JCMCTA 2011; 22 (1) : 39-42

MANAGEMENT OF LARGE MANDIBULAR AMELOBLASTOMA : A CASE REPORT

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Summary

Ameloblastoma is painless and slow growing true neoplasm of enamel organ. This may occur either in mandible or maxilla but most common site is posterior part of mandible and ramus. Diagnosis depends on clinical features, radiograph and histology. Management and reconstruction of this tumor is to be done by wide excision with healthy tissue. Secondly the bony & soft tissue defect is to reconstructed for cosmetic and functional outcome of the patient.

Key words

Ameloblastoma; hemimandibulectomy; disarticulation

Introduction

An ameloblastoma is the odontogenic tumor of epithelial origin and locally aggressive. At first recognized by Guzack in 1826.Then Falkson established it in 1879 . The lesion was further studied in 1885 by Malassez, who called it an adamantinoma[1] The term ameloblastoma was coined by Ivy and Churchill in 1933[2]. Ameloblastoma may develop at any age, but its peak incidence (20 to 40 years of age)[3]. About 85% of ameloblastomas occur in the mandible, most of them are found in the molar and ascending ramus[4]. An ameloblastoma is typically asymptomatic, slow growing and therefore seldom diagnosed in the early stage of development. Remaining undiscovered until the tumour produce intra and extra oral jaws swelling. . It is clinically hard, non tender while it becomes large in size it causes discomfort, malocclusion,mobility teeth, paresthesia& loss of function .It also develops facial asymtry[1]. Radiologically in most cases ameloblastomas present either unilocular or multilocular radiolucency with honeycomb or soap bubble appearance and moderately well defined margin.

Resorption of the apex of the roots and teeth displacement are common feature. Buccal & lingual cortical bone expansion is seen[5]. It may solid or cystic.Sometimes it may combine. Ameloblastoma is similar to the enamel organ.

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It is composed of nest, strands & cords of ameloblastic epithelial cell, separated by relatively small amount of fibrous connective tissue stroma. The cells are cuboidal or columnar type whose nucleoli are generally well polarized .The .Histologic pattern of the ameloblastoma are numerous types.They are follicular, plexiform, acanthomatus,baal cell and granular pattern.In addition unicystic lesion has been reported[6]. In developed countries, ameloblastoma is usually treated early, so these tumors do not normally reach sufficient size to qualify as a huge enlarge ameloblastoma. This case is reported because of its rarity, size, and management.

Case report

A 22 years old man presenting with a huge swelling on right side of the face for 12 years visited in oral and maxillofacial surgery department of Chittagong Medial College and Hospital. He had a huge swelling on right side of face. Presenting history revealed that swelling was slow growing over a period of 12 years. It was painless and discomfort initially. His swelling distorted facial contour due to huge enlargement. Recently developed a discharging point over the swelling .Now he faced difficulty in mastication & swallowing. Horizontally swelling extended from left angle of mouth to right in front of sternocledoid muscle & vertically from upper border of zygomatic arch to cervical margin of neck . Tumour was 18 cm/16 cm.in size (Fig-1) The skin of overlying mass was normal in colour but numerous dilated superficial veins were present. Overlying mucosa of the tumour was smooth & normal in colour. Introral huge growth was seen through mouth opening.Teeth of the affected side were mobile & divergent crowns. On palpation swelling was hard & some parts were soft & crackle in nature.There was no enlarging lymhnode in the neck region.

Fig 1 : Pre operative photogragph

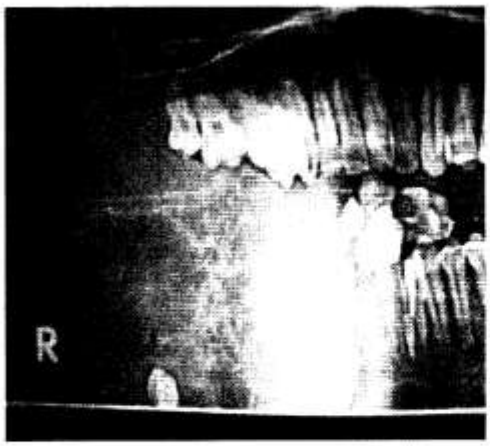

Fig 2 : Orthopantomogram

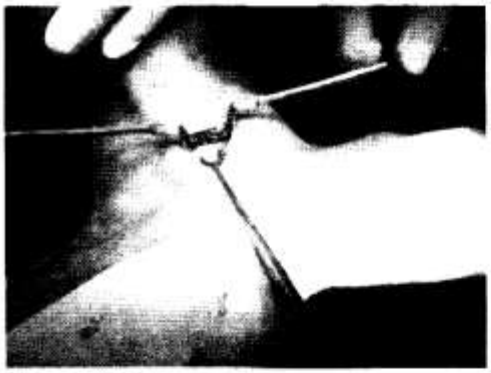

Fig 3 : Photograph of tracheostomy

Fig 4 : Peroparative Photograph

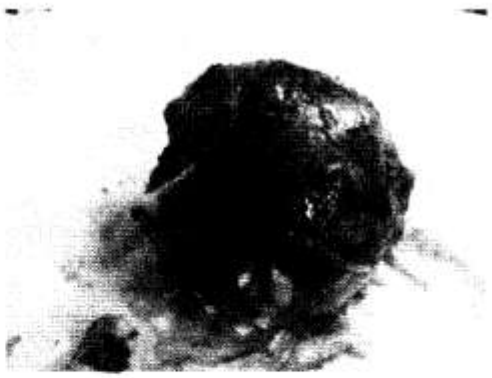

Fig 5 : Excised tumour

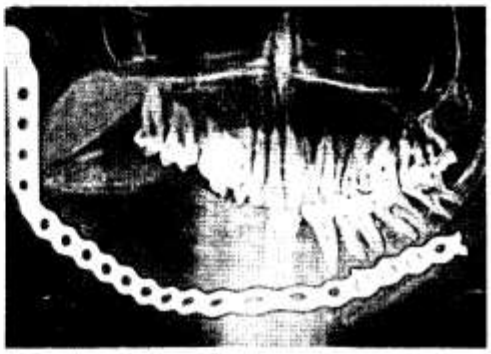

Fig 6 : Reconsnstruction plate

Radiological investigation included lateral view of face& orthopentomogram revealed a diffuse,ill defined margin and large radiopaque lesion with fine radiolucent area producing a honeycomb appearance.Area extended from left premolar region of mandible to right condyle & coronoid process. Roots resorbtion were present. Teeth were malaliged (Fig-2). Patient's presenting history, physical examination& radiograph pointed that tumour might be ameloblastoma But confirm diagnosed was done by histopathological examination. Pre-operative histological examination revealed plexiform type ameloblastoma, consist of thin trabeculae of darkly staining epithelial cells in a sparsely cellular connective tissue stroma.Epithelial components showed peripheral columner cell. Investigation reports revealed all were in normal limit, Hb% 13.0gm/dl, RBS 109mg/dl,Serum urea 24/dl,Serum creatinine 0.7mg/dl,ESR 40 mm 1st hour and TC WBC 9500/cu.Tracheostomy was done for good intubation (Fig-3). Treatment plan was made of hemimandibulectomy with disarticulation via lip splitting (Fig-4).

The rest part of mandible & soft tissue was stabilized by fixation of stainless steel reconstruction plate (Fig-6). Closed the wound in layer by layer (Fig-7). The excised tumour was 16/14cm in size include part of body of mandible, ramus & right coronoid process (Fig-5). Macroscopically mass was reddish soft tissue, teeth were embedded in mass but dark fluid was present in small part of tumour mass .On palpation the mass was firm in consistency. The specimen was sent for histopathlogical examination & report revealed that tumour was ameloblastoma. Nasogastic feeding tube was given postoperatively for better feeding. & maintain oral hygiene. Soft diet had been given orally at 7th day.Decanulation of tracheal tube was started at 6th post operative day. Tube was completely removed at 7th post operative day. Stitched has been removed at 9th post operative day. Follow up the patient, he has been found after eight month later a shun ken right face& scaring at the mid face. Now he felt better& also happy (Fig-8).

Discussion

Ameloblastoma is benign tumor of enamel type tissue origin that occurs either in jaws and usually asymptomatic. It expands buccal and lingual cortical plate of mandible and also invaded soft tissue. It has been reported that all age group may be affected and it is rare to seeing under 12 years but it peak incidence is in between 20 to 30 years age. There is no gender predilection. Mandible is more frequently affected 75% than maxilla 25%[6].The most common sites are third molar region of either jaw and ramus .It can be developed to a huge large size and causes facial deformity, malocclusion,It is usally painless when infected cause pain. Crowns of the teeth are divergent Radio logically ameloblastoma appears radiolucent that may be unilocular or multiloccular like soap bubble or honeycomb appearance[7]. The ameloblastoma is closely resemble the enamel organ. Different type of ameloblastoma are commonly found The most frequent pattern is follicular ameloblastoma. Epithelial islands are present within a variable quantity of fibrous stroma.The epithelial islands are mimic the enamel organ. The periphery is lined by columnar type cell, resemble preameloblast. The second major pattern of ameloblastoma is plexiform. The epitheliam proliferate in a network anastomosing strands the epithelial components are columnar cell with nuclei polarized away from the basement membrane toward the central stellate reticulum.

Incase acanthomatous ameloblastomam, within stellate reticulum, a variety of change may observed.Squamous metaplasia is seen.Sometimes keratin formation in the central portion of the tumour island is noticed.Occasionally, the cells within stellate reticulum are large and contain granular eosinophillic cytoplasm.This variant is called granular ameloblastoma composed of small discrete island of tumor. Peripheral cell layer is cuboidal or columnar.Nucleulous are generally well polarized .In flexiform type tumour ameloblast like cells are arranged as a network or interconnecting strands of cells. Each of these masses or strands is bounded by a layer of columnar cell.. Basal cell type of ameloblastoma bears considerable

Fig 7 : Photograph of finishing stage

Fig 8 : Photograph after eight month

resembles to the basal cell carcinoma of the skin.Plexiform unicystic ameloblastoma is termed to designate a plexiform type of epithelial proliferation occurring in dentigerous cyst[4].Further more the cystic lining can be trans formed to the ameloblastoma [6,8] A number of treatment modalities have been proposed in the treatment of ameloblastoma, like wide excision, curettage, enucleation, cryotherapy cautery laser usage, radiotherapy & chemotherapy[9].

Enucleation & curettage has been reported high recurrence rate[10].Best results have been obtained by wide surgical excision with a 10-20 mm margin of normal tissue[11]. Cryo therapy has not been proved as effective treatment regime. Chemo & radio combination therapy has been proved more successes full[12].Hemimandibulectomy is through lip splitting incision is good approach to give a better expose for complete removal of huge ameloblastoma. It has been reported that use a less invasive incision to avoid troublesome outcome of the lip splitting [13].They utilized risdon and intraoral incision which give less post operative morbidity and cosmetic out come[14].In the review of 60 mandibular ameloblastoma cases have been shown that there was no recurrence of these cases treated via enbloc resection as comparison to enucleation & curettage[15]. Reconstruction: Mandible is both functionally & cosmetically important part of head neck region contributing to facial appearance, speech, mastication and swallowing. So reconstruction of large mandibular defect is essential part of surgery but it is a challenging work.There are different method of mandibular reconstruction for large defect that have been described in different literature. Micro vascular bone grafts & free bone graft is usually done. Donor sites are iliac crest, radial forearm, and scapula. Some authors have done a comparative study between vascularised free fibular bone graft and iliac flap[16].It has been reported that reconstruction of surgical defect with free autogenous bone graft or vascularised autogenous bone graft which is carried out to obtain prosthetic restoration by means endogenous implant[17]. Recently developed another method of reconstruction is internal distraction osteogenesis has been popularized & they achieved successful distraction in eight patient& two patient were failed[18]. Researchers are now looking at other latest technique bio implant containing BMP-7 is being used in method of reconstruction. The resection defect was spanned with rigid reconstruction plate to hold the remaining mandibular segment in correct position. The defect was filled with bioimplant containing bone morphogenic protein-7 in a dematerialized bone matrix Hemimandibulectomy with disarticulation was done to remove massive tumour to reduce the chance of recurrence. A reconstruction plate was fixed to hold the soft tissue & rest part of mandible.

Conclusion

Surgical resection is an effective therapy in treating large mandibular ameloblastoma

Disclosure

All the authors declared no competing interestes.

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