Recurrent unicystic mural type ameloblastoma in a 9-year-old boy, 8 years follow-up

Burcu Sengüven, Emre Barış, Tülin Oygür, Akın Öztémel

Abstract

Unicystic ameloblastoma is not a rare odontogenic tumor in the pediatric population. A significant care should be given to unicystic ameloblastoma if it has mural invasions due to its local aggressiveness, high recurrence rates and radical management options as in conventional ameloblastoma. Fine needle aspiration (FNA) cytology is a rapid, non-traumatic diagnostic method that provides a required attention prior to surgery. We present an excisional biopsy proved FNA diagnosed mural type unicystic ameloblastoma in a 9-year-old child recurred as a solid ameloblastoma after 8 years. When distinctive features of ameloblastoma are known, an accurate diagnosis can be made by FNA cytology, in combination with clinicoradiological findings. This method gives benefit to the patients especially the younger ones both for the pre-operative surgical planning and the post-operative follow-up.

Keywords: Ameloblastoma, fine needle aspiration, odontogenic tumor

Introduction

Odontogenic tumors are a large group of neoplasm or hamartomatous growth of the jaws arises from tooth forming tissues.[1] During the dentinogenesis ectomesenchimal cells migrates from the neural crest to the jaws while overlying ectoderm send down to form enamel organ. Among all odontogenic tumors ameloblastoma is the most significant one;[2] although, it represents only 1% of all cystic or tumoral lesions in jaws, not more frequent than other odontogenic tumors, excluding odontomas.[3] Ameloblastomas derives from odontogenic epithelium, cellular remnants of the enamel organ or the basal layer of the oral mucosa.[4] Ameloblastomas are characterized by its histological resemblance to the enamel organ. Partial maxillectomy or mandibulectomy is the most common treatment choice with a safety margin of healthy bone to avoid recurrence.[5] Unicystic ameloblastoma is one of the four different clinicopathologic subtypes of ameloblastoma [Table 1]. In various studies, it accounts 10-40% of all intraosseous ameloblastomas. Approximately, 70% of unicystic ameloblastomas are presenting during the second and third decades.[6] Ord et al. reported a higher percentage of unicystic ameloblastoma in children with only 2.2% under 10 years old.[7] Unless the lesion is not large, it is often asymptomatic. The clinical and radiologic findings may suggest a cystic lesion and microscopical examination is needed for the diagnosis of ameloblastoma.[8]

The aim of this paper is to describe the clinicopathologic features of a unicystic ameloblastoma located in the posterior mandible of a child, which recurred after 8 years as a solid ameloblastoma. We also aimed to call attention to this relatively common and radical management options requiring tumor in the differential diagnosis and highlight the significance of fine needle aspiration (FNA) biopsies and long-term follow-up especially in pediatric group.

Case Report

A 9-year-old boy presented with large, well-defined unicystic radiolucency with unerupted second premolar permanent tooth in left mandible base [Figure 1]. On intraoral examination, there were no sign of lesion such as bone expansion or ulceration. After the radiologic and intraoral examination, FNA procedure was performed by a dental surgeon with initial diagnosis of dentigerous cyst.

The slides were wet fixed and stained with hematoxylin and eosin. Cytological evaluation of FNA demonstrated individual cell with eosinophilic cytoplasm and cantered nucleolus, on the other hand tightly packed groups of oval and elongated epithelial cells of basaloid appearance with thin chromatin pattern without any cellular atypia or pleomorphism [Figure 2]. The cytological findings were diagnostic for “odontogenic epithelial lesion without atypia.” The therapeutic approach to the lesion was excision with the extraction of molar primary tooth.
and the first and second permanent premolar teeth. Histopathological examination of excisional material was performed by Oral Pathology Department of Gazi University Dental Faculty. Macroscopically, the specimen was brownish cystic lesion measuring 3.5 cm × 2.5 cm × 2.0 cm. Cyst has multiple, small intraluminal cystic spaces. Multiple sections of the tumor were prepared routinely and stained with hematoxylin and eosin. Histologically, the lesion was nearly

**Figure 1:** A large, well-defined unilocular radiolucency of the initial lesion in left posterior mandible enclosing the permanent second premolar

**Figure 2:** Epithelial cells arranged in clusters with characteristic peripheral palisading as a uniform population and without any nuclear atypia from the initial fine needle aspiration (H and E, ×40)

**Figure 3:** Ameloblastomatous epithelium lining cyst from the initial excisional biopsy. The fibrous wall has tumor islands (arrow) (H and E, ×40)

**Figure 4:** Panoramic radiographic examination of the recurrent lesion showing unilocular radiolucency in posterior mandible

**Figure 5:** Histopathological examination of recurrent lesion showing typical reverse polarization of basal cells and stellate reticulum cells of follicular ameloblastoma (H and E, ×100)

**Figure 6:** Tumor free surgical bone margin (arrow) (H and E, ×40)
completely cystic, lined by ameloblastomatous epithelium with additional intraluminal extensions. Cyst epithelium was composed of peripherally palisaded basaloid cells and stellate reticulum cells, which exhibits follicular pattern. There were also tumor islands on the cyst wall [Figure 3]. According to these histopathological findings case was diagnosed as “unicystic ameloblastoma, mural type.” Dental surgeons approved no further treatment because of the continuing bone development of patient.

The 8 years post-operative follow-up revealed a recurrence at the same location [Figure 4]. The only clinical symptom was a slight bone expansion, which patient did not aware of. Radiographically the lesion was well-defined and FNA could not be performed due to firm and solid texture of the lesion. After the intraoperative “conventional ameloblastoma, follicular type” diagnosis from frozen sections, further bone resection was applied with safety margins. The diagnosis was approved by paraffin block sections of the lesion [Figures 5 and 6]. Figure 7 shows recovering bone defect after 8 months of surgery, free of tumor.

Discussion

It is not always easy to estimate the actual incidence of ameloblastoma in children because of the differences of age limit defining pediatric population and also the inconsistency of histological classifications in earlier publications. However, it is known that the mean age is approximately 25.3 years for unicystic ameloblastoma and it peaks in the second and third decades. In younger age group, unicystic ameloblastomas are seen more than solid-multicystic ameloblastomas.[7,8]

FNA has been used increasingly as a diagnostic procedure to evaluate neoplastic and non-neoplastic lesions in various tissues. It is a safe, cost-effective and well-tolerated method with few complications. In a small number of cases, FNA has been followed by varying degrees of necrosis in some organs.[9] Ameloblastomas can be aspirated easily and the cytological features are sufficiently distinctive. FNA provides a pre-surgical diagnosis and a reliable method for the diagnosis of ameloblastoma. Usually, FNA is not the first choice for diagnose in ameloblastomas, probably due the fact that incisional biopsy is not difficult. Nevertheless, it is still a very useful tool in the young patients, permitting a rapid, innocuous, economic, reliable and the most imported less traumatic diagnosis.[10]

At the microscopical level ameloblastoma demonstrates some typical features like tightly cohesive groups of neoplastic epithelial cells with basaloid appearances and scanty or moderate amount of cytoplasm. The centrally located nuclei were round to oval and squamous metaplastic changes might be present. Multinuclear foreign body-type giant cells in the background might be seen believe to be belong to the surrounding granulation tissue.[10]

Most unicystic ameloblastomas are enucleated with the pre-operative clinical diagnosis of dentigerous cyst due to the high frequency of unerupted tooth relation of the lesion.[8] Ameloblastomas do not have a unique clinical course and their clinical and radiological findings are similar to other odontogenic lesions, or even non-odontogenic lesions like brown tumor, giant-cell granuloma or eosinophilic granuloma. Pathologic examination is the only way to determine its true nature. On the other hand, although they have benign cytohistologic appearance, ameloblastomas are locally aggressive and tend to recur. Mural type unicystic ameloblastoma does infiltrate surrounding bone and further treatment might be required for these tumors. Although invasion is limited as in our case, which confirmed by sufficient sectioning, careful follow-up is recommended.[6,8]

The recurrent lesion showed a different clinicopathologic subtype than was encountered in the primary. A change from unicystic to solid ameloblastomas was noticed which is not only very rare, but also should warn both clinicians and pathologist about their differential diagnosis, follow-up periods and the extraordinary nature of this disease.

Conclusion

As conclusion FNA has several benefits compared with open surgical biopsy, especially in young patients for the pre-operative surgical planning and the post-operative follow-up. Therefore, FNA is a valuable diagnostic tool for children with a risk or history of ameloblastoma.
References

1. da-Costa DO, Maurício AS, de-Faria PA, da-Silva LE, Mosqueda-Taylor A, Lourenço SD. Odontogenic tumors: A retrospective study of four Brazilian diagnostic pathology centers. Med Oral Patol Oral Cir Bucal 2012;17:e389-94.
2. Arotiba GT, Effiom AO, Ayodele AS, Goundana MO, Gbotolorun MO, Olasoji HO, et al. A classification system for recurrent ameloblastoma of the jaws – Review of 30 cases in Nigerians. Nig Q J Hosp Med 2012;22:44-51.
3. Kruse AL, Zwahlen RA, Grätz KW. New classification of maxillary ameloblastic carcinoma based on an evidence-based literature review over the last 60 years. Head Neck Oncol 2009;1:31.
4. Reichart PA, Philipsen HP, Sonner S. Ameloblastoma: Biological profile of 3677 cases. Eur J Cancer B Oral Oncol 1995;31B: 86‑99.
5. Sannomiya EK, Silva JV, Brito AA, Saez DM, Angelleri F, Dalben Gda S. Surgical planning for resection of an ameloblastoma and reconstruction of the mandible using a selective laser sintering 3D biomodel. Oral Surg Oral Med Oral Pathol Oral Radiol Endod 2008;106:e36-40.
6. Li TJ, Wu YT, Yu SF, Yu GY. Unicystic ameloblastoma: A clinicopathologic study of 33 Chinese patients. Am J Surg Pathol 2000;24:1385‑92.
7. Ord RA, Blancaert RH Jr, Nikitakis NG, Sauk JJ. Ameloblastoma in children. J Oral Maxillofac Surg 2002;60:762‑70.
8. Li T, Wu Y, Yu S, Yu G. Clinicopathological features of unicystic ameloblastoma with special reference to its recurrence. Zhonghua Kou Qiang Yi Xue Za Zhi 2002;37:210‑2.
9. Singh Nanda KD, Mehta A, Nanda J. Fine-needle aspiration cytology: A reliable tool in the diagnosis of salivary gland lesions. J Oral Pathol Med 2012;41:106‑12.
10. Uçok O, Doğan N, Uçok C, Günhan O. Role of fine needle aspiration cytology in the preoperative presumptive diagnosis of ameloblastoma. Acta Cytol 2005;49:38‑42.

How to cite this article: Sengüven B, Baris E, Oygür T, Oztemel A. Recurrent unicystic mural type ameloblastoma in a 9-year-old boy, 8 years follow-up. Contemp Clin Dent 2013;4:569‑72.

Source of Support: Nil. Conflict of Interest: None declared.