

## Stage Surgery on Inverted Papilloma which Invaded Lacrimal Sac, Periorbita, Ethmoid and Frontal Sinus

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### — ABSTRACT —

Inverted papilloma of the nasal cavity and the paranasal sinuses is a benign epithelial tumor with a high rate of recurrence, local aggressiveness, and malignant transformation. For these reasons, inverted papilloma has been treated like malignant tumors with extensive surgical resection. With the help of endoscopic sinus surgery technique, it is now available to treat inverted papilloma with stage surgery without severe complications which usually resulted from extensive one stage resection. We report a case of stage surgery on inverted papilloma which invaded lacrimal sac, periorbita, ethmoid and frontal sinus. (*J Clinical Otolaryngol* 2016;27:143-147)

**KEY WORDS** : Inverted papilloma · Lacrimal sac · Periorbita · Surgery.

### Introduction

Inverted papilloma (IP) of the nasal cavity and the paranasal sinuses is a benign epithelial tumor with a high rate of recurrence, local aggressiveness, and malignant transformation.<sup>1,2)</sup> For these reasons, IP has been treated like malignant tumors with extensive surgical resection.

IP of lacrimal sac and periorbita is rarely reported and treatment strategies may differ according to the surgeon's philosophy on surgical strategy of IPs. Also the surgical strategy for the lacrimal sac tumor must be highly individualized based on tumor grade, aggressiveness, histopathological pattern and patient's age and preference.

Authors present a successful endoscopic stage surgery on IP which invaded lacrimal sac, periorbita, ethmoid and frontal sinus with the literature review.

### Case Report

A 41-year-old female presented in outpatient clinic with a complaint of tender swelling mass on the inner side of her right eye for 5 years which suddenly aggravated 2 months ago. She complained of epiphora but not diplopia nor visual disturbance. Patient is a nonsmoker and drinks only socially. She had a history of embolization of unruptured paraclinoid internal carotid artery aneurysm 5 months ago. Otherwise, she had no past medical history. On physical examination, there was bulging of right lateral wall of nasal cavity. Externally there was asymmetry of right lower eyelid because of the fixed mass at right medial canthus (Fig. 1). Intra-ocular pressure was 18 in the right side and 14 in the left side. Eye examination showed hyperdeviation on the right orbit and no exophthalmos.

Paranasal sinus computed tomography (PNS CT) showed a 39×37×24 mm sized lobulated hyperdense

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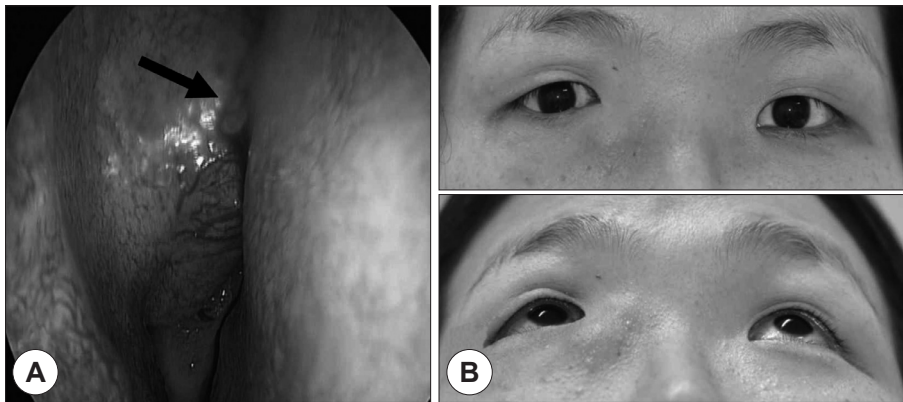
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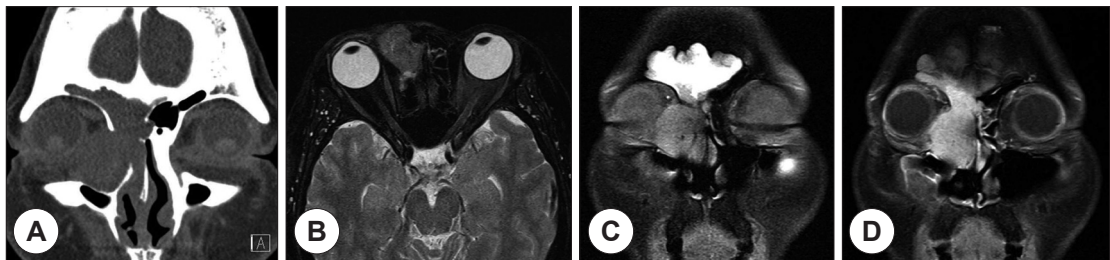
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**Fig. 1.** Preoperative physical findings. The endoscopic examination reveals mass bulging from the lateral wall of the right nasal cavity (arrow) (A). External view shows enlarged right lacrimal sac compared to left side (B).



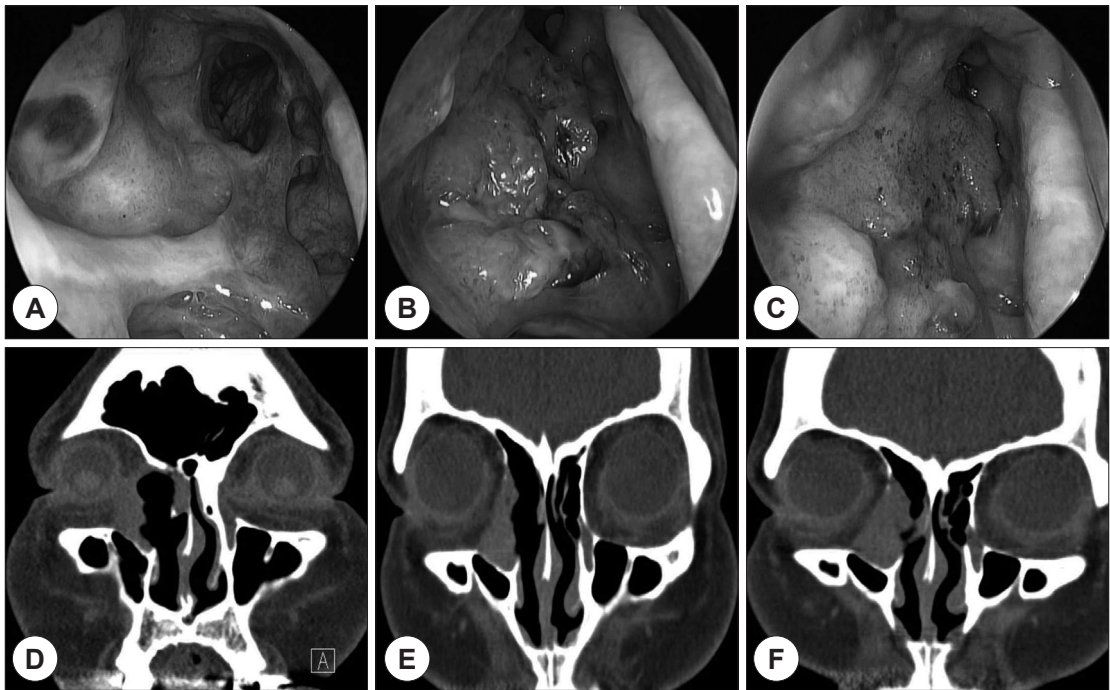
**Fig. 2.** Preoperative imaging findings. Preoperative coronal CT shows soft tissue density lesion invading into the right orbit and frontal sinus (A). T2 axial MRI shows mass with intermediate signal intensity (SI) in the anterior ethmoid to lacrimal sac (B). T1 coronal MRI shows mass with intermediate SI which is encroaching the periorbita and high SI in the frontal sinus (C). T1 coronal gadolinium enhanced MRI shows strong homogenous enhancing mass lesion (D).

lesion occupying right frontal sinus, ethmoid sinus, superior nasal cavity, ethmoid infundibulum and the periorbita with upward and lateral displacement of the globe. Magnetic resonance imaging (MRI) revealed intermediate signal intensity on T1 and T2-weighted image in the right ethmoid sinus, lower part of frontal sinus, periorbita and the lacrimal sac, and high signal intensity on upper frontal sinus and maxillary sinus on T2-weighted image which suggested inflammatory change. The mass was enhanced on the gadolinium contrast image (Fig. 2).

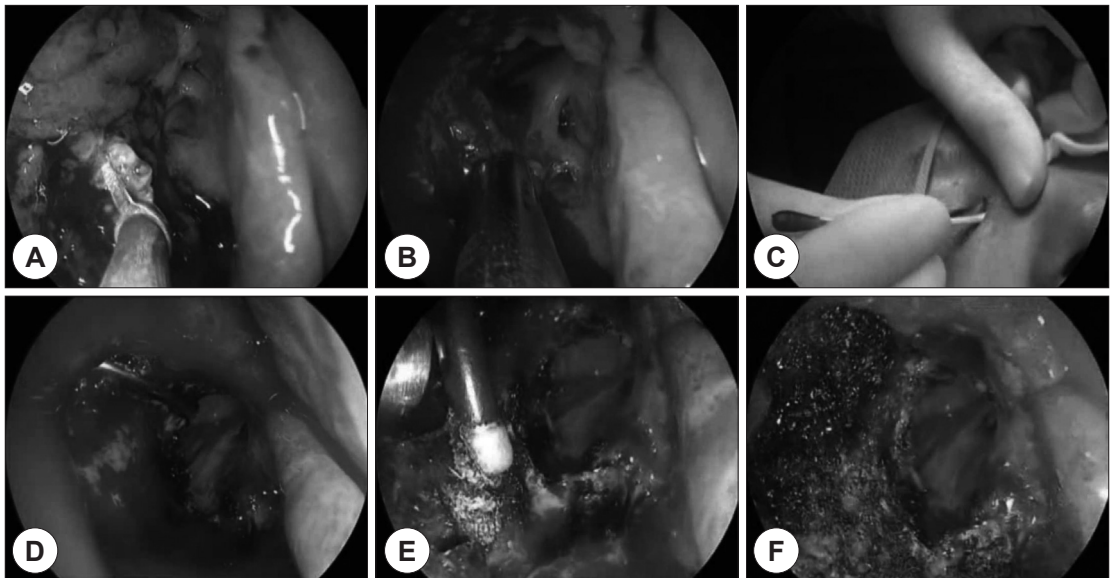
Endoscopic sinus surgery for the purpose of biopsy and debulking was performed under general anesthesia. IP was diagnosed via frozen and permanent biopsy. Lamina papyracea was eroded and periorbita was infiltrated. The tumor origin seemed to be the periorbita and the frontal recess. After surgical deb-

ulking of the mass in right frontal and ethmoid sinus, staged operation was planned whenever there is an evidence of tumor recurrence during the later follow-up. Symptom of the patient relieved immediately after the surgery. The patient was followed up regularly after 1st operation with endoscope and PNS CT. After 17 months, the mass around lacrimal sac has increased in both endoscopic and PNS CT findings (Fig. 3).

The 2nd stage operation was performed with ophthalmologist under general anesthesia. Lacrimal sac was removed with external dacryocystorhinostomy after lower eyelid incision. Ethmoidal and periorbital mass was exposed and removed via external (above) and endoscopic (below) approach (Fig. 4). IP was diagnosed without carcinoma in situ transformation. The mass near lacrimal bone was left for later surgery.



**Fig. 3.** Postoperative endoscopic and CT findings. Postoperative 1 month endoscopic and CT findings (A, D). The mass was removed except for the lacrimal sac. Postoperative 1 year endoscopic and CT findings (B, E). Postoperative 17 month endoscopic and CT findings (C, F). The examination revealed localized increased mass lesion in the ethmoid sinus.



**Fig. 4.** Intraoperative findings. The ethmoid mass is removed via bipolar coagulation nasal forceps and debrider (below approach) (A, B). Lower lid incision is made by ophthalmologist (C). The mass around lacrimal sac is removed via curved debrider and suction monopolar cauterization (above approach) (D, E). After tumor removal (F).

After 9 months, the mass recurred in the lacrimal bone area and was removed under local anesthesia (3rd stage operation). The nasal cavity seemed to be no evidence of tumor after 18 months of 3rd stage operation and the patient is free of symptom. Still, there is a possibility of reoperation for locally increased mass and long-term follow up is needed with regular endoscopic and CT examination.

## Discussion

IP is one of the most common benign tumors of the nasal cavity and the paranasal sinuses. Although it is known to be benign tumor, as it tends to recur about 15% to 20%, the treatment of choice for IPs has been complete resection.<sup>1)</sup>

The most common IP origin site in Korea is maxillary sinus followed by anterior ethmoid sinus and nasal cavity.<sup>3)</sup> However, IPs may present outside the sinonasal tract. It is occasionally reported that IP cases which developed in the lacrimal drainage apparatus.<sup>4-11)</sup> The occurrences of IP in lacrimal sac may be due to the direct invasion of a disease into the lacrimal drainage apparatus or ectopic migration of Schneiderian membrane during the period of embryology. Recent review of literature presented that the most commonly presenting symptoms of IPs in lacrimal sac were medial canthal mass followed by epiphora. The median age of onset was 34.5 with slight female preponderance. Among the 6 cases reviewed, orbital invasion was found in 2 cases with more aggressive behavior.<sup>11)</sup>

Most of the treatment strategies were complete resection which included open approach by lateral rhinotomy or combined open and endoscopic approach. Radiation therapy was also added when there was an evidence for malignant transformation. Still, there is no comparative study on which surgery could lower the recurrence rate after the primary surgery and there is even a case report of tumor recurrence after extensive radical surgery including orbital exenteration.<sup>10)</sup> Without evidence that extensive radical surgery can

lower the recurrence rate, the case which invades lacrimal apparatus system raises the question as to what is the most appropriate management of IPs. There is still a controversy on the oncological safety of orbit or functional and psychosocial loss of orbit.

IP that invades lacrimal sac and periorbita is uncommon and treatment strategy may differ depends on surgeon's philosophy on orbit invading tumor. With abundance of experience in surgical strategy for IPs, authors decided to perform stage surgery on this patient and had a good outcome until 18 months after the surgery.<sup>12-14)</sup> These days, endoscopic resection of sinonasal IP has been gaining popularity around the world.<sup>1)</sup> The current concept of the management of IP is dissection of the involved mucosa along the subperiosteal plane and drill out of underlying bone. With the help of endoscopic instruments and techniques, surgeon can verify a tumor with excellent visualization and remove the tumor. Also the recurrence can be detected early with regular endoscopic follow ups.

Authors encountered first IP which invaded lacrimal sac and periorbita in Korea and performed the endoscopic stage surgery while sparing the globe considering patient's morbidity. With the help of brilliant illumination with endoscope, IP around the lacrimal sac was successfully removed and there was no evidence of carcinoma in situ transformation.

Still, as the IP recurrence is reported after 3 years of surgery,<sup>3)</sup> it is essential to maintain clinical and radiographic follow-up of the patient to ensure continued tumor control.

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