with the left maxillary tuberosity involvement. X-ray also revealed discontinuity along the lower border of left side of the mandible suggestive of pathological fracture (Figure 4).

A plain CT scan showed the internal characteristics and extent of the lesion. It revealed a solid, expansile, ill-defined, mixed radiodense and radiolucent areas (Figure 5). Based on the clinical history and physical examination along with radiological findings the lesion was provisionally diagnosed as ameloblastoma.

The incisional biopsy of the oral cavity mass was done under local anaesthesia with minimal bleeding and histopathological diagnose was given as follicular ameloblastoma.
In view of the extensive tumour involvement, a left hemimandibulectomy and left posterior maxillectomy via lip splitting incision was planned (Figure 6). Intraoperatively, after temporary tracheostomy was performed, the skin flap was raised and retracted laterally and superiorly exposing the mandible underneath. Hemimandibulectomy was performed across the midline and left hemimandible was removed en bloc with the tumour (Figure 7) and a left posterior maxillectomy was performed (Figure 8, 9). Post-operative period was uneventful and tracheostomy tube was decannulated on day six and sutures were removed on day seven post operatively. Post-operative histopathology was interpreted as follicular ameloblastoma.

The Patient had no significant complaint at his first follow up, one month later. There was small depression over his left cheek but the patient was not much concerned about it. Follow up at the end of one year revealed no recurrence and he had no difficulties in swallowing, chewing and also maintained good voice quality. He was happy with his present status and refused our advice to undergo reconstruction of the cheek defect.

Discussion

Ameloblastoma is a benign but aggressive neoplasm of odontogenic origin. However, no enamel or hard tissue is formed by the tumour cells. The term ameloblastoma is coined by Churchill in 1933 but, the first detailed description of this lesion was by Falkson in 1879. It represents about 1% of all oral ectodermal tumours and 9% of all odontogenic tumours. The lesion may arise most commonly from cell rests of enamel organ, epithelium of odontogenic cysts, disturbances of the developing enamel organ and basal cells of oral epithelium.

Clinical Presentation:

Ameloblastomas, though predominantly seen in the middle age group (4th – 5th decade of life), it is also known to occur in children (8.7% to 15%). It may appear anywhere in the jaws, although approximately 85% of the lesion arise in the mandible, especially in the molar ramus region. Typically ameloblastoma present as painless slow growing mass as in our case, it took about one and half year before he developed symptoms such as significant facial asymmetry, difficulty in chewing and deglutition. Around 13.3% of lesion presents with paraesthesia of the innervated region of mandible. This patient also had paraesthesia over the left cheek particularly over the distribution of mandibular division of trigeminal nerve.

Radiographic features

The radiographic presentation of ameloblastoma may vary from unilocular to multilocular pericoronal radiolucency often associated with impacted tooth. They characteristically exhibit slow but unrelenting and destructive growth. The multilocular radiolucency is found to be more common than the unilocular type. The implication of certain radiological presentation to the surgical management of ameloblastoma has been documented. The unilocular or multilocular lesions may require conservative or aggressive management depending on the presence or absence of cortication and root resorption. The presence of cortication means that the marginal and/or adjacent bone to the lesion may be normal and therefore, the further 1 cm is expected to be actually normal bone – hence the need for less aggressive treatment. Whereas, the less well corticated lesions do not have the certainty of
normal adjacent bone and as such would require more aggressive management in terms of the amount of normal bone to be taken along with the lesion.\textsuperscript{11}

**Management**

Ameloblastoma was well known for its high recurrence rate if excision was incomplete. Inadequate diagnosis and treatment of this lesion although followed by initial healing, may lead to a recurrence years later, but with more aggressive behaviours. Therefore the treatment of choice is surgical excision with wide free margins, if not resection but, the growth characteristics of ameloblastoma vary from other benign lesions and tumour resection is often advocated regardless of the type of ameloblastoma.\textsuperscript{6,7} The appropriate amount of normal bone beyond the radiographic boundary required for a tumour-free margin has not been definitely established. Some clinician suggest a resection of no less than 1-2 cm of normal appearing bone beyond the radiographic tumour margin.\textsuperscript{11} When tumour has perforated bone, removal of adjacent soft tissue extending to the next adjacent anatomic boundary must be performed to ensure complete tumour-free soft tissue margins. Preservation of the inferior alveolar nerve is no prudent when it is involved by the tumour and it should be resected enbloc with the specimen.\textsuperscript{6,7,13} In our case the extensive lesion with decorticated margin along with discontinuity of lower border of mandible and its multilocular character directed us to plan for a hemimandibulectomy and a partial posterior segmental maxillectomy.

**Reconstruction**

The mandible is both functionally and cosmetically important structure of the head and neck region and hence reconstruction of large mandibular defects represents a challenge to head and neck reconstructive surgeon. Among the different methods of mandibular reconstruction for large defects, microvascular surgery has become the preferred option.\textsuperscript{14} Four donor sites i.e. fibula, iliac crest, radial forearm and scapula have become the primary source of vascularized bone and soft tissue for the oral reconstruction.\textsuperscript{5,14} In our case secondary surgery was planned for a reconstruction but the patient was happy with his appearance and refused to undergo the surgery.

**Recurrence**

Ameloblastoma is a tumour with a well-known propensity to recur due to its capacity to infiltrate trabecular bone.\textsuperscript{5} Several factors have been identified influencing the rate of recurrence. The most important factor is the clinicopathologic variant of the tumour. It is generally accepted that there are three variants of the benign ameloblastoma, designated as solid or multicystic, unicystic and peripheral.\textsuperscript{4} The solid variety has high propensity for local infiltration and therefore the highest potential for recurrence.\textsuperscript{5,15} It erodes the cortical bone and also spread in to the medullary space well beyond radiographic margins\textsuperscript{16} resulting in recurrence rate of 75-90\%,\textsuperscript{5} requiring more aggressive treatment. In contrast unicystic ameloblastoma are generally less aggressive in their growth characteristics resulting in far lower (approximately 15\%) rate of recurrence even when treated by conservative means. The second factor influencing the rate of recurrence is the anatomic site. The dense cortical bone of mandible prevents the tumour from spreading extensively for several years, although spread in the central cancellous bone is beyond the radiographic margins of the tumour.\textsuperscript{15} Adequate surgical margins are relatively easy to obtain in such cases. In contrast, the thin bone of maxilla provides a poor barrier to spread and the potential for inadequate resection is great increasing the recurrence rate five times than that of mandibular tumours. Third factor that should be considered is the adequacy of surgery. Conservative treatment has found to result in more recurrence rate that aggressive treatment.\textsuperscript{7} Finally, the histologic variant of the ameloblastoma has been suggested to be of prognostic significant in terms of recurrence. Certain histologic variants has been thought to behave more aggressively with a greater tendency to metastasize.\textsuperscript{16} Several authors contradict this statement, because no correlation has been found between histologic type and clinical behaviours.\textsuperscript{4} Consensus is that histologic variant should not modify treatment.
Conclusion

The challenge in the management of large ameloblastoma of the mandible is not only to excise the tumour completely in order to prevent recurrence but also to provide the best reconstruction method. Hence it is most emphasize, to both the clinician and the patient, the need for a definite treatment protocol and lifetime periodic follow-up for detection of recurrence as even a five year tumour-free period does not necessarily mean a cure.

References