Rare Case of hamartoma in nasolacrimal duct

Sangwon Jung¹, Gyeong Min Lee² Yeon Bi Han,³ Namju Kim⁴

¹Department of Ophthalmology, Seoul National University College of Medicine, Seoul National University Hospital, Seoul, Korea.
²Department of Ophthalmology, Dongguk University Ilsan Medical Center, Goyang, Republic of Korea.
³Department of Pathology and Translational Medicine, Seoul National University Bundang Hospital, Seongnam, Korea
⁴Department of Ophthalmology, Seoul National University Bundang Hospital, Seongnam, Korea

Corresponding author: Namju Kim, M.D., Ph.D.
Professor, Department of Ophthalmology, Seoul National University Bundang Hospital, Seongnam, Korea
E-mail: kimnamju@snubh.org

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Dear Editor,

Nasolacrimal duct (NLD) tumors are rare and [1] incidence was reported 0.02% of all tumors arising in the head and neck, and only 1% of patients receiving endoscopic dacryocystorhinostomy were found to have NLD tumor [1, 2]. They present with obstructive symptoms like epiphora, and symptoms may be present even before radiographic abnormalities become evident [3]. Among NLD tumors, papilloma was the most frequently reported benign tumor and lymphoma was the most common malignant tumor [1,3]. Hamartoma of NLD is extremely rare; to our knowledge, there have been no reported cases before. Here we report a case of hamartoma in the NLD.

A 35-year-old woman was referred to our clinic for epiphora of the right eye that lasted for 2 months. She had no underlying disease except for a unilateral thyroidectomy due to a benign thyroid nodule 18 years ago. On examination, the best corrected visual acuity was 20/20 in both eyes and the tear meniscus height was high on the right eye. Dacryocystography showed right intraluminal filling defects and orbital computed tomography showed a 1.7 cm-sized enhancing mass in the right NLD (Fig. 1A-C). To clarify the nature of NLD mass, orbital magnetic resonance imaging was performed and (Fig. 1D-E) mass was suspected to be a benign tumor such as papilloma or oncocytoma.

Surgery was planned as a diagnostic dacryoendoscopy followed by endonasal dacryocystorhinostomy (DCR). Intraoperative dacryendoscopy revealed a yellowish mass in the NLD, and the likelihood of malignant tumor such as melanoma was low (Fig. 1F). Endonasal DCR was performed and after removing lacrimal bone and incising along the lacrimal sac and NLD, a yellowish mass in the NLD was exposed. Using microcup forceps, mass was easily pulled out. Histological examination showed a benign polypoid lesion with bland-looking mature stromal and mucinous glandular proliferation in disorganized pattern, consistent with hamartoma (Fig. 1G-I). Two months after the surgery, tear meniscus height was normalized and symptoms disappeared.

We first reported hamartoma in the NLD. There have been a few reports of NLD obstruction due to compression of NLD from surrounding tissues with hamartoma [4,5]. However, hamartoma in the lumen of the NLD is first reported.

NLD tumors are very rare and can be presented early as obstructive symptoms such as epiphora. Epiphora caused by NLD obstruction is frequent in old age and relatively rare in young age. Therefore, if young patients present with epiphora, although rare, NLD tumor should be suspected and thorough imaging should be performed. Especially, if epiphora is intermittent or bloody tearing is present, more attention should be paid [1, 2].
REFERENCES


Figure 1. Preoperative examination. A, Dacryocystography showed right intraluminal filling defects. B-C, Computed tomography images demonstrated about 1.7 cm-sized enhancing mass in the right NLD. D-E, Magnetic resonance images demonstrated soft tissue tumor in the lower part of the right NLD. Intraoperative pictures. F, dacryoendoscopy revealed a yellowish mass in the NLD. G, Gross specimen of the excised lesion. H and I, Histopathologic examination showed benign polypoid lesion with bland-looking mature stromal and mucinous glandular proliferation in disorganized pattern. Scale bar = 5mm. Scale bar = 300 µm