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A single cervical lymph node metastasis of malignant ameloblastoma

Yoori Kim [a], Sung-Weon Choi [b], Jong-Ho Lee [c], Kang-Min Ahn [a, *]

[a] Department of Oral and Maxillofacial Surgery, College of Medicine, University of Ulsan, Asan Medical Center, Seoul, South Korea
[b] Oral Oncology Clinic, National Cancer Center, Gyeonggi-do, South Korea
[c] Department of Oral and Maxillofacial Surgery, School of Dentistry, Seoul National University, Seoul, South Korea

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ABSTRACT

Introduction: Cervical node metastasis of malignant ameloblastoma is extremely rare. Because of its rarity, there is no standard treatment modality in a single lymph node metastasis in malignant ameloblastoma.
Materials and methods: Eleven patients of malignant ameloblastoma involving a single cervical lymph node metastasis and one new case were reviewed. Neck treatment was classified into neck dissection and simple excision. Local nodal recurrence, distant metastasis and follow-up periods were investigated.
Results: Eight patients were treated with neck dissection (group A) and four patients underwent a simple node excision (group B). Two patients in group A experienced multiple organ metastases such as liver and lung seven months and 13 years after neck dissection respectively. The other patients showed no recurrence and metastasis. In group B, there was no report of a regional neck recurrence and distant metastasis during follow-up of 1–7 years.
Conclusion: Multiple nodes metastasis requires a radical neck dissection; however, simple excision with close follow-up may be used in a single node metastasis in malignant ameloblastoma.

1. Introduction

Ameloblastoma is a slowly growing, benign, but locally invasive odontogenic neoplasm with a high recurrence rate if the initial lesion is treated by simple enucleation or curettage (Shin et al., 2011). Despite the fact of its benign histologic characteristics, ameloblastoma has the ability to develop metastasizing lesions in organs such as the lungs (Chou et al., 2013), cervical lymph nodes (Park et al., 2003; Jayaraj et al., 2013), spine (Nguyen, 2005), myocardium (Zwahlen et al., 2003), skull (Hayashi et al., 1997), kidney (Hayakawa et al., 2004) and skin (White and Patterson, 1986). The two malignant counterparts of ameloblastoma are malignant ameloblastoma and ameloblastic carcinoma, and they compose less than 1% of all ameloblastomas. Malignant ameloblastoma is defined by the World Health Organization as an ameloblastoma which has metastasized, and the metastatic lesion must maintain the same benign histopathologic feature as the primary tumor. On the other hand, ameloblastic carcinoma is an ameloblastic tumor characterized by distinct cytologic atypia, which can show microscopic evidence of malignancy, regardless of whether it has metastasized. Ameloblastic carcinoma also presents aggressive clinical characteristics which include rapid growth, perforation of the cortex, and painful swelling (Bruce and Jackson, 1991; Ryu et al., 2002).

Treatment of the metastatic lymph node in malignant ameloblastoma is controversial because of the rarity of cases. There have only been 23 articles reported since 1928 about ameloblastoma with lymph node metastasis (Simmons, 1928; Masson et al., 1959; Eda et al., 1972; Ikemura et al., 1972; Brandenburg et al., 1976; Lanham, 1987; Ueda et al., 1989; Houston et al., 1993; Duffey et al., 1995; Takeda, 1996; Witterick et al., 1996; Narozny et al., 1999; Sugiyama et al., 1999; Verneuil et al., 2002; Goldenberg et al., 2004; Gilijamse et al., 2007; Cardoso et al., 2009; Dao et al., 2009; Reid-Nicholson et al., 2009; Dissanayake et al., 2011; Bansal et al., 2012; Golubovic et al., 2012; Jayaraj et al., 2013). Goldenberg et al. (2004) and Dissanayake et al. (2011) suggested a radical neck dissection for cervical lymph node metastasis; however, Houston et al. (1993) and Dao et al. (2009) performed simple lymph node excision and reported no recurrence during their follow-up period.

The purpose of this review is to suggest a treatment modality of a single cervical lymph node metastasis in malignant ameloblastoma and to report our experience of simple lymph node excision with ten years follow-up.

* Corresponding author. Department of Oral and Maxillofacial Surgery, College of Medicine, University of Ulsan, Asan Medical Center, 88 Olympic-ro, 43-gil, Songpa-gu, Seoul, 137-736, South Korea. Tel.: +82 2 3010 5901; fax: +82 2 3010 6967.
E-mail address: ahnkangmin@hanmail.net (K.-M. Ahn).

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2. Materials and methods

2.1. Review of the literature

2.1.1. Search strategy and study selection

A computerized literature search on the subject of malignant ameloblastoma was conducted focusing on the database of Medical Literature Analysis and Retrieval System online (MEDLINE) from 1920 to 2013. The searching term was “malignant ameloblastoma” or “metastatic ameloblastoma” or “metastasizing ameloblastoma” or “malignant odontogenic tumor” or “adamantinoma”. Inclusion and exclusion criteria were as followed.

2.1.2. Inclusion criteria

Ameloblastoma with metastasis to a single cervical lymph node was included in this analysis. The additional inclusion criteria for the analysis were;

- Publications in the English literature
- Studies with follow-up periods and survival
- Studies with clearly defined surgical methods such as neck dissection or neck node excision
- Studies with live patients

2.1.3. Exclusion criteria

The reasons for exclusion were as followed:

- Non-English literature
- Ameloblastic carcinoma
- Other organ metastasis such as lung, liver, kidney, skin and spleen
- Multiple cervical lymph node metastasis
- No treatment due to patient's general condition
- Autopsy case
- Chemotherapy as a main treatment modality
- No follow-up periods and no report about survival

2.2. New case

In June 2004, a 20-year-old female was referred from a local clinic to the department of Oral and Maxillofacial Surgery with a complaint of painless swelling of the right mandible. The patient has been de-identified and approval from the Institutional Review Board (IRB) of Seoul Asan Medical Center was obtained. The patient denied any significant medical history existed beforehand. A computed tomographic scan (CT) depicted multilocular, radiolucent lesions with buccolingual expansion (Fig. 1A, B), involving the right premolar to molar area which was diagnosed as an ameloblastoma by incisional biopsy.

Different treatment options along with the advantages versus disadvantages of each option were presented to the patient. To decrease the possibility of recurrence, segmental mandibular resection surgery is recommended initially, but the patient chose to receive a more conservative treatment considering the young age of the patient. Thus, the patient underwent extraction of right lower canine to third molar, right marginal mandibulectomy with preservation of the inferior border and a free iliac bone reconstruction surgery under general anesthesia (Fig. 2). Biopsy confirmed the diagnosis of ameloblastoma (Fig. 3).

During the 2.5 years of follow up, no evidence of recurrence was found in the primary site. In January 2007, the patient presented for a regular annual check-up regarding neck swelling on the right side with discomfort. Clinical examination revealed one soft lump in the right submandibular region. CT of the neck region showed a well-demarcated submandibular node swelling which was suspected as a reactive lymph node or a metastatic lymph node (Fig. 4).

Ultrasono-guided fine needle aspiration biopsy was performed to confirm that there were no malignant cells in the aspirated fluid and tissues. Single node excision surgery or modified radical neck dissection surgery are considered as possible treatment options. Because the lesion was a well-demarcated single node and the patient strongly wanted to keep functionality of her neck as much as possible along with minimizing post-op impairment, a single node excision surgery was performed under general anesthesia in January 2007. The submandibular lymph node was removed without disruption of the capsule and it consisted of one soft tissue nodule measuring 4.0 × 2.5 × 2.5 cm (Fig. 5A, B). The lymph node revealed the identical histopathologic morphology as the primary lesion, which confirmed the diagnosis of a malignant ameloblastoma (Fig. 6). The patient has been regularly monitored each year for follow-up visits, presently has been free of symptoms for 10 years after mandibular surgery and seven years after neck node excision, and will be under close evaluation to detect any recurrences or metastasis.

3. Results

Of the 587 articles were examined and 11 studies were selected for this study. In the English literature, there are 23 cases of cervical

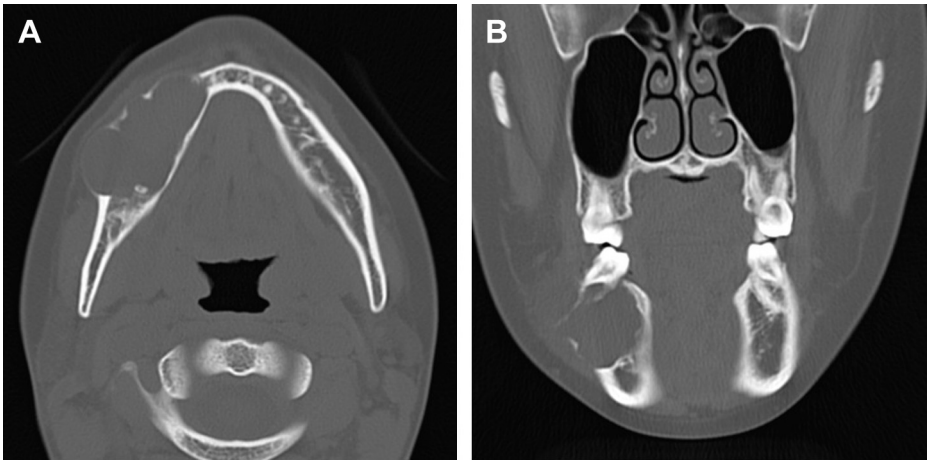

Fig. 1. Computed tomography showing multilocular radiolucency with destruction of buccal cortical bone of right mandible in June 2004 (A: axial section, B: coronal section).