An Unusual Case of Unicystic Ameloblastoma Involving the Mandible

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Citation

V Dewoolkar, D Belekar, A Desai, J Anam, M Parab. *An Unusual Case of Unicystic Ameloblastoma Involving the Mandible*. The Internet Journal of Surgery. 2008 Volume 20 Number 1.

Abstract

Ameloblastoma is a true neoplasm of odontogenic epithelial origin. It is the second most common odontogenic neoplasm, and only odontoma outnumbers it in reported frequency of occurrence.1 Its incidence, combined with its clinical behavior, makes ameloblastoma the most significant odontogenic neoplasm Unicystic ameloblastoma (UA) refers to those cystic lesions that show clinical, radiographic, or gross features of a mandibular cyst, but on histologic examination show a typical ameloblastomatous epithelium lining part of the cyst cavity, with or without luminal and/or mural tumor growth. 2 It accounts for 10-15% of all intraosseous ameloblastomas.3We would like to present such a case of unicystic ameloblastoma in a 28-year-old male at our institute.

CASE REPORT

A 28-year-old Hindu male, tailor by occupation, came with complaints of a swelling in the right lower jaw. The patient noticed the swelling incidentally, 2 months prior to presentation, when it was small with a size of approximately 2cm in diameter. It rapidly increased in the next 2 months to about 5cm in diameter.

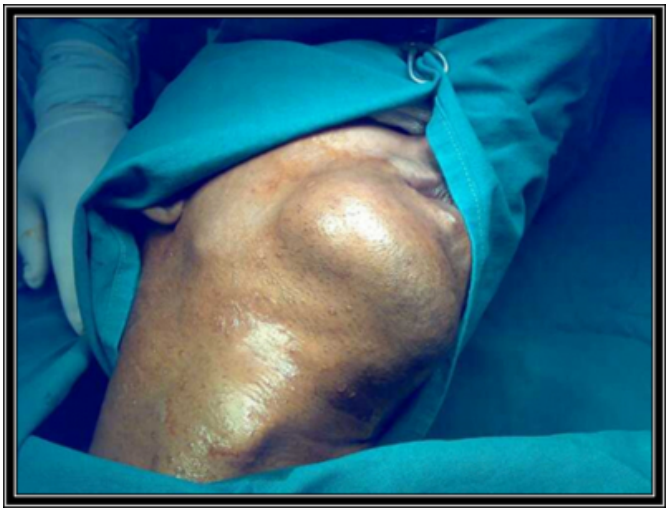
The patient did not give any history of local trauma. He used to consume tobacco in form of ‘gutka’ and also smoked over 10 cigarettes per day for the past 10 years. He did not have any other swelling in the neck. He had not undergone any dental procedures in the past. There was no history of any unerupted tooth in the past. He did not have any major medical or surgical illness in the past.

On examination, the patient had a solitary swelling of 5 x 4.5 x 1cm on the right lower mandible body extending from the right lower lateral incisor to the right lower first molar tooth. The skin overlying the swelling was normal with no dilated veins, scars or sinuses. The swelling was hard on the external surface but was tense and cystic on the intraoral mucosal surface. It appeared to have arisen from the bone. Overlying teeth were not loose. He had dental caries and poor oral hygiene, but no ulcers.

Right submandibular lymph nodes were palpable, but no other groups of cervical lymph nodes were palpable. All his blood investigations were within normal limits. He was not immunocompromised. A dental surgeon’s opinion was taken to consider for prosthesis but he advised it at a later date after mandible excision. So a decision to do segmental mandibulectomy with Kirschner wire (K-wire) fixation was taken.

Figure 1

Figure 1: Pre-operative view of mandibular tumor.



The patient was subjected to right lower segmental mandibulectomy with K- wire fixation of the ends. Intra-operative findings: a cystic swelling of 5 x 4.5 x 1.5cm was noted extending from the right lower lateral incisor to the right lower first molar, containing thick greenish viscous fluid. This segment was excised with an extra-oral approach.

Inter-dental wiring was done. The K-wire was fixed to bridge the gap between the two cut ends. The patient was extubated on the operation table. Post-operative recovery was uneventful.

At pathology, on gross examination, there was a cystic swelling 5 x 4.5 x 1cm with mucosal flap and 6 teeth. Cut section showed a cyst involving the body of the mandible containing sticky material.

Figure 2

Figure 2: Internal view of the specimen after segmental mandibulectomy.

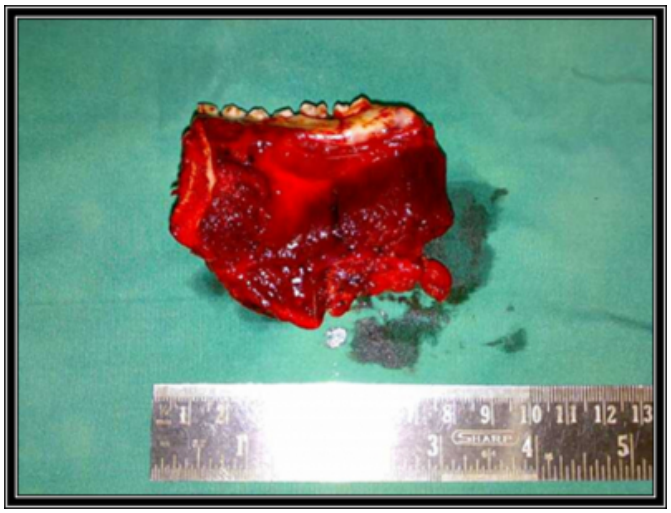
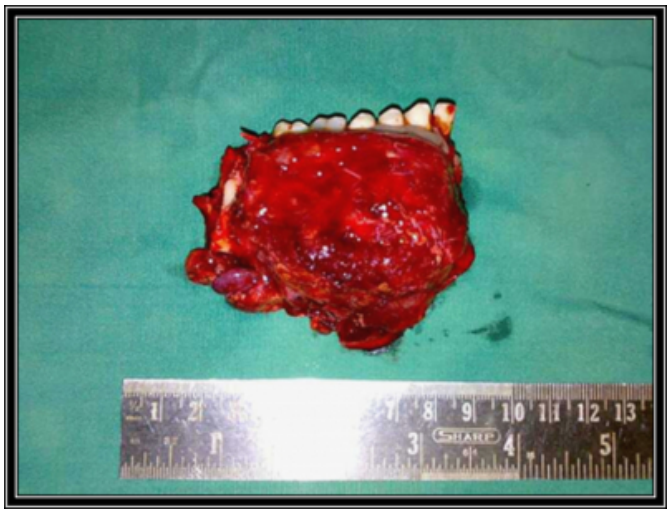


Figure 3

Figure 3: External view of the specimen after segmental mandibulectomy.



The histopathological report revealed a cystic mass lined by ameloblastic epithelium, showing basal cells palisading with reticulum cells above it. Focal squamous metaplasia was also noted. The diagnosis of a unicystic ameloblatoma was given.

The patient recovered well after surgery. He was started on nasogastric tube feeds. The drain was removed on the 4th post-operative day and sutures were removed on the 10th post-operative day. The patient's inter-dental wiring was removed after 4 weeks and he could open his mouth normally. He is scheduled for a dental prosthesis after complete healing of the wound after 3 months.

DISCUSSION

Ameloblastoma is divided into four types.[4]

- The classic solid/multicystic ameloblastoma (SMA) (83%).
- The UA (6%).
- The peripheral ameloblastoma (PA) (2%).
- The desmoplastic ameloblastoma (DA), including the so-called hybrid lesions (9%).

UA occurs in a younger age group, with slightly more than 50% of cases occurring in patients in the second decade of life. In more than 90% of cases, the UA is located in the mandible, with 77% located in the molar ramus region of the mandible. The mandible to maxilla ratio is 13:1.[5,6,7] Between 50 and 80% of cases are associated with tooth impaction, the mandibular third molar being most often involved. The 'dentigerous' type occurs 8 years earlier on average than the 'non-dentigerous' variant.

Patients most commonly present with chief complaints of swelling and facial asymmetry. Although the swelling is typically asymptomatic, pain is an occasional presenting sign. A chief complaint of painless swelling often indicates a lesion of long duration and significant size. Continued growth of the tumor and enlargement of the involved area may eventuate in ulceration of the mucosa overlying the lesion. Small lesions tend to be discovered more often on routine radiographic screening examinations or as a result of local effects produced by the tumor. Such local effects include tooth mobility, occlusal alterations and failure of eruption of teeth.[8]

Histologically, the minimum criterion for diagnosing a lesion as UA is the demonstration of a single cystic sac lined by odontogenic (ameloblastomatous) epithelium often seen only in focal areas. UA should be differentiated from odontogenic cysts because the former has a higher rate of

recurrence than the latter.[9] In a clinicopathologic study of 57 cases of unicystic ameloblastoma, Ackermann et al. classified this entity into 3 histologic groups: [3] 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).

Histologic subgrouping (modified after Ackermann et al.) by Philipsen and Reichart:[5]Subgroup 1: Luminal UA Subgroup 1.2: Luminal and intraluminal Subgroup 1.2.3: Luminal, intraluminal and intramural Subgroup 1.3: Luminal and intramural

The UAs diagnosed as subgroups 1 and 1.2 may be treated conservatively (careful enucleation), whereas subgroups 1.2.3 and 1.3 showing intramural growths must be treated radically, i.e., as a solid or multicystic ameloblastoma.[5] Vigorous curettage of the bone is discouraged since it may implant foci of ameloblastoma more deeply into bone. Chemical cauterization with Carnoy's solution is also advocated for subgroups 1 and 1.2. Subgroups 1.2.3 and 1.3, in which the cystic wall is involved with islands of ameloblastoma tumor cells and in which there is possible penetration into the surrounding cancellous bone, are thought to be associated with a high risk for recurrence, requiring more aggressive surgical procedures.[10,11,12]

The average interval of recurrence is 7 years. Recurrence is also related to histologic subtypes of UA, with those invading the fibrous wall having a rate of 35.7%, but others only 6.7%.[12]Recurrence rates were 3.6% for resection, 30.5% for enucleation alone, 16% for enucleation followed by Carnoy's solution application, and 18% by marsupialization followed by enucleation (where the lesion reduced in size) or resection.[13]

After resection, the patient's wires were removed after one month. He could masticate well all type of food products. He has been provided with dental prosthesis now after 3 months.

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