Conservative approach for an ameloblastoma in an adolescent patient who did not accept resection therapy: A case report and review of litratures

Keywords: Ameloblastoma, Reconstructive tumor surgery, Conservative ameloblastoma treatment

#### Abstract

Ameloblastoma is the most common epithelial odontogenic tumor after odontoma in oral and maxillofacial region with invasive and benign histopathologic appearance but clinical behavior, Clinicopathologic variant, location, age, sex and availability to close long-term follow up of patient must be assessed before deciding treatment. Several surgical techniques described for treating ameloblastoma; Enucleation alone with peripheral or ostectomy/curettage, cauterization, marginal resection with keeping of mandibular lower border and radical resection with taking about 1-2 cm of uninvolved bone. In recurrent cases, facial deformity and swelling are most common symptoms. Some cited that extensive resection of the mandible in all ameloblastoma of children may be too radical and will make concern about the deformity, dysfunction, influence on facial growth, shape asymmetry, masticator dysfunction, and psychological impact.

## Introduction

Ameloblastoma is a slowly developing epithelial odontogenic tumor that first described by Koning in 1825. It is most common local aggressive benign tumor with a potential for recurrence if treatment was not enough. A few cases of malignancy noted and distant metastasis in case reports <sup>1</sup>. The screening panoramic view of jaws that is taken by practitioners becomes one of the primary steps, which are more common for detecting such pathologies.

Solids/multicystics are most common variant but unicystics and peripherals ameloblastoma are less common. Current opinions spotting those multicystics and solid types are locally aggressive with high recurrence rate if treated locally with enucleation or curettage. Unicystic ameloblastoma has less aggressive characteristics and may treat with enucleation and/or curettage <sup>2,3</sup>.

There is a serious attention for saving the quality of life following resection of such pathologies regarding feeding, speech, appearance and saliva control as lack of tissue support in the face <sup>4</sup>. Some authors advocating conservative therapy whereas the others persist of radical and absolute resection of involved jaw with extremely safe borders due to mean 4.5 mm histological extensions of tumor beyond its radiologic appearance<sup>2</sup>.

#### **Case presentation**

In July 2010, A 16 years old otherwise healthy, white female referred by a dentist whom recommended get a panoramic for evaluation of intra-oral swelling in right mandibular body plan.

We noted a slight expansion and asymmetry in right aspect of the face regard to mild mandibular swelling in angle and body area. Not any abnormal sensation nor any dynamic abnormality were detected in primary evaluation of patient. In Intra-oral exam, we noted absence of right mandibular third molar and slight hard expansion of buccal cortex of this area to the anterior portion of the right buccal vestibule was seen. The covering mucosa of this site was neither ulcerated nor erosive. Right mandibular premolars are not aligning well and had some degree of buccal and lingual drift. In existing panoramic, a unilateral, single, multilocular and well-defined radiolucent lesion extended from upper one third of ascending ramus to parasymphysis of same side involving a far displaced impacted third molar was dazzling. (Fig.1) Effects on roots of the first and second molar as resorption of them and some degree of divergency in premolar area also noted. Other structures of the ipsilateral side of mandible and total maxillary skeleton were normal. A cone beam CT showed  $25.7 \times 56.4$  mm radiolucent expanded lesion in greatest dimensions with thin buccal ,lingual and lower border cortex (Fig.2 a,b). Differential diagnoses for this lesion were dentigerous cyst, ameloblastoma and odontogenic keratocyst.



Fig. 1 Large multilocular radiolucency in panoramic view



**Fig.** 2 - a: Large expanded radiolucency in cbct view. **b**: thin cortical bone and impacted third molar is seen in cbct view.

Aspiration of site was not positive. Incisional biopsy pathology report showed ameloblastoma (plexiform type ). Regarded to literatures or reliable reference texts, this tumor due to its extensions should be treating by partial or marginal resection of the mandible(**5,6**)But due to disabilities and deformities those will be make the quality of life for this patient worse and of course because there were not any acceptance for wide resection onside her parents nor her, we decided enucleating this lesion with adequate curettage and informing the patient for close observation needed to follow-up.In surgical procedure cortical perforation did not detect as previously noted in cone beam CT (CBCT) .Lesion removed entirely and complete peripheral ostectomy was done(Fig.3). The surrounding bones of the lesion did seem to be intact and lower border of mandible also extracted due to involvement of roots in the lesion. Inferior alveolar nerve was not visualized, had cortical sheath, and did not seem to be involved. After irrigation with diluted hydrogen peroxide, surgical mesh smeary with tetracycline ointment placed at the site, secured, and scheduled to gradual removal in next appointments .



a b c

*Fig 3–a:* Cavity made by removing tumor mass, complete peripheral ostectomy was done *b:tumor bulge*, lesion removed entirely *c:* surgical mesh smeary with tetracycline ointment placed at the site

Excisional biopsy pathology report showed ameloblastoma (plexiform type ) and all surgical margins of tumor free of tumor .

On the follow up, radiographies opaque bone formation in matrix of bony container of site seen gradually in 3 months period for the first year postoperatively (Fig 4).



b



С

Fig. 4 -a: 2 months after surgery b: 8 month after surgery c: 12 months after surgery

Next Incisional biopsy performed after 6 months that did not show any ameloblastic component. In recall appointment for follow up we noted that the formation of bone in the right inferior border of mandible. We planned to restore the Mandibular missed teeth for patient by her demand and also need for having occlusion on site and prevention of occlusal plan complication. With the advent of Prosthodontics consultation, She underwent implant insertion in site after getting incisional biopsy and also shaving of right inferior border. Then three Intra-Lock implant were scheduled to insertion in sites for restoration first premolar, first and second molar and second premolar. Sites prepared and implants inserted with good primary stability. Bone consistency was good in palpation and inspection. Patient followed up for about 1 year after implantation. At the time of uncovering, excellent osteointegration clinically noted and observed in radiography. Also complete bone submerging noted in distal implant and a dense cortical bone has been covered the cover screw of implant (Fig.5).



*Fig. 5* – *Panoramic view just before uncovering,18 months after first surgery,6 months after implants insertion* 

# Discussion

In clinical impression and diagnosis of ameloblastoma, we encounter important and amazing results for clinical judgment. In a report by of 38 cases of ameloblastoma in young persons, 15 cases have dentigerous cyst impression, while only three cases were thought to be ameloblastoma and three susceptible between ameloblastoma and dentigerous cyst and the remains presented other patterns like OKC, myxoma and etc <sup>7</sup>. In some report mentioned that unicystic ameloblastoma and dentigerous cyst have a similar clinical and radiologic appearance <sup>8</sup> It will make us thinking that while ameloblastoma can personate as dentigerous cysts, more cares should taken to expanded dental follicle around the crown of impacted teeth rather than simply spotting it as a expanded follicle or dentigerous cyst.

So the patient and legally more important, her/his parents understand clean and clearly to make the decision not only through the opinion of surgeons or pathologists but also by themselves <sup>7</sup>. We think that recurrence is probably is not most important consideration for children, because we can observe them very well and detect any type of change in pattern of healing, and should not be keep in mind as equivalent to failure. Tümer reported a 24 years old female with plexiform ameloblastoma in right posterior mandible, treated with enucleation, curettage, and using allograft bone for reinforcing place (Tutoplast microchips). He followed her for 8 years and finally restored her missed tooth by an ITI implant and concluded that more conservative surgical enucleation with sufficient bone curettage and use of osseointegrated implants for prosthetic rehabilitation could be useful as predictable treatment of unicystic ameloblastoma. Becker and Wong also used five implants 2 years after removal of tumor<sup>8</sup>.

5 years follow up of implant rehabilitation by fibula graft and implantation by Giacomo Oteri et al. No peri-implantitis noted. Also very limited resorption of peri-implant bone was found at the end of the follow up time<sup>9</sup>. Case series done by Cheung et al for alveolar distraction following fibular reconstruction for implant restoration in four patients with Mandibular ameloblastoma. All implants had a minimal bone loss with good gingival health with 100% success rate <sup>10</sup>. Natashekar and et al rehabilitated 56-year-old male with distractor and dental implants following the resection of recurrent follicular unicystic ameloblastoma<sup>11</sup>. A large series of implants, 252 of them inserted in grafted bone and 454 in remnant bone were in 111 patients, 21 of them were ameloblastoma. Follow up period was 6 months to 9 years. Several different methods used to treat lesions. Of 706 implants 348(49.3%) were inserted in Mandibular reconstruction bony flaps <sup>12</sup>. As we see, all of above have some good results but also complications in post operation period. Using the other site of patient's body for harvesting bone, the disabilities and patient esthetic and functional concerns about them are the other materials that lead us to have another choice whenever we can treat the tumor more conservatively. In addition using of such appliances as distractor, pins, screws, plate fixations and hydroxyapatite could be increase overall cost and facilitates a good environment for bacterial reproduction. Some judge these

foreigners could compromise the competence of radiation therapies if indicated, that would be better rehash in a distinct topic.<sup>11</sup>

If any suspicious event found, checking every 6 months with radiography and biopsy should performed whenever the lesion increases even only a little in size, in an attempt to treat recurrence at the early stages <sup>7</sup>.

# Conclusion

Our findings consider being gentler for treatment of ameloblastoma in adolescent patient, suggesting an exact differential diagnosis, and using less aggressive procedure to decrease the extent of lesion and to get optimal specimens for serial section examination in condition we definitely ensure for follow-up not only by surgeon but also by patient and parents demand.

## References

1. Olaitan AA, Arole G, Adekeye EO. Recurrent ameloblastoma of the jaws. A follow-up study. *Int. J. Oral Maxillofac. Surg. 1998; 27: 456-460.* 

2. Takahashi K, Miyauchi K, Sato K Br. Treatment of Ameloblastoma in Children. J. Oral Maxillofac Surg 1998;36:453

3. Carlson ER, Marx RE. The Ameloblastoma: Primary, Curative Surgical Management. *J Oral Maxillofac Surg 2006; 64: 484-494* 

4. Chukwuneke FN, Ajuzieogu O, Chukwuka A, Okwuowulu T, Nnodi P, Oji C. Surgical challenges in the treatment of advanced cases of ameloblastoma in the developing world: The authors' experience. *Int. J. Oral Maxillofac. Surg. 2010; 39: 150–155.* 

5. Badal S. Management of Plexiform Ameloblastoma in a 12 year old female: A Case Report. WebmedCentral maxillofacial surgery. 2011;2(12):2593

6. Silva Toro JL, Ghersi MHD, Cabrera Gomez EA. Conservative treatment of the unicystic ameloblastoma: report of a case. *Int. J. of Oral Maxillofac Surg. 2011;7: 1171* 

7. Huang I-Y, Lai S-T, Chen C-H, Chen C-M, Wu H-W, Shen Y-H. Surgical Management of ameloblastoma in children. *Int. J. of Oral Maxillofac Surg.* 2007; 4: 478-485

8. Tümer C, Meral G. Unicystic Ameloblastoma: Implant-Supported Reconstruction and Long-Term Follow-up. J. of Hacettepe Faculty of Dentistry. 2007; 31: 49-53

9. Oteri G, Saverio DPF, Pisano M, Cicciù M. Five years follow-up of implant-prosthetic rehabilitation on a patient after mandibular ameloblastoma removal and ridge reconstruction by fibula graft and bone distraction. *Dent Res J (Isfahan). 2012 Mar-Apr; 9(2): 226–232.* 

10. Cheung LK, Chua HDP, Hariri F. Alveolar Distraction Osteogenesis for Dental Implant Rehabilitation Following Fibular Reconstruction: A Case Series. *J Oral Maxillofac Surg. 2013;* 71:255-271.

11. Natashekar M, Chowdhary R, Chandraker NK. Rehabilitation of recurrent unicystic ameloblastoma using distraction osteogenesis and dental implants. Niger J Clin Pract .2011;14:486-91

12. Cuesta-Gi M, Ochandiano CS, Riba-Garcí F. Oral Rehabilitation With Osseointegrated Implants in Oncologic Patients. *J Oral Maxillofac Surg.* 2009; 67:2485-2496.

