Case Report

rare tumors

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A case of initially metastasizing pleomorphic adenoma of parotid gland

Sungchul Ko[1], Kye Hoon Park[1], Ji-Hye Lee[2] and Ki Nam Park[3]

Abstract

Metastasis of pleomorphic adenoma (PA) is rare and usually presented as a locoregional recurrence developed many years after excision of the primary tumor although the PA is the most common neoplasm in the parotid gland. We described a case of a 48-year-old male with a parotid tumor with multiple enlarged ipsilateral lymph nodes which suggested a malignancy. The tumors had been neither evaluated nor excised and preoperative evaluation revealed benign PA in both lesions. After the complete surgical excision, the final pathology was notable for benign PA with metastasis to regional lymph nodes. At 1 year follow up he was clinically and radiographically free of disease. This implies that pleomorphic adenoma can occur as initially metastasis to regional lymph node even though benign neoplasm.

Keywords

parotid neoplasms, metastasizing pleomorphic adenoma, parotid gland

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Introduction

Pleomorphic adenoma (PA) is the most common benign tumor of salivary gland commonly affecting parotid and submandibular gland and only 10% of PA arise at minor salivary gland.[1] PA can be converted to carcinoma ex pleomorphic adenoma, the risk of developing carcinoma ex pleomorphic adenoma is about 2–9%.[2] Main treatment of PA is complete excision and adequate resection margin should be secured because pseudopods from the tumor surface may be left near the resection margin and result in recurrence, which have been reported as the rate of 25–45% after incomplete resection.[2,3] Despite its benign pathological feature, PA rarely metastasizes outside the salivary gland and develops slowly after incomplete excision. Although metastasizing pleomorphic adenoma (MPA) has been reported as it progresses after incomplete resection, recently, we experienced a rare case of initially occurred MPA with no history of previous surgery or invasive procedure including fine needle aspiration cytology and core needle biopsy. Thus, we presented the case and relative literature review.

Case report

A 48-year-old male patient without any history of invasive procedure or surgery on head and neck area was referred for further evaluation addressing painless multiple neck masses on left side neck. Physical examination showed round firm non-tender neck masses on left parotid and level II. No specific finding was noted in laboratory tests and chest

[1]Department of Otorhinolaryngology-Head and Neck Surgery, Soonchunhyang University Bucheon Hospital, Cheonan, Republic of Korea
[2]Department of Pathology, College of Medicine, Soonchunhyang University Cheonan Hospital, Cheonan, Republic of Korea
[3]Department of Otorhinolaryngology-Head and Neck Surgery, Soonchunhyang University College of Medicine, Bucheon, Republic of Korea

Corresponding author:
Ki Nam Park, Department of Otorhinolaryngology-Head and Neck Surgery, Soonchunhyang University Bucheon Hospital, Soonchunhyang University College of Medicine, 170 Jomaru-ro, Wonmi-gu, Bucheon 420-767, Republic of Korea.
Email: man7140@schmc.ac.kr

radiology. Enhance neck computed tomography (CT) scan and ultrasonography showed that 25 × 15 mm sized parotid mass with variable sized multiple lymph nodes at left level II. (Figure 1) Ultrasound-guided core needle biopsy was performed on parotid and neck masses and histologic findings from each showed the admixture of epithelial and myoepithelial cells in a variable stroma, which suggested pleomorphic adenoma. Superficial parotidectomy (SP) with selective neck dissection of level II was performed under the diagnosis of initially metastasizing pleomorphic adenoma. (Figure 2(a) and (b)) Histologically, the primary parotid tumor was surrounded by a fibrous capsule and composed of the typical mixture of epithelial and myoepithelial cells with abundant mucinous material of a pleomorphic adenoma in variable chondromyxoid stroma. Several masses found in the left neck were lymph nodes, it showed the similar cytologic findings as the parotid mass diagnosed with pleomorphic adenoma diagnosed. The patient showed no significant complications including facial palsy and was discharged at postoperative day 4. After confirming diagnosis of metastasizing pleomorphic adenoma (MPA), the patient was submitted for chest CT scan for survey of another metastatic lesion. A 1 mm sized tiny lung nodule was noted at the right upper lobe and the nodule was very small in size, so regular follow-up was decided rather than immediate biopsy. After 2 years of follow-up, there was no evidence of recurrence on physical and radiologic exams.

Discussion

Pleomorphic adenoma is benign mass with typical histologic findings of mixture of epithelial and myoepithelial components surrounded by fibrous capsule which forms pseudopods. Incomplete resection may result in relapse or metastasis. 2–7% of untreated PA can be converted to malignant tumor known as carcinoma ex pleomorphic adenoma.[4] Although PA is a histologically benign tumor, however, it has been reported that PAs can occur synchronously or metachronously and have also been reported outside the salivary gland such as oral cavity, pharynx, larynx, nasal cavity paranasal sinus, cervical lymph nodes, soft tissues of neck, upper and lower limbs, trunk, lung, breast, sellar region, lacrimal glands and sac, ear, mediastinum, and intraosseous region. Due to the paucity of cases, the incidence of MPA has not been established yet. Nouraei et al. reported that only 42 cases had been documented in the literature over 50 years. In most cases reported previously, metastasis was related to incomplete resection near the

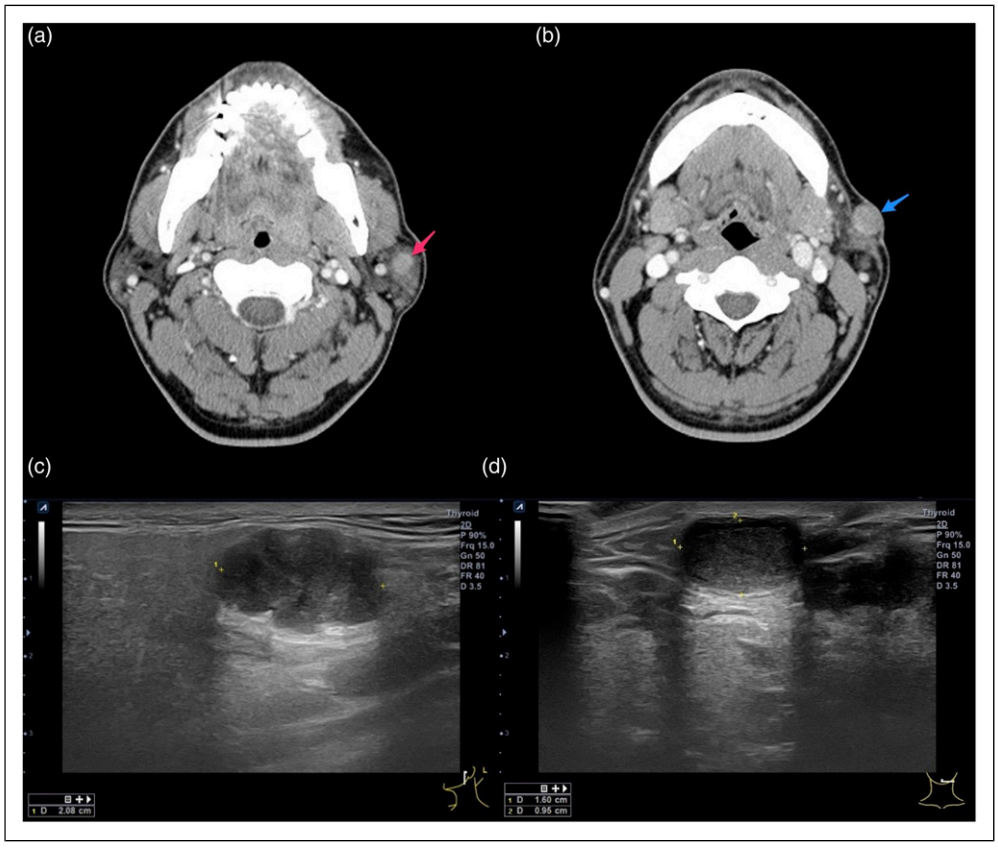

Figure 1. Preoperative evaluation of tumors. Enhanced neck CT (a, b) and ultrasound showed ovoid masses with slight enhancement in left parotid gland and lateral neck level II (c, d).

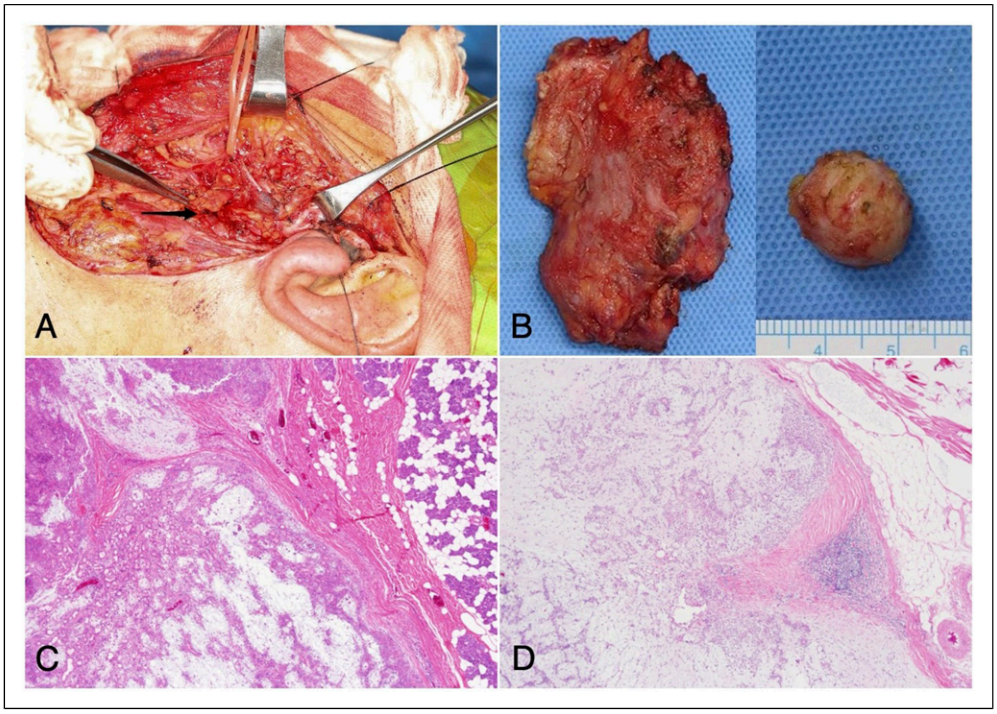

Figure 2. Complete excision of multiple tumors and pathologic findings. Left superficial parotidectomy with ipsilateral level II lymph node dissection was conducted. (a) Black arrow shows parotid tumor adjacent to facial nerve (a) and main mass and lymph node specimen is shown seperately (b). Pathologic finding of the parotid is compatible with benign pleomorphic adenoma showing benign epithelial cells in the myxoid stroma (H&E, x40) (c) and is also found in left level II lymph node. (H&E, x40) (d).

anatomical structures like facial nerve or violation of tumor capsule.[5] Synchronously occurred MPA is extremely rare.[6] The patient with MPA usually addresses no symptom until the mass can be palpated and histological confirmation is necessary for diagnosis.[7,8] Knight et al. reported that there was a considerable period of 0–51 years from the diagnosis of primary PA to the metastatic lesions, with an average of 14.9 years. There was no significant difference in gender regarding incidence, and most of the primary lesions were parotid gland (74.1%), followed by submandibular gland (14.8%) and palate (6.2%). The metastatic lesions that have been reported were bone (36.6%), lungs (33.8), lymph nodes (20.1%), kidneys (8.6%), liver (4.9%), and central nervous system (3.7%).[9]

Complete resection is the mainstay of treatment for MPA. Incomplete resection caused by enucleation increases the risk of local recurrence which may lead to the potential of distant metastases.[5] Benign salivary gland tumor of superficial lobe of parotid gland can be excised with SP.[10] Total parotidectomy was used in the majority of cases for recurrent metastasizing pleomorphic adenoma if a facial nerve was not involved,[11] however, we conducted the conventional SP for this patient because the pleomorphic adenoma was located only in superficial lobe and there was no history of surgical resection. Regarding treatment for regional neck metastasis, the formal neck dissection should be conducted for clinically evident nodal metastasis of parotid malignancy and elective neck dissection is recommended in the high-grade histology type of malignancy.[12] However, we conducted the selective neck dissection for the level II compartment rather than berry picking because there was no evidence in the other neck area and the PA is classified as a benign tumor.

Radiotherapy can be applied to the limited case of unresectable MPA.[2,13] There have been only a few reports regarding long-term prognosis, hence exact prognosis of MPA is uncertain. Multiple metastases and distant metastasis have been reported as important predictive values for poor prognosis.[5,9,14] Knight et al. reported that 8 out of 13 patients with more than three multiple metastases died within 3 years after diagnosis of MPA, and 5 others were not followed up.[9] Distant metastasis of MPA was reported as significantly associated with morbidity and mortality rate and the 5-year disease-specific and disease-free survival rate of MPA with distant metastasis were 58% and 50%, respectively.[5]

The duration of 2 years of follow-up seemed to be not enough because the median recurrence time was 10.3 years after initial treatment.[15] The patient had been fully explained the possibility of recurrence and malignant transformation. We recommended the regular imaging work-up including the neck CT and chest X-ray over 10 years after surgery.

Conclusion

The etiology of MPA has not been clearly revealed. Since most of the cases documented in the literature occurred after surgery or invasive procedure, the hypothesis that surgical manipulation caused tumor cell migration to blood vessels and hematogenous spread can lead to metastasis. Lymphatic metastasis and aspiration of tumor cells have also been reported as possible causes of lung metastasis.[2,8] This presenting case is extremely unusual in that metastasis occurred without prior surgical history or invasive biopsy on primary PA. It can be assumed that there might have been cellular migration from tumor into lymphatics or blood vessels caused by local compression or blunt trauma. When diagnosing PA, as a screening, imaging study on common site of metastasis is worth suggesting.

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Declaration of Conflicting Interests

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Ethical approval

(include full name of committee approving the research and if available mention reference number of that approval) Institutional Review Board of Soonchunhyang University approved the presentation of a single patient's case. (2022-01-029)

Informed Consent

Informed verbal consent was obtained from the patient reported in this manuscript.

Contributorship

All authors made significant contributions and have approved the final version of manuscript.

ORCID iD

Ki Nam Park https://orcid.org/0000-0001-6641-3981

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