



UNICYSTIC AMELOBLASTOMA WITH MURAL PROLIFERATION: A SUCCESSFUL CONSERVATIVE APPROACH

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ABSTRACT

Unicystic ameloblastoma is one of the types of ameloblastomas, which is often asymptomatic, slow growing and locally aggressive. There are high rates of recurrence if the tumor is inadequately treated. Biological behavior of Ameloblastoma depends on histopathological variations which in turn affect treatment plan, and prognosis. Hence, there is difficulty in determining the most appropriate form of treatment for unicystic ameloblastoma. We present a case of unicystic Ameloblastoma with mural proliferation that occurred in the left posterior mandible of 25-year-old woman, which was treated successfully by conservative approach and did not recur till today.

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INTRODUCTION

Ameloblastoma is an odontogenic epithelial tumor that accounts for approximately 10% of Odontogenic tumors and 1% of all tumors and cysts of the jaws. (Small et al 1955) Its etiology, clinical behaviour, histologic patterns, and treatment modalities have been extensively discussed in the literature. (Sehdev et al 1974, Gardner et al 1980, Williams et al 1993, Feinberg et al 1996) Clinically, though ameloblastoma appears as an aggressive Odontogenic tumour, is often asymptomatic and slow growing, with no evidence of swelling. It can sometimes cause symptoms such as swelling, dental malocclusion, pain and paresthesia of the affected area. As it is slow growing, recurrences of ameloblastoma generally present many years and even decades after primary surgery. (Ferretti et al 2000) If inadequately treated, there is a possibility of developing into malignancy. (Becelli et al 2002) Three histological types (luminal, intraluminal, and mural) are recognized according to the degree of ameloblastomatous epithelial extension. This classification helps to decide biological behavior, treatment and prognosis. However,

deciding appropriate form of treatment of unicystic Ameloblastoma is quite difficult. (Garcia et al 2016) Treatment modalities for ameloblastoma can be have divided into conservative and radical therapies (Feinberg et al 1996) As it is slow growing and tendency to local invasion of bone and soft tissue, there are high rates of recurrence if the tumor is inadequately treated. Ameloblastomas have been treated with exclusive radiation therapy or in combination with chemotherapy (Gardner et al 1988, Atldnson et al 1994). Approximately 10–15% of all ameloblastoma occur in children and adolescents. The treatment of ameloblastoma in children and adolescents remains controversial as a more conservative approach, such as enucleation and curettage, has shown a high recurrence rate of 90% in the mandible and 100% in the maxilla and Radical approach which consists of partial and total maxillectomy or resections, with or without continuity defect in the mandible may lead to facial deformities. (Li et al 2012) Here is a case report of a 25yr old patient who was diagnosed with unicystic ameloblastoma of mural proliferation was treated conservatively with good result.

CASE REPORT

A 25yr old female patient reported to the Department of Oral and Maxillofacial Surgery with the chief complaint of mild discomfort in left lower back region of jaw since 4 months. Past medical, dental and family history of patient was non-contributory. There was no history of trauma, sinus opening or pus discharge. No signs of facial asymmetry were noted on extraoral examination. (Fig.1) Intraoral examination revealed missing left mandibular third molar with mild swelling in the region of 38. (Fig.2) On radiographic examination, OPG revealed a well defined, unilocular solitary radiolucency involving distal root of 36, 37, impacted 38 and involving whole of the ramus of mandible, sigmoid notch and condylar process with intact lower border of mandible. (Fig.3) A differential diagnosis of Odontogenic Keratocyst, Dentigerous cyst or Ameloblastoma was considered and Incisional biopsy was carried out. On histological examination, the diagnosis was given as unicystic ameloblastoma with mural proliferation. Considering the age of the patient it was decided to manage the case conservatively inspite of having mural proliferation. Surgical enucleation with extraction of involved teeth was carried out. Chemical cauterization was done using Carnoy's solution and postoperative dressings of bismuth subnitrate, Iodo form and paraffin paste was given (Fig.4). The dressing was changed every week.

Follow up was carried out every week for a month and every month for a year. Postoperative radiographs showed diffuse radiopacity which confirms bone formation.(Fig.5,6,7)



Figure- 1 No signs of facial asymmetry were noted on extraoral examination



Figure- 2 Intraoral examination revealed missing left mandibular third molar with mild swelling in the region of 38



Figure 3 Pre-operative OPG revealed a well defined, unilocular solitary radiolucency involving distal root of 36, 37, impacted 38 and involving whole of the ramus of mandible, sigmoid notch and condylar process with intact lower border of mandible



Figure 4 Immediate post-operative surgical enucleation with extraction of involved teeth was carried out. Chemical cauterization was done using Carnoy's solution and postoperative dressings of bismuth subnitrate, Iodoform and paraffin paste



Figure 5 Post – Operative OPG after 1 month



Figure 6- Post- operative OPG after 3 months.



Figure 7 Post- operative OPG after one and half year showed diffuse radiopacity which confirms bone formation.



Figure 8 Aspiration and excised specimen

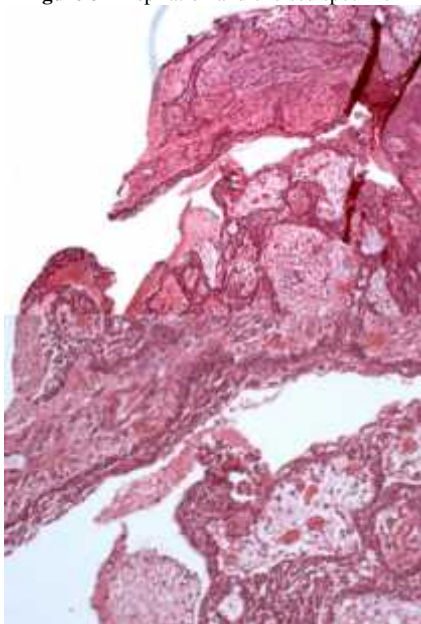


Figure 9 Histopathological slides of unicystic Ameloblastoma with mural proliferation

Histopathological studies

Histology slides show tissue showing cystic lumen lined by a basal cell layer of ameloblasts like cells with overlying stellate reticulum like cells. Connective tissue wall is infiltrated by odontogenic epithelium in the form of strands & cords which are bounded by ameloblastic cells surrounding stellate reticulum like cells. Some areas of squamous metaplasia are also seen. Final diagnosis was made as Unicystic Ameloblastoma with Mural Proliferation (Fig.8 & 9)

DISCUSSION

Ameloblastoma is divided into 3 clinico-radiographic groups: solid or multicystic, unicystic and peripheral. The solid ameloblastoma is the most common form of the lesion (86%). It has a tendency to be more aggressive than the other types and has a higher incidence of recurrence. (Hollows *et al* 2000) Unicystic ameloblastoma has a large cystic cavity with luminal, intraluminal or mural proliferation of ameloblastic cells. It is a less aggressive variant and it has a low rate of recurrence, (Kim *et al* & Rosenstein *et al* 2001,) although lesions showing mural invasion are an exception and should be treated more aggressively. (Rosenstein *et al* 2001) Various treatment modalities for Unicystic ameloblastoma have been used such as segmental or marginal resection, more conservative treatment such as enucleation and curettage, marsupialization to reduce the size of the lesion, followed by second stage surgery. (Robinson *et al* 1977 & Nakamura *et al* 2002) These treatments can be followed by adjunctive therapy including cryotherapy, thermal or chemical cauterization, and even radiotherapy or chemotherapy. (Holland *et al* 1981 & Lee *et al* 2004) The reported recurrence rate after treatment for unicystic ameloblastoma ranges from 10% to 25%. (Robinson *et al* 1977) There is no adequate evidence to prove which treatment modality is more effective. Resection of Unicystic ameloblastoma results in the lowest recurrence rate (3.6%) if adequate bone margins are removed. Despite high success rate for resection of UA more conservative treatment in order to optimize quality of life is generally favoured. (Lau *et al* 2006) Marsupialization together with other treatment resulted in an 18% recurrence. The aim of Marsupialization is to reduce the size of UA so that less extensive surgery is required. The treatment modality is not so popular method, but most benefit is expected in severely ill patients or those with a huge lesion. (Atldnson *et al* 1994, Lau *et al* 2006) Enucleation alone yielded the highest recurrence rate among all treatments (30.5%). Two possible explanations: firstly, cystic lining of the tumor is inadequately removed; secondly, ameloblastic tumor cells can invade the cancellous bone to a certain extent. (Lau *et al* 2006) Enucleation followed by application of Carnoy's solution has resulted in a recurrence rate of 16.0% which is the best except for resection. The recurrence rate could even lower than reported, if the closely related teeth with tumor are extracted. Because in an attempt to preserve the tooth without damage, tumor remnants may be left around the tooth apex or root and these may lead to recurrence. (Lee *et al* 2004 & Lau *et al* 2006). In the present case report, teeth in close relation of tumor were extracted. Carnoy's solution, a powerful fixative penetrates the cancellous spaces and thus fixes the remaining tumor cells. Usually, Carnoy's solutions are applied for 3-5 min. However, (Frerich *et al* 1994) suggested that the application of Carnoy's solutions should not exceed by 3 min and should not be

directly applied over the nerve as it could lead to nerve impairment.

CONCLUSION

Although unicystic Ameloblastoma is benign but is life threatening, Due to airway obstruction, &starvation from restriction of feeding. We should be aware of the unilocular radiolucencies of the jaws as these lesions could be UA. Timely intervention and conservative surgical treatment followed by application of Carnoy's solution and the extraction of closely related teeth may improve treatment outcome and potential complications associated with larger resection. Considering the characteristics of the tumour, the priority of the treatment should be from the point of morbidity and quality of life of the patients, noting that the rate of recurrence is not always the primary factor.

References

- Small IA, Waldron CA. Ameloblastoma of jaws. *Oral Surg* 1955;8:281-97.
- Sehdev MK, Huvos AG, Strong EW, Gerold FP, Willis GW. Ameloblastoma of maxilla and mandible. *Cancer* 1974;33:324-33.
- Gardner DG, Pecak AMJ. The treatment of ameloblastoma based on pathologic and anatomic principles. *Cancer* 1980;46:2514-9.
- Williams TP. Management of ameloblastoma: a changing perspective. *J Oral Maxillofac Surg* 1993;51:1064-70.
- Feinberg SE, Steinberg B. Surgical management of ameloblastoma: current status of the literature. *Oral Surg Oral Med Oral Pathol* 1996;81:383-8.
- Ferretti C, Polakow R, Coleman H. Recurrent ameloblastoma: report of 2 cases. *J Oral Maxillofac Surg* 2000; 58(7):800-4.
- Becelli R, Carboni A, Cerulli G, Perugini M, Iannetti G. Mandibular ameloblastoma: analysis of surgical treatment carried out in 60 patients between 1977 and 1998. *J Craniofac Surg* 2002; 13(3):395-400.
- Feinberg SE, Steinberg B. Surgical management of ameloblastoma: current status of the literature. *Oral Surg Oral Med Oral Pathol* 1996;81:383-8.
- Gardner DG. Radiotherapy in the treatment of ameloblastoma. *Int J Oral Maxillofac Surg* 1988;17:201-5.
- Garcia NG, Oliveira DT, Rodrigues MT. Unicystic Ameloblastoma with Mural Proliferation Managed by Conservative Treatment. *Case Rep Pathol*. 2016;2016:3089540. doi: 10.1155/2016/3089540.
- Atldnson CH, Harwood AR, Cummings BJ. Ameloblastoma of the jaw: a reappraisal of the role of megavoltage irradiation. *Cancer* 1994;53:869-73.
- Wenlu Li, Fayu Liu, Zhongfei Xu, Shaohui Huang, Wei Zhu and Changfu Sun1 Treatment of Ameloblastoma in Children and Adolescents *Journal of Hard Tissue Biology* 21[2] (2012) p121-126
- Hollows P, Fasanmade A, Hayter JP. Ameloblastoma – a diagnostic problem. *Br Dent J* 2000; 188(5):243-4.
- Kim SG, Jang HS. Ameloblastoma: a clinical, radiographic and histopathologic analysis of 71 cases. *Oral Surg Oral Med Oral Pathol Oral Radiol Endod* 2001; 91(6):649-53.
- Rosenstein T, Pogrel MA, Smith RA, Regezi JA. Cystic ameloblastoma—behaviour and treatment of 21 cases. *J Oral Maxillofac Surg* 2001; 59(11):1311-6
- Robinson L, Martinez MG. Unicystic Ameloblastoma: A Prognostic Distinct Entity. *Cancer*. 1977;40:2278-85.
- Nakamura N, Higuchi Y, Mitsuyasu T, Sandra F, Ohishi M. Comparison of long term result between different approaches of ameloblastoma. *Oral Surg Oral Med Oral Pathol Oral Radiol Endod*. 2002;93:13-20.
- Holland PS, Mellor WC. The conservative treatment of ameloblastoma using diathermy or cryosurgery. A 29 year review. *Int J Oral Surg*. 1981;10:32-6.
- Lee PK, Samman N, Ng IO. Unicystic ameloblastoma- Use of Carnoy's solution after enucleation. *Int J Oral Maxillofac Surg*. 2004;33:263-7
- Lau SL, Samman N. Recurrence related to treatment modalities of unicystic ameloblastoma: A systematic review. *Int J Oral Maxillofac Surg*. 2006;35:681-90.
- Frerich B, Cornilious CP, Wietholter H. Critical time of exposure of rabbit inferior alveolar nerve to Carnoy's solution. *Int J Oral Maxillofac Surg*. 1994;52:599-606.

