https://doi.org/10.36570/jduhs.2024.2.1981

JDUHS

CASE REPORT

Delayed Recurrence of Mandibular Ameloblastoma in Soft Tissues of Neck: A Case Report of a Rare Entity

Nabeela Riaz, Samreen Younas

Department of Oral and Maxillofacial Surgery at King Edward Medical University/Mayo Hospital Lahore, Pakistan.

Correspondence to: Dr. Samreen Younas Email: samreenyounas1@gmail.com, ORCiD: 0009-0000-6666-3913

ABSTRACT

Ameloblastoma is the second most common odontogenic neoplasm and is locally aggressive. Various treatment approaches exist, with resection involving safe margins associated with the lowest recurrence rates. However, ameloblastoma still has a propensity for delayed recurrences, necessitating lifelong follow-up for patients. Recurrences usually develop at the primary site of the tumor but have also been reported in reconstructive bone grafts. We present a unique case of a very unusual, delayed recurrence of mandibular ameloblastoma in the soft tissues of the neck, 16 years after the primary surgery. The recurrence presented as a large cystic swelling in the neck with a solid component, posing a diagnostic challenge for the team. The final histopathological examination after excision under general anesthesia confirmed it to be ameloblastoma.

Keywords: Ameloblastoma, Local Neoplasm Recurrence, Odontogenic Tumors.

INTRODUCTION

Ameloblastoma is a benign, locally aggressive neoplasm of odontogenic epithelial origin. It affects both males and females equally and is most common in patients aged 30 to 60 years. The mandible is more commonly affected than the maxilla. According to the World Health Organization (2017), ameloblastoma is classified into three types: conventional ameloblas-toma (solid/multicystic), unicystic ameloblas-toma, and extraosseous/peripheral ameloblastoma.[1] The mandible is the most frequent site for ameloblastoma, with 80% of tumors occurring there and only 20% in the maxilla. The solid/multicystic type of ameloblastoma is associated with much higher recurrence rates.[2]

The primary management of ameloblastoma remains surgical, despite many emerging therapies. Radical surgery is associated with higher recurrence-free survival rates. Patients require lifelong screening due to the propensity of ameloblastoma for delayed recurrences.[3] Recurrences mostly develop at the site of the primary tumor, but cases of recurrence in soft tissues and even in reconstructive bone grafts are rare. The management of recurrent ameloblastoma presents a surgical challenge.[4]

We present a rare case of recurrent ameloblastoma in the neck soft tissues 16 years after surgical resection of mandibular solid ameloblastoma. To our knowledge, no such case has been reported in the literature where a large soft tissue recurrence presented in the neck, well away from the initial tumor site.

CASE REPORT

A 32-year-old female patient presented to the outpatient department of Oral and Maxillofacial Surgery in May 2023 with a large swelling on the right lateral side of her neck, which had developed about two months prior (Figure 1). The patient reported a history of developing a swelling on the right side of her lower jaw 16 years ago, at the age of 16. She was diagnosed with ameloblastoma at a tertiary care hospital following an incisional biopsy. She underwent tumor resection with safe margins-disarticulation on the right side and resection up to the second premolar region on the contralateral side-under general anesthesia in 2007. The mandibular bone defect was reconstructed with a free fibula flap by a plastic surgery team in 2017.

Sixteen years after the initial surgery, the patient developed a swelling on the right side of her neck. The swelling measured 13 x 7 cm in cranio-caudal and transverse dimensions, extending from the supra-clavicular region to the submandibular region. It was soft to firm in consistency, with preserved color and texture of the overlying skin. Aspiration revealed dark straw-colored fluid. There was no restriction of mouth

opening, and no significant findings on intraoral examination.

Computed tomography (CT) revealed a large cystic lesion with thick walls, causing a mass effect on the carotid sheath components and significantly compressing the internal jugular vein (Figure 2). Excision of the lesion was planned under general anesthesia. The patient's vital parameters and baseline investigations were within normal ranges. The lesion was approached using a horizontal neck crease incision, raising the skin flaps in the subplatysmal plane (Figure 3). Intraoperatively, a solid component of the tumor was found extending superiorly to the mastoid bone from the superior pole of its cystic component, prompting a modification in the initial incision. Resection of the tumor necessitated ligation of the internal jugular vein, as it was inseparable from the tumor. The tumor was delivered en bloc without puncturing its cystic component, incorporating a small area of overlying skin (Figure 4). All vital structures in the region were preserved, and the skin was closed primarily after achieving hemostasis and securing a suction drain in place.

Histopathological examination of the lesion revealed a neoplasm composed of islands of stellate reticulum with peripheral palisading epithelium, with focal areas of granular cells and fibrotic walls in the cystic component, confirming the diagnosis of recurrent ameloblastoma. The patient had been recurrence-free until the last follow-up, seven months post-surgery, with no functional deficits observed.

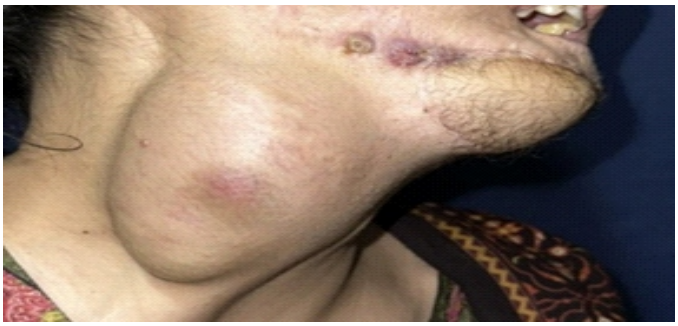

Figure 1: A large cystic swelling at right side of neck

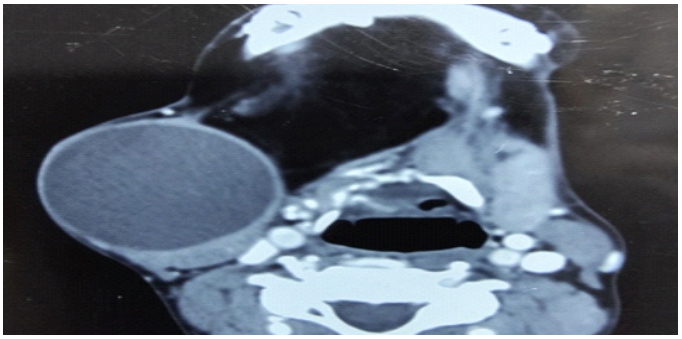

Figure 2: C.T showing a well-defined cystic lesion

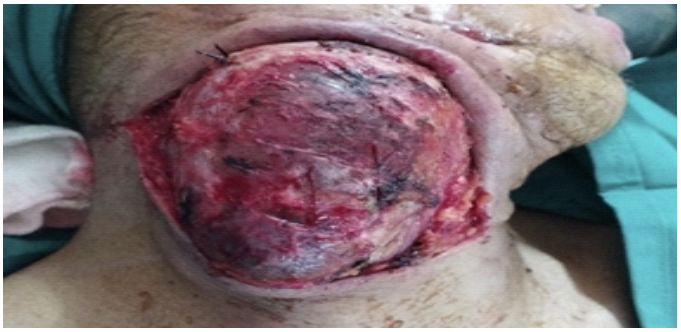

Figure 3: Surgical exposure of the lesion using horizontal neck incision

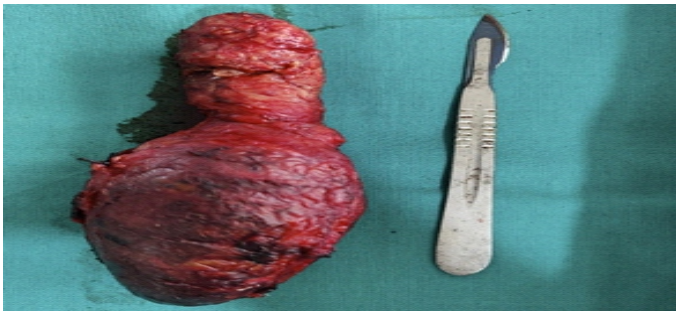

Figure 4: Excised specimen

DISCUSSION

Ameloblastoma is the second most common odontogenic neoplasm after odontoma, being locally aggressive and rare malignant transformations have also been reported. It is more commonly reported in developed countries than the developing countries.[5]

The mean age of presentation is between 3rd and 6th decades of life. In the present case report the patient was diagnosed for ameloblastoma at an age of 16 years which is younger than the peak age.

The extent of surgical resection of the tumor is debatable and ranges from local curettage and conservative management to wide resection with 1-3cm margins. The reported rate of recurrence after conservative management of amelolastoma ranges from 55%-90% for solid and multi-cystic tumors. Therefore the treatment of choice for solid variant of ameloblastoma is wide surgical resection to minimize the rate of recurrence.[6] In a retrospective study conducted on 158 patients local recurrence rate of 13.2% was recorded and 10.76% of the recurrences were recorded within first 5 year of follow-up period. It was concluded that choice of treatment modality has a significant relation with the rate of recurrence and resection with adequate safe margins remains the mainstay of treatment as the tumor cells may extend 5-8mm away of radiographic extent of disease.[7] Another study suggested that majority of the recurrences present within first 5 year, however may present up to

10-15 years after the initial treatment. The main factors which were identified contributing to recurrence in this study were conservative treatment, follicular histopathological pattern and multi-locular type of ameloblastoma.[8] In the present case the tumor was managed with wide surgical resection and presented with recurrence in soft tissues after 16 years of surgery. A similar case presented with recurrence in the reconstructive graft material and surgical bed after 17 year of initial treatment for ameloblastoma.[9]

Ameloblastoma is a disease that needs lifelong surveillance. Although most of the recurrences are present during the first 5 years of treatment but late recurrences are not uncommon as in the present case report. The reported rate of developing ameloblastic carcinoma and malignant ameloblastoma is about 2%.[10] The three possible explanations for the recurrences include; recurrence from the bony stumps left after resection, recurrence from the adjacent soft tissues and from the tumor cells seeding in surgical field during the primary surgery.[11] The most acceptable explanation for the present case is seeding of the tumor cells intra-operatively as the recurrence was well away from the original site of the tumor excluding the first and second possibilities.

The recurrences that appear asymptomatic at the initial stages may lead to extensive destruction, therefore the pivotal role of regular long-term follow-up should not be understated. Clinical examinations should be augmented with appropriate radiographs. I has been suggested that special measures should be taken while treating the recurrences to prevent further recurrence. In the present case the tumor was excised en-block and a small area of overlying skin was also removed to minimize chances of further recurrence.

CONCLUSION

The reported case highlights the importance of en bloc resection of ameloblastoma to minimize chances of tumor cell seeding followed by thorough irrigation of surgical field to reduce chances of tumor cells implantation and long term follow up.

LIMITATION: The patient was initially operated at a different center and the exact information of previous surgery was not available.

PATIENT'S CONSENT: The authors certify that they have obtained all appropriate patient consent forms. In the form patient has given her consent for her clinical information to be reported in journal. The patient understands that her name and initials will not be published.

CONFLICT OF INTEREST: No

Received: October 12, 2023
Accepted: May 15, 2024

REFERENCES

1. Cadavid AM, Araujo JP, Coutinho-Camillo CM, Bologna S, Junior CA, Lourenco SV. Ameloblastomas: current aspects of the new WHO classification in an analysis of 136 cases. Surg Exp Pathol 2019; 2:1-6. doi:10.1186/s42047-019-0041-z
2. Neagu D, Escuder-de la Torre O, Vazquez-Mahia I, Carral-Roura N, Rubin-Roger G, Penedo-Vazquez A, et al. Surgical management of ameloblastoma. Review of literature. J Clin Exp Dent 2019; 11:e70-e5. doi:10.4317/jced.55452
3. Milman T, Ying GS, Pan W, LiVolsi V. Ameloblastoma: 25 Year Experience at a Single Institution. Head Neck Pathol 2016; 10:513-20. doi:10.1007/s12105-016-0734-5
4. Effiom OA, Ogundana OM, Akinshipo AO, Akintoye SO. Ameloblastoma: current etiopathological concepts and management. Oral Dis 2018; 24:307-16. doi:10.1111/odi.12646
5. Aramanadka C, Kamath AT, Kudva A. Recurrent Ameloblastoma: A Surgical Challenge. Case Rep Dent 2018; 8271205. doi:10.1155/2018/8271205
6. Yang R, Liu Z, Gokavarapu S, Peng C, Ji T, Cao W. Recurrence and cancerization of ameloblastoma: multivariate analysis of 87 recurrent craniofacial ameloblastoma to assess risk factors associated with early recurrence and secondary ameloblastic carcinoma. Chin J Cancer Res 2017;29:189-95.doi:10.21147/j.issn.1000-9604.2017.03.04
7. Bi L, Wei D, Hong D, Wang J, Qian K, Wang H, et al. A Retrospective Study of 158 Cases on the Risk Factors for Recurrence in Ameloblastoma. Int J Med Sci 2021; 18:3326-332. doi:10.7150/ijms.61500
8. Ajila V, Hegde S. Ameloblastomas vs recurrent ameloblastomas: A systematic review. J Oral Med Oral Surg. 2022; 28:11. doi:10.1051/mbcb/2021044
9. Hamdy O, Raafat S, Saleh GA, Awny S, Shebl AM, Hegazy MA. Recurrent mandibular ameloblastoma in soft tissue and rib graft 17 years after initial presentation. J Egypt Natl Canc Inst 2020; 32:1. doi:10.1186/s43046-019-0012-1
10. McClary AC, West RB, McClary AC, Pollack JR, Fischbein NJ, Holsinger CF, et al. Ameloblastoma: a clinical review and trends in management. Eur Arch Otorhinolaryngol 2016; 273:1649-61. doi:10.1007/s00405-015-3631-8
11. Adebayo ET, Fomete B, Adekeye EO. Delayed soft tissue recurrence after treatment of ameloblastoma in a black African: case report and review of the literature. J Craniomaxillofac Surg 2011; 39:615-8. doi:10.1016/j.jcms.2010.05.010