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Phyllodes tumor metastasis to the tonsil with synchronous undifferentiated carcinoma

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ABSTRACT

INTRODUCTION: Breast tumor metastasis to the tonsil is extremely rare.**PRESENTATION OF CASE:** A 54-year-old woman underwent resection of a breast malignant phyllodes tumor and later presented with metastasis to the lung and the left tonsil. She underwent left lower lobectomy and resection of the left tonsillar tumor. She subsequently developed undifferentiated carcinoma of the right tonsil. She underwent resection of the right tonsillar tumor and chemotherapy was started. The central venous catheter became infected with methicillin-resistant *Staphylococcus aureus*. Finally, she died.**DISCUSSION:** To the best of our knowledge, this is the first reported case of a phyllodes tumor metastasizing to the tonsil. Furthermore, morphological and immunohistochemical study revealed that the right tonsillar tumor was irrelevant to the phyllodes tumors.**CONCLUSION:** We report a case of phyllodes tumor metastasis to the left tonsil which developed undifferentiated carcinoma in the other side of tonsil later. Breast tumor metastasis to the tonsil is rare but it should be considered as a possible diagnosis.© 2014 The Authors. Published by Elsevier Ltd. on behalf of Surgical Associates Ltd. This is an open access article under the CC BY-NC-ND license (<http://creativecommons.org/licenses/by-nc-nd/3.0/>).

1. Introduction

Metastasis to the tonsil is extremely rare, accounting for only 0.8% of all tonsillar tumors.¹ The cases reported involved metastasis from cutaneous or mucosal melanoma,^{2,3} gastric adenocarcinoma,^{4,5} hypernephroma,^{6,7} and various lung neoplasms.⁸ The less common metastatic tonsillar tumors originate from mesothelioma,⁹ hepatocellular carcinoma,¹⁰ ovarian endometrioid carcinoma,¹¹ seminoma,¹² anaplastic thyroid carcinoma,¹³ pancreatic adenocarcinoma,¹⁴ prostatic adenocarcinoma,¹⁵ and gall bladder carcinoma.¹⁶ Breast carcinoma is one of the rarest primary tumors that can metastasize to the tonsil; only eight cases have been reported in the literature,¹⁷ and only one case of tonsillar metastasis of breast sarcoma has been reported.¹⁸

Tonsillar metastasis rarely becomes apparent before the diagnosis of the primary neoplasm, and only a few cases of tonsillar metastasis as a presenting feature have been reported in the

literature.¹⁹ In most of these cases, the presenting symptoms were sore throat, an oral globus sensation, and different degrees of dysphagia and odynophagia. In other cases, patients were asymptomatic, and their tonsillar neoplasm was detected incidentally during a routine oral examination. Tonsillar metastasis can be bilateral or unilateral depending on the nature of the primary neoplasm. The prognosis for patients with tonsillar metastasis is rather poor.⁶ In this report, we present a case of a phyllodes tumor that metastasized to the tonsil; we have included the clinical features and pathological findings.

2. Presentation of case

A 54-year-old woman underwent right mastectomy for a breast tumor 3 years before she was referred to our department. The tumor was diagnosed pathologically as a malignant phyllodes tumor. There was no evidence of axillary metastasis. Histopathological examination showed a tumor larger than 10 cm in diameter that had slightly invaded the surrounding tissues and was characterized by moderate nuclei pleomorphism, moderate cell density, and moderate mitosis (Fig. 1A and B). The final pathology revealed that no tumor was identified on the margin of the resected specimen.

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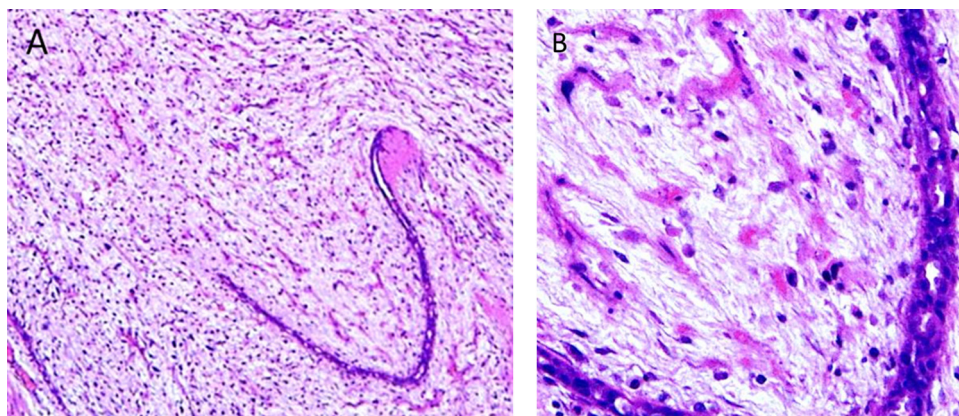


Fig. 1. (A) The breast surgical specimen shows spindle cell proliferation with evident stromal overgrowth and numerous bizarre cells (HE stain, 40 \times). (B) Higher magnification shows malignant spindle cell proliferation arranged in long intersecting fascicles. The cells show evident pleomorphism, hyperchromasia, and mitotic activity (HE stain, 200 \times).

Free margin was 900 μ m. Immunostaining showed that the tissue was negative for cytokeratin AE1/AE3, MNF116, and CAM5.2.

Although she was informed of possible recurrence, she stopped ambulatory care 4 months after the operation. Two years and 10 months after the operation, she presented with cough and dyspnea. Physical examination revealed a tumor in the left lung. A biopsy of the lung tumor was performed with bronchoscopy, and the sample was diagnosed as metastasis of the phyllodes tumor. She subsequently underwent left lower lobectomy for the lung metastasis. Pathology analysis of the lung specimen indicated metastasis of the malignant phyllodes tumor with incomplete resection. Three years and 1 month after the lung operation, the patient was referred to our department for throat pain and presented with left tonsil swelling. Physical examination revealed a left tonsillar tumor measuring 20 mm in diameter. Computed tomography images showed a left tonsillar tumor that did not spread outside the pharyngeal constrictor muscles. No neck lymphadenopathy was present. Pathology analysis of the biopsy of the left tonsil showed metastasis of the malignant phyllodes tumor of the breast. No tumor recurrence was identified in the left lung. She underwent resection of the left tonsillar tumor. Histopathological analysis of the left tonsillar tumor showed growth of spindle cells, necrosis, and atypical nuclei (Fig. 2A and B). Immunostaining showed that the tissue was negative for cytokeratin AE1/AE3, MNF116, and CAM5.2. Because the pathological features of the left tonsillar tumor were similar to those of the primary breast tumor, the left tonsillar tumor was diagnosed as a metastasis of the primary phyllodes tumor. The post-operative course was favorable, and she was discharged 1 week later.

Three months after the left tonsil resection, she presented with right tonsil swelling. Physical examination revealed a right tonsillar tumor. Biopsy of the right tonsillar tumor showed undifferentiated carcinoma of the oropharynx. One week after the biopsy, she underwent resection of the right tonsillar tumor. Histopathology analysis of the right tonsillar tumor showed growth of spindle cells, necrosis, and atypical nuclei, which differed from the findings for the primary breast phyllodes tumor (Fig. 3). Immunostaining showed that the tissue was positive for cytokeratin AE1/AE3, MNF116, and CAM5.2. In situ hybridization showed that the tissue was negative for Epstein–Barr virus. The pathological features of the resected right tonsil were similar to those of undifferentiated carcinoma of the oropharynx. Three histopathologists consulted and agreed on the diagnosis.

No recurrence of the tonsillar tumors or deterioration of the lung lesion had been observed, but the breast tumor regrew. Chemotherapy, including doxorubicin and ifosfamide, was started. The central venous catheter became infected with methicillin-resistant *Staphylococcus aureus* during chemotherapy, and the chemotherapy was stopped. Finally, she died 1 month after the cessation of chemotherapy.

3. Discussion

Phyllodes tumor is an uncommon breast neoplasm that accounts for less than 1.0% of all breast tumors and has the ability to recur and metastasize.²⁰ Phyllodes tumor comprises both epithelial and stromal elements. Phyllodes tumor is graded as benign, borderline, or malignant according to a set of histological data including

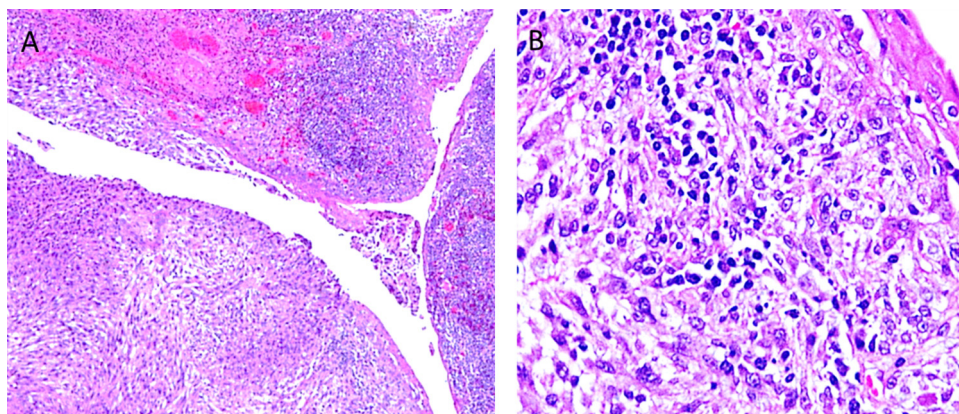


Fig. 2. (A) Section from the left tonsillar tumor covered by stratified squamous epithelium shows remnants of lymphoid tissue and wide areas of hemorrhage and necrosis (HE stain, 40 \times). (B) Higher magnification shows brisk mitotic activity and multinucleated tumor giant cells (HE stain, 200 \times).

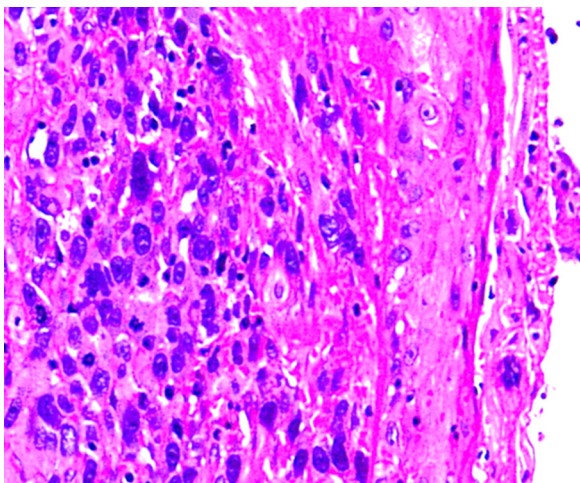


Fig. 3. The histopathological findings of the right tonsillar tumor show growth of spindle cells, necrosis, and atypical nuclei. These differed from the findings for the breast tumor: HE stain 200 \times .

stromal cellularity, stromal cell atypia, mitotic activity, stromal overgrowth, necrosis, and whether the margin is rounded or infiltrative and shows malignant heterologous elements. A review of the literature suggests that malignancy occurs in 14–50% of cases.²¹ The overall incidence of metastasis of phyllodes tumor is 3–12%, most commonly to the lung.²²

The treatment of choice is excision with a 10-mm margin, although malignant lesions require mastectomy. However, mastectomy is not effective in preventing metastatic disease,²² and the optimal treatment of metastatic disease has not been established. Phyllodes tumors are thought to be resistant to both chemotherapy and radiation.²³

This case is extremely rare from two perspectives. First, to our knowledge, this is the first reported case of a phyllodes tumor metastasizing to the tonsil. A review of the literature found several reports of phyllodes tumors metastasizing to the head and neck region, including the right ramus,²⁴ posterior right maxilla,²⁵ left mandibular posterior quadrant,²⁶ and left angle and ramus of the mandible.²⁷ Other cases involved gingival metastasis and metastasis of the temporomandibular region, parotid gland, thyroid adenoma.^{28–31} There is no report of phyllodes tumor metastasizing to the tonsil.

Second, this is the first reported case of secondary primary right tonsillar tumor which was developed in the patient with a metastatic left tonsillar tumor. The clinical course suggested that the right tonsillar tumor should be a metastasis of a phyllodes tumor. However, the right tonsillar tumor was diagnosed as undifferentiated carcinoma because the histopathology clearly differed from those of the left tonsillar tumor. Secondary neoplasms occurring in cases of phyllodes tumor were reported in one paper.³² In that report, eight tumors were diagnosed in 6/32 patients as secondary tumors of duodenal cancer, cervical cancer, laryngeal cancer, alveolar cell adenocarcinoma, metastatic colon cancer, astrocytoma, esophageal cancer, and lung adenocarcinoma. But these cases were not simultaneously occurred. The diagnosis of the right tonsillar tumor in this case remains controversial. Another hypothesis is anaplastic transformation of phyllodes tumor (i.e., stromal elements may have changed to epithelial elements). Further investigation is necessary to understand the metastasis process in cases of phyllodes tumor.

Breast tumor metastasis to the tonsil is rare but it should be considered as a possible diagnosis.

4. Conclusion

We report a case of phyllodes tumor metastasis to the left tonsil which developed undifferentiated carcinoma in the other side of tonsil later. Breast tumor metastasis to the tonsil is rare but it should be considered as a possible diagnosis.

Conflict of interest statement

None declared.

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None.

Ethical approval

This was an institutional review board-approved retrospective review. Written informed consent was obtained from the patient for publication of this case report and accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal on request.

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