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Unicystic Mural Ameloblastoma: A Case Report

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Abstract: ***Unicystic ameloblastoma (UA) are cystic lesions that shows clinical, radiographic and gross features similar to a mandibular cyst, and can be definitely diagnosed only with the help of histopathological examination. They usually present as a painless, slowly enlarging swelling which commonly occurs in mandible and accounts for approximately 1% of all oral tumors. Though its recurrence rate is low, it may recur after a long time period which necessitates the need for long term follow up. Here we present a case of unicystic mural ameloblastoma in a 45 years old female patient who was treated with surgical enucleation.***

Keywords: Enucleation, Unicystic Ameloblastoma, Mural type, Mandibular cyst.

1. Introduction

Ameloblastoma was defined by World Health Organization in the year 1991 as a benign but locally aggressive tumor with a high tendency to recur, consisting of proliferating odontogenic epithelium lying in a fibrous stroma [2]. Unicystic ameloblastoma (UA) are cystic lesions that shows clinical and radiographic features similar to a mandibular cyst, and can be definitely diagnosed only with the help of histopathological examination [6]. Unicystic ameloblastoma is considered to be a less aggressive form compared with solid or multicystic type and accounts for 5 to 15% of all ameloblastomas [1], [4]. Based on histopathologic examination three variants of unicystic ameloblastoma were described, which are luminal, intraluminal, and mural [8]. Following enucleation and curettage, the recurrence rate of all unicystic ameloblastomas were reported as 10% to 20% [5]. Here we report a case of unicystic ameloblastoma type III (transmural) affecting the left anterior mandibular region in a 45-year-old female patient along with its radiological and histological findings.

2. Case Report

A 45 year old female patient presented with a swelling in lower left side of mouth since 4 months. There is no associated pain, difficulty in chewing/mouth opening. On extraoral examination minimal to no swelling is evident.

Figure 1: Pre op Profile

On intraoral examination, a swelling is noted in the left anterior mandibular region extending from left mandibular central incisor to left mandibular first premolar region with buccolingual expansion. The mucosa appears normal in colour and texture. No bleed/ooze noted from the swelling. Left mandibular canine is missing and left mandibular lateral incisor shows grade I mobility. On palpation, the inspectory findings are confirmed. It is tender and non indurated. Also lymph nodes were not palpable. No other abnormalities detected on systemic examination.

Figure 1: Pre op Intraoral photograph

An orthopantamogram(OPG) was done which showed a unilocular expansile lesion extending from left mandibular central incisor to left mandibular first premolar region region with well defined, corticated, non scalloped border. Resorption seen with respect to left mandibular central and lateral incisor also left mandibular first premolar. Small area in the crestal margin between left mandibular lateral incisor and left mandibular first premolar shows honey comb appearance in the superior aspect of unilocular lesion.

Figure 2: Pre operative OPG

It was provisionally diagnosed as residual cyst and treated by surgical enucleation. Patient was taken for surgery under general anesthesia. Cyst was surgically removed in toto along with the cystic lining and extraction done with respect

to mandibular central and lateral incisor, mandibular first and second premolar. Closure done with 3-0 vicryl. Also drain placed in the operated site and secured.

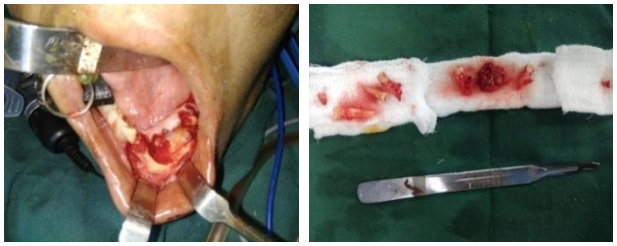

Figure 3: Intraoperative photographs

Postoperatively follow up done after 1 week, 2 weeks, 1 month and 3 months clinically and radiographically(OPG). Healing is satisfactory and no evidence of recurrence noted.

Figure 4: Post op Clinically – 6 months

Figure 5: Post operative OPG – 6 months

The H & E stained and studied section from the submitted excised specimen show predominantly thin non keratinized 2-3 cell thick ameloblastomatous proliferative lining associated with luminal and mural proliferations in the form of islands, tiny clusters and cords supported by connective tissue wall. Foci of resorbing host bone also noted with evidence of lesional cell infiltration.

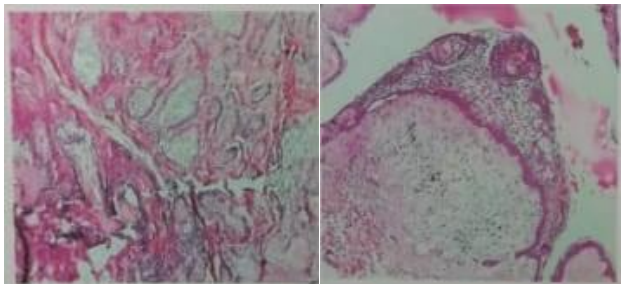

Figure 6: Histopathology report

3. Discussion

Ameloblastoma is a benign tumor of odontogenic origin which is slow growing, locally aggressive, common in mandible and accounts for approximately 1% of all oral tumors [11]. Robinson in 1937 defined ameloblastoma, as a benign tumor that is “unicentric, nonfunctional, intermittent in growth, anatomically benign and clinically persistent.” Clinically ameloblastomas are classified in three types: 1) solid and multicystic ameloblastoma, 2) unicystic ameloblastoma (UA) and 3) peripheral ameloblastoma [12].

Unicystic ameloblastoma (UA) are cystic lesions that shows clinical, radiographic and gross features similar to a mandibular cyst, and can be definitely diagnosed only with the help of histopathological examination [6]. Three histologic subtypes of unicystic ameloblastoma are described, based on the character and extent of tumor cell proliferation within the cyst wall, 1. Luminal, which is of simple cystic nature, 2. Intraluminal, which is those with intraluminal proliferation nodules, and 3. Mural type, which contains infiltrative tumor islands in the cyst walls [8].

As unicystic ameloblastoma shows similarity in clinical, radiographic and gross features to other odontogenic cyst, it becomes sometimes difficult to arrive at a conclusive diagnosis. Different treatment modalities are described in the literature which includes enucleation with or without the application of carnoy’s solution, marsipulization, resection [12].

Lau and Samman in their study reported that recurrence rates of 3.6% is there after resection, 30.5% following enucleation alone, 16% on enucleation followed by Carnoy’s solution application, and 18% if marsupialisation is done followed by enucleation [13].

The decision to choose a particular treatment whether surgical or conservative depends on the character and extent of the lesion and considering its tendency to recur. According to Marx and Stern, Invasive ameloblastoma arising from the epithelial lining and proliferating through the complete thickness of the connective tissue layer of a cyst should be treated with resection, and ameloblastoma “in situ” developing in, which is limited to the epithelial lining of a cyst, microinvasive ameloblastoma arising from the epithelial lining and proliferating into the connective tissue layer of the cyst, enucleation can be done [9]. Lee et al [10] showed that there is lower recurrence rate when carnoy’s solution is used after enucleation of type III unicystic ameloblastoma.

This case was preoperatively diagnosed as residual cyst and careful surgical enucleation was done. These kind of lesions require histopathological examination to arrive at proper diagnosis [7]. After histopathology reporting was done, the lesion was defined as unicystic mural ameloblastoma. Therefore because of its tendency to recur, the patient was kept on follow up.

Three months follow up was done and there was no signs of any recurrence. Though the recurrence rate is low, Unicystic ameloblastoma may recur after a long time period. So a long-term postoperative follow up is advocated for detecting the lesion at an early stage and to prevent local invasion.

4. Conclusion

Usually diagnosis of ameloblastoma may be delayed because of its low grade symptoms and prevalence. Moreover unicystic ameloblastomas are very difficult to diagnose with the help of clinical and radiographic examination alone because of its close similarity to cysts. Histopathological examination of the excised specimen is necessary to rule out such lesions, to decide on optimal treatment and further follow up. Enucleation though it's a good modality of treatment, strict follow up is necessary to detect any signs of recurrence. Unicystic ameloblastoma because of its difficulty to diagnose clinically and radiographically and because of its tendency to recur is a dilemma to surgeons.

References

[1] Kramer IR, Pindborg JJ, Shear M. 2nd ed. Berlin: Springer-Verlag; 1992. Histological Typing of Odontogenic Tumours. WHO International Histological Classification of Tumours; pp. 11–14.
[2] Reichart HP, Philipsen P. Unicystic ameloblastoma. A review of 193 cases from the literature. Oral Oncology 1998; 34: 317-25
[3] Harnet JC, Raybaud H, Rocca JP, et al. Unicystic ameloblastoma in a child: a case study and review of the literature. Int J Pediatr Otorhinolaryngol 2008;3:192–196
[4] Robinson L, Martinez MG. Unicystic ameloblastoma: a prognostic distinct entity. Cancer 1977;40:2278–2285
[5] Gardner DG, Corio RL. Plexiform unicystic ameloblastoma. A variant of ameloblastoma with a low recurrence rate after enucleation. Cancer 1984;53:1730–1735
[6] Rakesh S Ramesh, Suraj Manjunath, Tanveer H Ustad, Saira Pais, and K Shivakumar. Unicystic ameloblastoma of the mandible – an unusal case report and review of literature. Head Neck Oncology 2010, 2:1
[7] Alexander d. Rapidis, Angelos p. Angelopoulos, Cris A. Skouteris and Stavros Papanicolaou. Mural (intracystic) ameloblastoma. Int. J. Oral surg. 1982: 11: 166-174
[8] B. M. Rudagi, R. Halli, Y. Kini, V. R. Kharkar, and S. Asnani, "Unicystic ameloblastoma of the mandible managed by conservative approach: a case report," Journal of the Indian Dental Association, vol. 5, no. 6, pp. 743–745, 2011.
[9] R. E. Marx and D. Stern, "Odontogenic tumors: hamartomas and neoplasms," in Oral andMaxillofacil Pathology: A Rationale for Diagnosis and Treatment, pp. 671–678, Quintessence, 2012.
[10] Lee PK, Samman N, Ng IO. Unicystic ameloblastoma-Use of Carnoy's solution after enucleation. Intl J Oral Maxillofac Surg 2004;33:263-7
[11] K. M. K. Masthan, N Anitha, Jayasri Krupaa, and Sudha Manikkam. Ameloblastoma. J Pharm Bioallied Sci. 2015 Apr; 7(Suppl 1): S167–S170.
[12] David Neagu,Oscar Escuder-de la Torre, Inés Vázquez-Mahía, Nicolás Carral-Roura, Guillermo Rubín-Roger, Ángel Penedo-Vázquez, Ramón Luaces-Rey, and José-Luis López-Cedrún. Surgical management of ameloblastoma. Review of literature. J Clin Exp Dent. 2019 Jan; 11(1): e70–e75.
[13] S. L. Lau and N. Samman, "Recurrence related to treatment modalities of unicystic ameloblastoma: a systematic review," International Journal of Oral and Maxillofacial Surgery, vol. 35, no. 8, pp. 681–690, 2006.
[14] G. L. Ackermann, M. Altini, and M. Shear, "The unicystic ameloblastoma: a clinicopathological study of 57 cases," Journal of Oral Pathology, vol. 17, no. 9-10, pp. 541–546, 1988.

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