Recurrent Ameloblastoma in the Free Fibula Flap: Review of Literature and an Unusual Case Report

Salih Onur Basat · Ali Rıza Öreroğlu ·
Cagdas Orman · Tolga Aksan · İlker Üşçetin ·
Mithat Akan

Received: 12 November 2013 / Accepted: 15 September 2014 / Published online: 4 January 2015
© The Association of Oral and Maxillofacial Surgeons of India 2014

Abstract Ameloblastoma is the second most common odontogenic tumor of the oral cavity with the primary site being the mandible. The ratio of maxillomandibular involvement however is 5:1 in favor of the mandible. The most common complaint is a painless swelling over the mandibular area. Despite its benign nature, ameloblastoma has a high local recurrence rate, with the most recurrences seen within 5 years after operation. Biopsy and radiological evaluation may be helpful in differentiating the subtypes of ameloblastoma. Differentiation is important because some subtypes are more aggressive than the others. Preoperative planning may be done according to this classification, which can help decrease the recurrence rate. In our case, a 26-year-old female patient with recurrent ameloblastoma which developed on the fibular flap is presented. The free fibular flap and the left parasymphiseal part of the mandible were totally excised. Ameloblastoma was confirmed on pathological examination. We reconstructed the left mandibular site with a reconstruction plate and recurrence was not seen during follow up period.

Keywords Recurrent ameloblastoma · Free fibular flap · Reconstruction of mandible · Ameloblastoma classification

Introduction

Ameloblastoma is a rare benign odontogenic tumor [1], with its name derived from the early English word “amel” meaning enamel and the Greek word “blastos” meaning germ [2]. Ameloblastoma is an odontogenic tumor of the jaws, derived from the dental embryonic remnants possibly from the epithelial lining of an odontogenic cyst, dental lamina or enamel organ, stratified squamous epithelium of the oral cavity or displaced epithelial remnants [3].

Ameloblastoma was first described by Cusack [4]. Malassez [5] introduced another name “adamantinoma” that is now used to name a rare form of bone cancer. Churchill [6] renamed the ameloblastoma to its widely used denomination.

Ameloblastoma represents 1 % of all tumors and cysts that involve the maxillomandibular area and about 10 % of the odontogenic tumors [7]. The ratio of maxillomandibular involvement is 5:1 in favor of the mandible, and the most common site of occurrence is the mandibular molar region [8, 9]. It is primarily seen in adults in the third decade of life, with equal sex predilection [7].

Because of the locally aggressive behavior, ameloblastoma has a high recurrence rate [10]. More than 50 % of the recurrences occur within the first 5 years after primary surgery [8]. According to the World Health Organization (WHO) classification, a distinction has been made between ameloblastoma, malignant ameloblastoma and ameloblastic carcinoma [10]. Malignant ameloblastoma differs from ameloblastoma by the presence of metastases. However, they both possess the same benign histology [11]. Ameloblastic carcinoma has malignant cytological features regardless of the presence of metastases. In ameloblastoma, metastases are uncommon. When they occur, lungs are the most frequently site involved [12].

S. O. Basat · C. Orman (✉) · T. Aksan · İ. Üşçetin
Plastic, Reconstructive and Aesthetic Surgery Clinic,
Okmeydani Training and Research Hospital, Darulaceze cad.
No: 25 Şişli, Istanbul, Turkey
e-mail: cagdasorman@gmail.com

A. R. Öreroğlu
Acibadem Fulya Hospital, Istanbul, Turkey

M. Akan
Department of Plastic, Reconstructive and Aesthetic Surgery,
Medipol University, Istanbul, Turkey

Fig. 1 Preoperative, 26-year-old woman with recurrent ameloblastoma

Case Report

Our case is a 26-year-old female with a complaint of swelling on the left side of the mandible in May 2012. The history revealed initial presentation of the swelling in 1998 with cyst curettage done in another medical center. Upon relapse, curettages had been repeated in 1999 and 2000 at the same center however the treatment was not successful. The patient did not want any other operation until 2005, after which she had been admitted to the Medical Faculty of Istanbul with swelling on the left hemimandibular area where hemimandibulectomy and simultaneous reconstruction with free fibular flap had been performed.

Our examination of the patient revealed a hard and painless tumor that extended from the left parasymphiseal area of the mandible involving the body and the ramus of the mandible and further extending into the temporal region. Intraoral examination revealed severe expansion of both lingual and buccal cortices of the mandible without any mucosal destruction (Fig. 1).

CT evaluation revealed a well-defined unilocular radiolucency extending from the left parasymphiseal region to the left glenoid fossa. The tumor mass extending under the zygomatic arch also resulted in displacement of the zygomatic arch in the anterior and superior direction. The free fibular flap was totally disrupted by the cystic lesion. The miniplates and screws that had been used for fixation of the free fibula to the mandible were located over the cortical bone and the medullar part of the bone was totally filled with the lesion (Fig. 2).

The patient was operated again in May 2012. The free fibular flap and the left parasymphiseal part of the mandible were totally excised. Ameloblastoma was confirmed on pathological examination, reporting a macroscopic $8 \times 5 \times 5$ cm size unilocular cystic lesion (Fig. 3a). The

microscopic evaluation reported no tumoral invasion at both the soft tissue and bone margin. The patient did not choose the reconstruction with free fibula because of fear of disease recurrence. We reconstructed the left mandibular site with a reconstruction plate (Figs. 3b, 4).

Discussion

After odontoma, ameloblastoma is the second most common odontogenic neoplasm of the oral cavity [10]. The average age of patients with ameloblastoma is 34 years with a range of 5–74 years [13]. The most common complaint at time of diagnosis is a painless swelling over the mandibular area [14].

In radiological terms, ameloblastoma can be a unilocular or multilocular radiolucent lesion with a honeycomb or soap bubble appearance [15]. There are several subtypes including the plexiform, follicular, acanthomatous, basaloid, granular cell, cystic and desmoplastic forms. The follicular entity is the most common form. There are also various clinical types of ameloblastoma. These are the solid or multicystic type, unicystic type, malignant type and the rare peripheral type [16]. Peripheral ameloblastoma involves the gingival and alveolar mucosae, infiltrating the gingival connective tissue specially. The peripheral variant however does not involve the underlying bone [17].

Clinical and histopathological classification of ameloblastoma is the mainstay concept for surgical treatment. According to current data, unicystic and peripheral subtypes have a better prognosis than the solid type after a limited surgical treatment [18].

The current surgical options for treatment of ameloblastoma extend from curettage to radical excision. According to recent studies on unicystic ameloblastoma,

Fig. 2 Computerized tomography of coronal and axial plane. Expansion of ameloblastoma

Fig. 3 Excised ameloblastoma (a) and intra-operative photos (b)

infiltration of the tumor can be seen within 0.25 cm of bone margin with no infiltration seen after 0.50 cm. The safe surgical margin for follicular subtype is 0.75 cm but in granular ameloblastoma infiltration can also be seen up to 0.75 cm of bone margin [16]. The clinical and pathological classification of ameloblastoma is therefore of great importance for correct surgical planning.

Ameloblastoma is a tumor with well-known propensity for recurrence [19]. There is a 50–72 % incidence of local

recurrence after initial therapy [20]. Several factors can influence this rate with the first and the most important one being the clinicopathologic variant of the tumor. The solid or multicystic variant has the highest recurrence potential because of the great propensity for local infiltration [14, 20, 21]. The second factor is the anatomic localization of the tumor. The most common anatomic location for ameloblastoma is the mandible; which has a dense cortical bone structure that prevents spreading for several years [22]. The

Fig. 4 Postoperative 10-month follow up photos

tumor can however extend beyond the radiological margin by spreading through the central cancellous bone [23]. The third factor contributing to recurrence is the inadequacy of surgery.

Radical surgical excision is the more proper treatment solution for recurrent ameloblastoma. Tumoral excision and mandibular reconstruction with bone graft is the preferred choice for recurrent cases by most maxillofacial surgeons. Some recurrences of ameloblastoma over bone grafts have been reported in the medical literature [22]. In these cases one should ask himself “whether a more radical excision could have been done” to prevent the recurrence. Recurrence of the ameloblastoma over a free fibular flap however is very rare.

Conclusion

In our case, radical excision (hemimandibulectomy) had been performed with reconstruction of the mandibular with a free fibula flap. In spite of these radical interventions, relapse of the tumor has been observed 7 years after radical excision. Histopathological and clinicopathological classification reveals a less aggressive type than the other subtypes but relapse has been observed despite the radical excision. This case may be suggestive in that different factors may contribute to the relapse of ameloblastoma other than what we know today.

References

1. Amzerin M, Fadoukhair Z, Belbaraka R, Iraqui M, Boutayeb S, M'rabti H, Kebdani T, Hassouni K, Benjaafar N, El Gueddari BK, Errihani H (2011) Metastatic ameloblastoma responding to combination chemotherapy: case report and review of the literature. *J Med Case Rep* 3(5):491
2. Brazis PW, Miller NR, Lee AG, Holliday MJ (1995) Neuro-ophthalmologic aspects of ameloblastoma. *Skull Base Surg* 5(4):233–244
3. Avon SL, McComb J, Clokie C (2003) Ameloblastic carcinoma: case report and literature review. *J Can Dent Assoc* 69(9):573–576
4. Cusack JW (1827) Report of the amputation of the lower jaw. *Dublin Hosp Rec* 4:1–38
5. Malassez L (1885) Sur Le rôle des debris epitheliaux papdentaires. *Arch Physiol Norm Pathol* 5:309–340, 6:379–449
6. Ivery RH, Churchill L (1930) The need of a standardized surgical and pathological classification of tumors and anomalies of dental origin. *Am Assoc Dent Sch Trans* 7:240–245
7. Chauhan DS, Guruprasad Y (2011) Plexiform ameloblastoma of the mandible. *J Clin Imaging Sci* 1:61
8. Reichart PA, Philipsen HP, Sonner S (1995) Ameloblastoma: biological profile of 3677 cases. *Eur J Cancer B Oral Oncol* 31B(2):86–99
9. Kim SG, Jang HS (2001) Ameloblastoma: a clinical, radiographic, and histopathologic analysis of 71 cases. *Oral Surg Oral Med Oral Pathol Oral Radiol Endod* 91(6):649–653
10. Jordan RCK, Speight PM (2009) Current concepts of odontogenic tumours. *Diagn Histopathol* 15(6):303–310
11. Ciment LM, Ciment AJ (2002) Malignant ameloblastoma metastatic to lungs 29 years after primary resection. *Chest* 121:1359–1361
12. Henderson JM, Sonnet JR, Schlesinger C, Ord RA (1999) Pulmonary metastasis of ameloblastoma: case report and review of the literature. *Oral Surg Oral Med Oral Pathol Oral Radiol Endod* 88(2):170–176
13. Gilijsamse M, Leemans CR, Winters HA, Schulten EA, van der Waal I (2007) Metastasizing ameloblastoma. *Int J Oral Maxillofac Surg* 36(5):462–464
14. Gardner DG (1994) Controversies in the nomenclature, diagnosis and treatment of ameloblastoma. In: Worthington P, Evans JR (eds) *Controversies in oral and maxillofac surgery*. Saunders, Philadelphia, p 362
15. Tozaki m, Hayashi K, Fukuda K (2001) Dynamic multislice helical CT of maxillomandibular lesion: distinction of ameloblastoma from other cystic lesions. *Radiat Med* 91:649–653
16. Rastogi V, Pandilwar PK, Maitra S (2010) Ameloblastoma: an evidence based study. *J Maxillofac Oral Surg* 9(2):173–177
17. Feinberg SE, Steinberg B (1996) Surgical management of ameloblastoma. Current status of the literature. *Oral Surg Oral Med Oral Pathol Oral Radiol Endod* 81(4):383–388

18. Gardner DG (1996) Some current concepts on the pathology of ameloblastoma. *Oral Surg Med Oral Pathol Oral Radiol Endod* 82(6):660–669
19. Olaitan AA, Arole G, Adekey EO (1998) Recurrent ameloblastoma of jaw: a follow-up study. *Int J Oral Maxillofac Surg* 27:456–460
20. William TP (1993) Management of ameloblastoma: A changing perspective. *J Oral Maxillofac Surg* 51:1064–1070
21. Adekeye EO, Lavery KM (1986) Recurrent ameloblastoma of the maxillofacial region. *J Maxillofac Surg* 14:153–157
22. Collings SJ, Harrison A (1993) Recurrent ameloblastoma? A historic case report and a review of literature. *Br Dent J* 174:202–206
23. Gardner DG, Pecak AM (1980) The treatment of ameloblastoma based on pathologic and anatomic principle. *Cancer* 46:2514–2519