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Mural Unicystic Ameloblastoma in Anterior Region of Mandible: A Case Report

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ABSTRACT

Ameloblastoma is a benign but locally invasive epithelial odontogenic tumor. Ameloblastoma usually presents in the posterior mandibular ramus region, while it is rare in the anterior mandibular region. It has been divided into three classic types: Unicystic Ameloblastoma (UA), Extraosseous/ Peripheral Ameloblastoma, and Conventional Ameloblastoma. The unicystic type accounts for approximately 5-15% of all cases and predominantly occurs in the younger population, typically in the second decade of their life, but is quite rare in older adults. We present a case of mandibular unicystic Ameloblastoma in the anterior area of a 62-year-old Iranian female with intraoral swelling of the gingiva. We reported the clinicoradiographic and histopathological features of the lesion with complete treatment intervention and follow-up.

Keywords: Ameloblastoma; Mural; Odontogenic tumor.

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Introduction

Ameloblastoma is the most recurrent tumor with an odontogenic origin. It is usually locally invasive and originates from epithelial cellular elements and dental tissues [1,2]. However, the tumor is slow-growing and often asymptomatic, which leads to its discovery in the early stages incidentally. About 80% of Ameloblastomas occur in the mandible: 70% of cases are in the molar region and ascending ramus, 20% in the premolar region, 10% in the incisor region, and about 20 % of all cases occur in the maxilla [3]. According to the World Health Organization (WHO) classification of head and neck tumors in 2022, there are five types of Ameloblastoma: Unicystic Ameloblastoma (UA), Extraosseous/Peripheral Ameloblastoma, Conventional Ameloblastoma, Adenoid Ameloblastoma, and Metastasizing Ameloblastoma [4]. The conventional type is the most prevalent one, and the average age based on statistics is about the 4th-5th decade of life [3,4]. The unicystic type accounts for approximately 5-15% of all cases. Which mainly involves the younger population in the second decade of their life. UA has three subtypes: luminal (simple), intraluminal, and mural [2,3]. Both simple and luminal subtypes are usually treated by enucleation, while the mural subtype requires more considerable treatments [5]. Unicystic Ameloblastoma is quite rare in older adults. We present a case of mandibular unicystic Ameloblastoma in the anterior area of a 62-year-old female and discuss clinical, radiographical, and histomorphological features of this tumor, especially in older adults.

Case Presentation

A 62-year-old female presented with a swelling of the mandible of unknown duration and was referred to the Oral and Maxillofacial Pathology Department of Tehran University of Medical Sciences (TUMS). The patient's past dental history (PDH) and past medical history (PMH) were not significant. In intraoral examinations, a mild, painless mandibular swelling in the anterior area without tooth mobility was evident. In the extraoral examination, there was no sign of swelling or asymmetry; she had neither tenderness nor pain, nor numbness in the region. In the radiographic view, a radiolucent lesion with well-defined borders and a maximum dimension of 5mm was observed on the mesial side of the right mandibular fourth tooth, extending to the region of the mental foramen. The lesion was unilocular on the right side and semilocular on the left side, and had caused expansion in the buccal side of the cortical jaw bone. The lesion also caused root resorption in anterior teeth (Figure 1). Differential diagnoses of the lesion based on radiographic features included glandular odontogenic cyst (GOC), Unicystic Ameloblastoma (UA), Odontogenic tumor, and low-grade intraosseous malignancies such as odontogenic cysts and mucoepidermoid carcinoma (less probable). Fine needle aspiration yielded bloody fluid, confirming the cystic nature of the lesion. Given the lesion's small size, its predominantly cystic nature, and the results of aspiration, the oral and maxillofacial surgeon opted for complete excision of the lesion as the initial treatment approach. An excisional biopsy and peripheral ostectomy were performed. The macroscopic view revealed several cystic tissues, consisting of several pieces of creamy, yellow cystic tissue measuring 3×2.5×1cm. The cut section revealed a homogenous cystic surface with a maximum thickness of 0.1cm. Microscopic examinations of the specimen revealed a cystic lesion lined by hyperplastic stratified epithelium, characterized by palisaded, reversed-polarized basal cells and loosely arranged superficial cells that proliferated toward the underlying fibrotic connective tissue. The mural nests revealed granular cell changes and basaloid features in some areas. The other pieces were a bony tissue consisting of four pieces of gray to black bony-hard tissue measuring 1.5×1×0.5cm. Microscopic examinations of the bony tissues showed sheets of lamellar bone trabeculae. Numerous tumoral nests and islands demonstrated peripheral palisaded cells, squamous metaplasia, and keratin formation were seen among the bone trabeculae (Figure 2). These histomorphologic features were compatible with the diagnosis of UA, mural type. Now, 32 months after surgical treatment, no evidence of recurrence was seen in the follow-up of the patient (Figure 3).

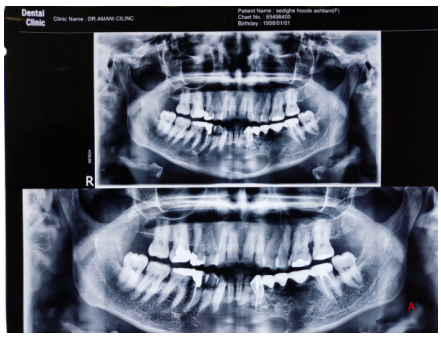

Figure 1. Radiographic view of the lesion. A) Panoramic view shows a radiolucent lesion in the anterior part of the mandible, which has crossed the midline with distinct borders. B) Cone-beam computed tomography shows root resorption and expansion in the buccal side of the mandible. C) Cone-beam computed tomography shows semi-multilocular behavior of the lesion in the left part of the lesion.

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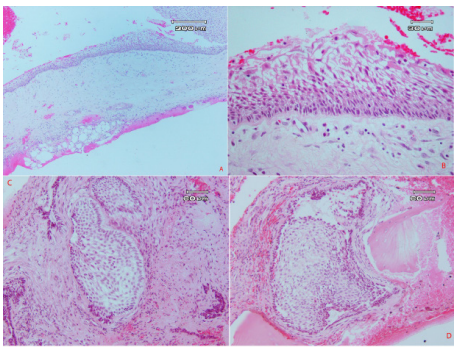

Figure 2. Histopathologic view of the lesion. A)Epithelial lining of the cyst (×100 magnification) shows palisaded reverse-polarized basal cells. B) Epithelial lining superficially loosely arranged cells (×400 magnification). C) Some mural nests and islands invaded fibrous connective tissue cyst wall (×200 magnification). D) a large mural nest close to regional bone trabeculae (×200 magnification).

Discussion

Ameloblastoma is a well-known locally invasive tumor that is about 1% of all oral tumors and about 13%-78% of all odontogenic tumors [2,6]. Ameloblastoma is an epithelial-originated neoplasm that may develop from the rest of the dental lamina, enamel apparatus, and the epithelial lining of an odontogenic cyst (mostly dentigerous cyst) or the basal epithelial cells of the oral mucosa [6]. Based on a systematic review by Michael P. Chae et al, the female to male ratio was (2.67:1) [3]. According to the 2022 World Health Organization (WHO) classification, ameloblastoma is divided into unicystic ameloblastoma, extraosseous/peripheral ameloblastoma, conventional ameloblastoma, adenoid ameloblastoma, and metastasizing ameloblastoma [4].

Unicystic type includes 5% to 15% of all Ameloblastomas and more than 90% of unicystic Ameloblastomas involve the mandible, usually the posterior region; and up to 80% are related to an unerupted mandibular third molar tooth [7]. In contrast, the anterior region of the mandible is considered to be rare and atypical. In our case, the UA occurs in the anterior site of the mandibular bone, an uncommon anatomical location. UA occurs across a wide age range, with a mean age of 35 years for non-impacted tooth-related cystic Ameloblastoma, in contrast to 16 years for the impacted tooth-related variant [7]. However, it is less common in the seventh decade of life. Our reported case was a UA in a 62-year-old female. Classification. It often appears clinically as a slow-growing, painless swelling, causing expansion of cortical bone, spreading of the lingual and/or buccal plates, and penetration of soft tissue [6].

The patients usually complain of draining sinuses, ulceration, mobile teeth, ill-fitting dentures, malocclusion, and nasal obstruction [8]. Facial asymmetry is a clinical late sign and the main reason patients seek dental care [7]. Our patient was complaining of an intraoral prominence, which appeared as painless swelling. Radiographically, Ameloblastoma may appear as a well-defined unilocular radio transparency with or without the presence of sclerotic margins, frequently associated with an unerupted tooth, or as multilocular radio transparency, which may be described in terms of “honeycombs” or “soap bubbles” [9]. In contrast, unicystic Ameloblastoma may appear as a unilocular cystic lesion with or without scalloped margins in radiographic views [10]. These findings resemble jaw cysts; however, histopathological analysis is required to confirm the final diagnosis [7].

The histopathological view of unicystic Ameloblastoma shows a single cystic lesion lined by odontogenic (Ameloblastomatous) epithelium [2]. Unicystic ameloblastoma itself has different classifications. Ackerman has classified it into three histologic groups as follows:

Group I: Luminal UA (tumor confined to the luminal surface of the cyst).

Group II: Intraluminal/plexiform UA (nodular proliferation into the lumen without infiltration of tumor cells into the connective tissue wall.

Group III: Mural UA (invasive islands of ameloblastomatous epithelium in the connective tissue wall not involving the entire epithelium) [1]. Another histologic subgrouping by Philipsen and Reichart has also been described: Subgroup 1: Luminal UA, Subgroup 1.2: Luminal and intraluminal, Subgroup 1.2.3: Luminal, intraluminal, and intramural, and finally Subgroup 1.3: Luminal and intramural [1]. The differential diagnosis of UA includes odontogenic lesions, both odontogenic cysts and tumors, and, less likely, non-odontogenic lesions [2]. The choice of treatment depends on the type of tumor and its clinical manifestations. We usually choose conservative operations for simple unicystic and peripheral Ameloblastoma, while solid or multicystic Ameloblastoma are often treated radically [6]. Curettage does not cause satisfactory results and has high rates of recurrence. The treatment of choice for Ameloblastomas is usually radical surgery; however, if it were possible to find a way to minimize sequelae, conservative treatment may be considered. For Ameloblastoma, a wide local excision with margins of 1.5 to 2 cm is recommended [9]. Marsupialization, followed by complete but conservative excision, may be considered

for luminal and intraluminal types of unicystic Ameloblastoma. At the same time, resection is recommended for the mural variant of unicystic Ameloblastoma [5,8]. Among UAs, luminal and intraluminal types are surrounded by the fibrous connective tissue wall of the cyst. Therefore, complete removal can be achieved through enucleation of the cyst. However, mural and invasive UAs involve the surrounding connective tissue wall of the cyst and can even extend beyond the surrounding bone. Therefore, resection is a better choice for these types of UAs [8]. In our case, due to a few small nests of ameloblastic tissue, mandibular peripheral ostectomy was the preferred treatment for the cyst wall.

Recurrence rates are also related to the type of initial treatment. Lau et al reported recurrence rates of 3.6% for resection, 30.5% for enucleation alone, 16% for enucleation followed by Carnoy's solution application, and 18% for marsupialization followed by enucleation (where the lesion reduced in size) [1]. In a study by Milman et al., the authors observed recurrence in 24% of patients with Ameloblastoma, with an average period to the first relapse of 4.6 years. However, Siar et al. found recurrence in 13.3% of cases with an average period of 7.3 years [9].

Antonoglou and Sandor (Antonoglou & Sandor, 2015) conducted a systematic review and meta-analysis on the recurrence rates of solid and unicystic Ameloblastomas based on studies published from 1977 to 2003. They found a lower risk of recurrence after radical compared to conservative treatment. However, due to the limited number of studies evaluating both treatment modalities in unicystic Ameloblastoma, conclusions can only be drawn for solid or conventional Ameloblastomas [6]. In our case, fortunately, there was no evidence of recurrence after conservative treatment, including enucleation and peripheral ostectomy, after more than two years.

Conclusion

In conclusion, UA is a type of Ameloblastoma, characterized by a variety of clinical, radiological, and histopathological features. Disclosure of a case in the seventh decade of life in the anterior region of the mouth is very rare, and fortunately, in this case, it was diagnosed successfully. After all, due to the possibility of recurrence, close follow-up is essential. We presented a 62-year-old female with a UA in the anterior site of the mandible, with a 32-month follow-up of the patient without any sign of recurrence.

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Conflict of Interest

There is no conflict of interest to declare.

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