

Giant Ameloblastoma Flexiform (A Case Report)

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Abstract: Background: Ameloblastoma is a true neoplasm of odontogenic epithelial origin, that exhibits a locally aggressive behavior with a high level of recurrence. Patients usually present late after the tumour achieved considerable size to cause facial disfigurement. Purpose: To describe understanding, pathogenesis, complication, and the management of giant ameloblastoma in the mandible. Case: A 43 y.o female patient was referred to Oral and maxillofacial surgery department with swelling on the left mandible extend to left cheek region with 20x15x10 cm in size, redness, febris, pain, fluctuation with spontaneous drainage. This patient was diagnosed with ameloblastoma flexiform. Case Management: After confirmation diagnosis, The patient was performed Left Hemimandibulectomy. Recurrence was not detected during the follow-up period. Conclusion: Correct surgical planning is the key for successful management of large ameloblastoma, which is best treated using radical resection.

Keywords: Giant Ameloblastoma, Management, Hemimandibulectomy

1. Introduction

Ameloblastoma is an odontogenous tumor derived from dental epithelium and is benign but local invasive and has a high recurrence rate. Ameloblastoma was first introduced by the name of Adamantinoma by a French physician named Louis-Charles Malassez in 1885, but had previously been discovered by Cusack in 1827. Finally, the modern name of ameloblastoma in 1933 by Ivey and Churchill.¹ The jaw ameloblastoma known as adamantinoma is a clinically rare odontogenous tumor. The incidence rate of ameloblastoma is 1-3% of the tumor and cyst in the jaw. Ameloblastoma is generally cystic, slowly growing, and locally invasive and benign, no pain so usually found in the later stages and there has been swelling and bone destruction.² Some experts argue that ameloblastoma originates from a variety of causes, only the initial stimulation that causes the process of the occurrence of ameloblastoma is not known for certain.³ Causative factors may be caused by (1) non-specific irritant factors such as extraction, caries, trauma, infection, inflammation or tooth eruption; (2) deficit nutritional defects; and (3) viral pathogenesis.⁴

2. Case Report

A 43 y.o female patient was referred to Oral and maxillofacial surgery department with swelling on the left mandible extend to left cheek region with 20x15x10 cm in size, redness, febris, pain, fluctuation with spontaneous drainage. ± 15 years prior to admission the patient complained of swelling on the left lower jaw with marble sized. ± 10 years prior to admission the swelling got bigger with rambutan sized, then the patient got an operation because of the complaint. But the swelling still got bigger and increased look like papaya sized. ± 6 months prior to admission patient complained spontaneous pus drainage on the left cheek.



Figure 1

On Extraoral examination, found lumps on the left mandible extend to the mentalist region, with facial asymmetry, lump color equal to approximately, solid lump, fix, diameter 20x15x10 cm (Figure 1), well defined, border of the left mandible not palpable, and there is spontaneous drainage in the left mandible.



Figure 2

In the Intraoral examination, there a mass of buccal and lingual region extends up to posterior left mandible, palpable, solid, hard, there are parts that seem cystic, the lingual portion is the same color mass as around, the impression lobus, no pain was obtained.

CT scan 3D photographs of 8 January 2016 showed bone destruction from region 42 extending towards sinistra mandible border, region of symphysis, to condylus sinistra of mandible. There not tooth appearance 42, 41, 31, 32, 33, 34, 35, 36, 37, 38 (Fig. 3).

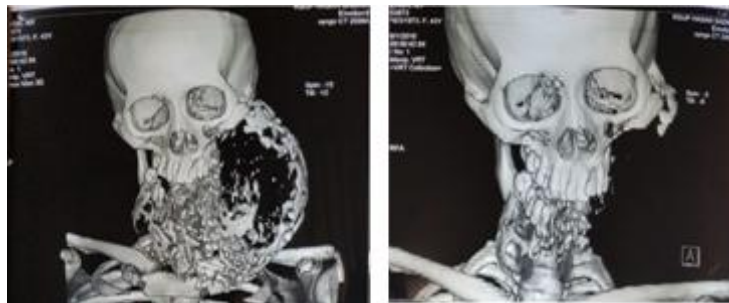


Figure 3

Biopsy results on sept 13, 2016, show as ameloblastomaflexiform ar left mandible. Patients were

planned to perform a left hemimandibulectomy and reconstruction with free fibula osteoseptocutaneous flap.



Figure 4

Recurrence was not detected during the follow-up period. It was found that patients had good esthetic and functional results after immediate reconstruction. The social activities of patients also were not affected.

from a developing enamel organ, from the epithelial lining of an odontogenic cyst, or from the basal cells of the oral mucosa. Ameloblastoma are slow-growing, locally invasive tumors.

Giant ameloblastoma has received very limited attention and reported in the literature.



Figure 5

A painless expansion of the jaws is the most common clinical presentation; neurosensory changes are uncommon, even with large tumors. Slow growth is the rule, with untreated tumors leading to tremendous facial disfigurement⁵. There was some report, Michael, et al. in 2010 reported a recurrent ameloblastoma of size 24cmx19cmx15cm in diameter. Hughes, et al from United States of America described a giant ameloblastoma which measured 15,2cm x11,4cm x12.0cm. Dunn, et al. Reported a giant ameloblastoma which measured 17cm x15cm x13 cm.⁶ In this case report, giant ameloblastoma was measured about 20cm x15cm x10 cm in size.

3. Discussion

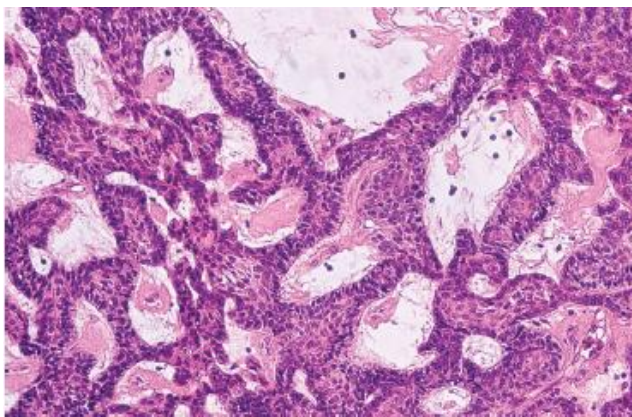
Ameloblastomas are tumors of odontogenic epithelial origin. Theoretically, they may arise from rests of dental lamina,

Table 2: A review of clinical features, tumor size, and histopathological type of giant ameloblastomas that were previously reported and the present case.

SL. no	Year	Age	Sex	Tumor size	Histopathological type
1	1977	57	M	Mental region to ramus of mandible	Follicular ameloblastoma
2	1977	62	F	Right wisdom tooth to left 1st premolar tooth region	Plexiform ameloblastoma
3	1985	30	F	15 × 14 × 12 cm	Plexiform ameloblastoma
4	1990	33	M	Right cuspid tooth to left mandibular condyle	Follicular ameloblastoma
5	1991	39	F	11 × 10 × 6 cm	Plexiform ameloblastoma
6	1995	73	M	11 × 11 × 14 cm	Plexiform ameloblastoma
7	1997	62	F	17 × 15 × 13 cm	Plexiform ameloblastoma
8	1999	53	F	15.2 × 11.4 × 12 cm	Plexiform ameloblastoma
9	2005	53	M	14 × 11 × 10 cm	Follicular ameloblastoma
10	2011	35	F	15 × 12 × 10 cm	Plexiform ameloblastoma
11	Present case	39	F	12 × 9 × 10 cm	Granular cell ameloblastoma

Figure 6

In the 2005 World Health Organization classification the benign ameloblastoma is divided into 1) solid/multicystic, 2) extra-osseous/peripheral, 3) desmoplastic, and 4) unicystic.⁷ The solid/multicystic ameloblastoma can histopathologically be divided into a follicular and a plexiform type. The plexiform type contains basal cells arranged in anastomosing strands with an inconspicuous stellate reticulum. The stroma is usually delicate, often with cystlike degeneration.

**Figure 7**

The plexiform type of ameloblastoma consists of long, anastomosing cords or larger sheets of odontogenic epithelium. The cords or sheets of epithelium are bounded by columnar or cuboidal ameloblast-like cells surrounding more loosely arranged epithelial cells. The supporting stroma tends to be loosely arranged and vascular. Cyst formation is relatively uncommon in this variety. When it occurs, it is more often associated with stromal degeneration rather than cystic change within the epithelium.⁸ 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.⁹ According to World Health Organization (WHO) ameloblastoma is a benign tumor. Metastases to the lungs or central nervous system (CNS) have rarely been reported. Complications of ameloblastoma include the following:

- Breathing difficulty
- Pain and facial deformity
- Secondary infection of the tumor

- The recurrence rate of these tumors is 25-30%. The tumor can recur after treatment and hence, a close follow-up is needed.

Treatment of ameloblastoma is primarily surgical. There has been some debate regarding the most appropriate method of surgical removal of ameloblastomas. These ranges from conservative to radical modes of treatment. The conservative modalities include curettage, enucleation and cryosurgery, while the radical modalities are marginal, segmental, and Hemimandibulectomy.¹⁰ Because of their tendency to be more aggressive than the other types and has a higher incidence of recurrence, the treatment are with a radical approach for these tumours. Resected all the tumours with a margin of at least 2 cm of healthy bone. Because of the giant size of these lesions all our cases had huge defect of soft tissue and bone post resection, we have used free fibular flap for reconstruction of these defects, free fibular flap offers lot of advantages in mandibular reconstruction.

In this cases, A 43 y.o female patient was referred to Oral and maxillofacial surgery department with swelling on the left mandible extend to left cheek region with 20x15x10 cm in size, redness, febris, pain, fluctuation with spontaneous drainage. Patient was performed a left hemimandibulectomy and reconstruction with free fibula osteoseptocutaneous flap. Recurrence was not detected during the follow-up period. It was found that patients had good esthetic and functional results after immediate reconstruction.

4. Conclusion

Ameloblastoma is a true neoplasm of odontogenic epithelial origin, that exhibits a locally aggressive behavior with a high level of recurrence. 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.⁴ Because of their tendency to be more aggressive than the other types and has a higher incidence of recurrence, the treatment are with a radical resection.

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