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Lacrimal Sac Metastasis from Phyllodes Tumor of the Breast

Dear Editor,

The phyllodes tumor belongs to the fibroepithelial tumors group, which includes a combination of neoplastic epithelial and mesenchymal components [1]. Breast cancer metastases have various distributions, and the most common metastatic sites are bone, regional lymphatic organs, lung, liver, and brain [2]. Given that metastasis from breast cancer to the nasolacrimal duct is extremely rare, the mechanisms involved remain unclear. Here, we describe an unusual metastatic spread of phyllodes tumor of the breast and discuss the nasolacrimal duct obstruction caused by this metastasis.

A 41-year-old female was referred to the Ophthalmology Department of Inha University Hospital owing to complaints of epiphora, bloody tears, and swelling of the left eyelid, which began a week prior to presentation. Her medical history was relevant to malignant phyllodes tumor of the breast, which was diagnosed via positron emission tomography-computed tomography and widespread, with gastric metastases. Four months before presentation, she had undergone mastectomy with skin graft and palliative chemotherapy with olaratumab and doxorubicin, to which her body responded poorly. Ophthalmologic examination confirmed elevation in the left medial canthal area with tenderness (Fig. 1A). Nasolacrimal sac irrigation revealed obstruction with blood regurgitation in the left lacrimal duct. Visual acuity, intraocular pressure, ocular motility, and the anterior segment were within normal limits. There

was no exophthalmos. Scans obtained via magnetic resonance imaging revealed that a tumor extended from the lacrimal fossa to the lamina papyracea, lacrimal bone, frontal process of the maxilla, and nasolacrimal duct (Fig. 1B). We considered performing a debulking surgery to reduce the tumor burden and relieve the patient's symptoms,

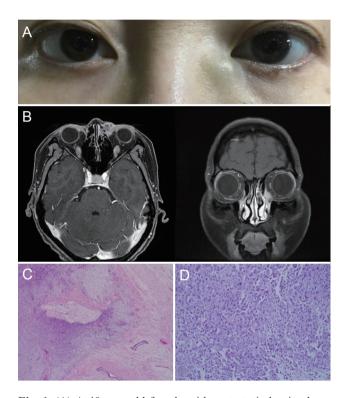


Fig. 1. (A) A 40-year-old female with metastatic lacrimal sac tumor. Note swelling of the left medial canthal area. (B) Axial and coronal views of gadolinium-enhanced T1-weighted fat-suppressed magnetic resonance imaging demonstrated a soft tissue mass in the left lacrimal fossa extending to the nasolacrimal duct. (C) Histological examination of the breast tumor revealed a malignant phyllodes tumor with marked stromal cellularity alternating with lower cellular area, nuclear pleomorphism, and stromal overgrowth (hematoxylin and eosin stain, ×100). (D) Histological examination of the metastatic phyllodes tumor in the lacrimal sac revealed a malignant stromal component with polygonal plump cytoplasm, nuclear pleomorphism, and necrosis without an epithelial component (hematoxylin and eosin stain, ×200). A written consent was obtained from the patient.

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but the hemato-oncologist reported that her general condition and life expectancy may be poor. Thus, we performed local excision and biopsy of the presumed lacrimal sac metastasis. Microscopically, the tumor comprised malignant epithelioid and spindled mesenchymal cells with large cytoplasm as well as nuclear pleomorphism, frequent mitosis, and focal necrosis without any epithelial component. The histological features were similar to those of the mesenchymal component of malignant phyllodes tumor of the breast. Therefore, a diagnosis of metastatic malignant phyllodes tumor was made (Fig. 1C, 1D). Palliative radiotherapy was recommended after surgery, and the complications were carefully explained. However, she chose against receiving further treatment. A few days after surgery, she complained of bloody discharge with tearing, swelling, redness, and tenderness in the medial canthal area of the left eve.

Breast cancer metastasizes to the orbit, and such an occurrence is relatively common [2]. However, acquired nasolacrimal duct obstruction caused by neoplasm is rare, and the majority of these neoplasms are malignant. Because patients with lacrimal sac tumor typically exhibit symptoms similar to those of chronic dacryocystitis, clinicians should consider the possibility of lacrimal sac malignancy in order to facilitate an early diagnosis and achieve better outcomes because of the unique location. Furthermore, a history of malignancy also warrants further investigation. The ophthalmologic signs that are particularly useful for diagnosing tumors of the lacrimal sac and nasolacrimal duct include epiphora, recurrent dacryocystitis, epistaxis, and swelling and/or redness of the skin overlying the lacrimal sac [3]. Phyllodes tumor of the breast has a peak incidence in women between the ages of 40 and 50 years [4]. It is regarded as a stroma-derived tumor and sub-classified as benign (10% to 40%), borderline (35% to 64%) or malignant (25%) [4]. Histologically, phyllodes tumor are fibroepithelial tumors which probably originate from the terminal ducto-lobular unit [1]. The World Health Organization has established criteria for the diagnosis and grading of phyllodes tumors. A benign phyllodes tumor has mildly increased stromal cellularity (compared with fibroadenoma), minimal nuclear atypia, pushing border, and mitoses of 4 / 10 high-power fields, with no stromal overgrowth. In contrast, a malignant phyllodes tumor has marked stromal cellularity and atypia, permeative margins, and mitotic activity of at least 10 / 10 high-power fields, with easily identifiable stromal overgrowth.

Malignant tumors of the nasolacrimal duct are potentially lethal because of their anatomical location. Complete and wide surgical excision of the tumor, whether benign or malignant, is the treatment of choice. However, if a metastatic malignant tumor is suspected, a systemic evaluation that includes radiologic imaging is necessary. A multidisciplinary treatment plan based on life expectancy should be offered to patients. Careful use of globe-sparing treatment could achieve reasonable visual outcomes [5]. Although lacrimal sac metastasis is rare, younger patients with atypical symptoms and a history of malignancy should undergo further evaluation for possible metastasis to the nasolacrimal duct and lacrimal sac.

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Conflict of Interest

No potential conflict of interest relevant to this article was reported.

References

- Macdonald OK, Lee CM, Tward JD, et al. Malignant phyllodes tumor of the female breast: association of primary therapy with cause-specific survival from the Surveillance, Epidemiology, and End Results (SEER) program. *Cancer* 2006:107:2127-33.
- Nickelsen MN, VON Holstein S, Hansen AB, et al. Breast carcinoma metastasis to the lacrimal gland: two case reports. Oncol Lett 2015;10:1031-5.

- Stefanyszyn MA, Hidayat AA, Pe'er JJ, Flanagan JC. Lacrimal sac tumors. *Ophthalmic Plast Reconstr Surg* 1994;10:169-84.
- 4. Norris HJ, Taylor HB. Relationship of histologic features to behavior of cystosarcoma phyllodes. Analysis of nine-
- ty-four cases. Cancer 1967;20:2090-9.
- 5. El-Sawy T, Frank SJ, Hanna E, et al. Multidisciplinary management of lacrimal sac/nasolacrimal duct carcinomas. *Ophthalmic Plast Reconstr Surg* 2013;29:454-7.