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Case Report

Inverted papilloma originating from the lacrimal sac and the nasolacrimal duct with marked FDG accumulation ☆

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ABSTRACT

Inverted papilloma originating from the lacrimal sac and the nasolacrimal duct is rare, although that in the sinonasal region is a relatively common lesion with local invasion, malignant potential and high recurrence rates after surgery. We report a 52-year-old woman with inverted papilloma of the right lacrimal sac and the nasolacrimal duct, who underwent CT, MR imaging and FDG-PET/CT preoperatively. In addition to CT and MR imaging features similar to those in previous reports, the inverted papilloma exhibited marked FDG accumulation with a maximum standardized uptake value of 7.34 and no other significant FDG accumulation was detected. In summary, our case of inverted papilloma originating from the right lacrimal sac and the nasolacrimal duct noted marked FDG accumulation on PET/CT, which enabled visualization of the localized tumor extension with no metastases.

☆ Competing Interests: The authors declare no conflicts of interest associated with this manuscript.

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Introduction

Inverted papilloma originating from the lacrimal sac and the nasolacrimal duct, which is rare as compared to that originating in the sinonasal region, shows high attenuation on CT and T1 iso-intensity on MR imaging in previous reports [1–8]. In addition to the CT and MR imaging findings, we describe FDG-PET/CT findings and clarify the usefulness of FDG-PET/CT for visualization of tumor extension and evaluation of metastases in FDG-avid inverted papilloma.

Case report

A 52-year-old woman presented to our hospital with a 3-year history of haemolacria and ophthalmalgia. Laboratory findings were not significant. CT revealed a highly attenuated mass of 3 cm in diameter in the right lacrimal sac and the nasolacrimal duct without bone destruction (Fig. 1A and B). The differential diagnoses of tumors in this region include benign epithelial tumors, such as papilloma, oncocytoma,

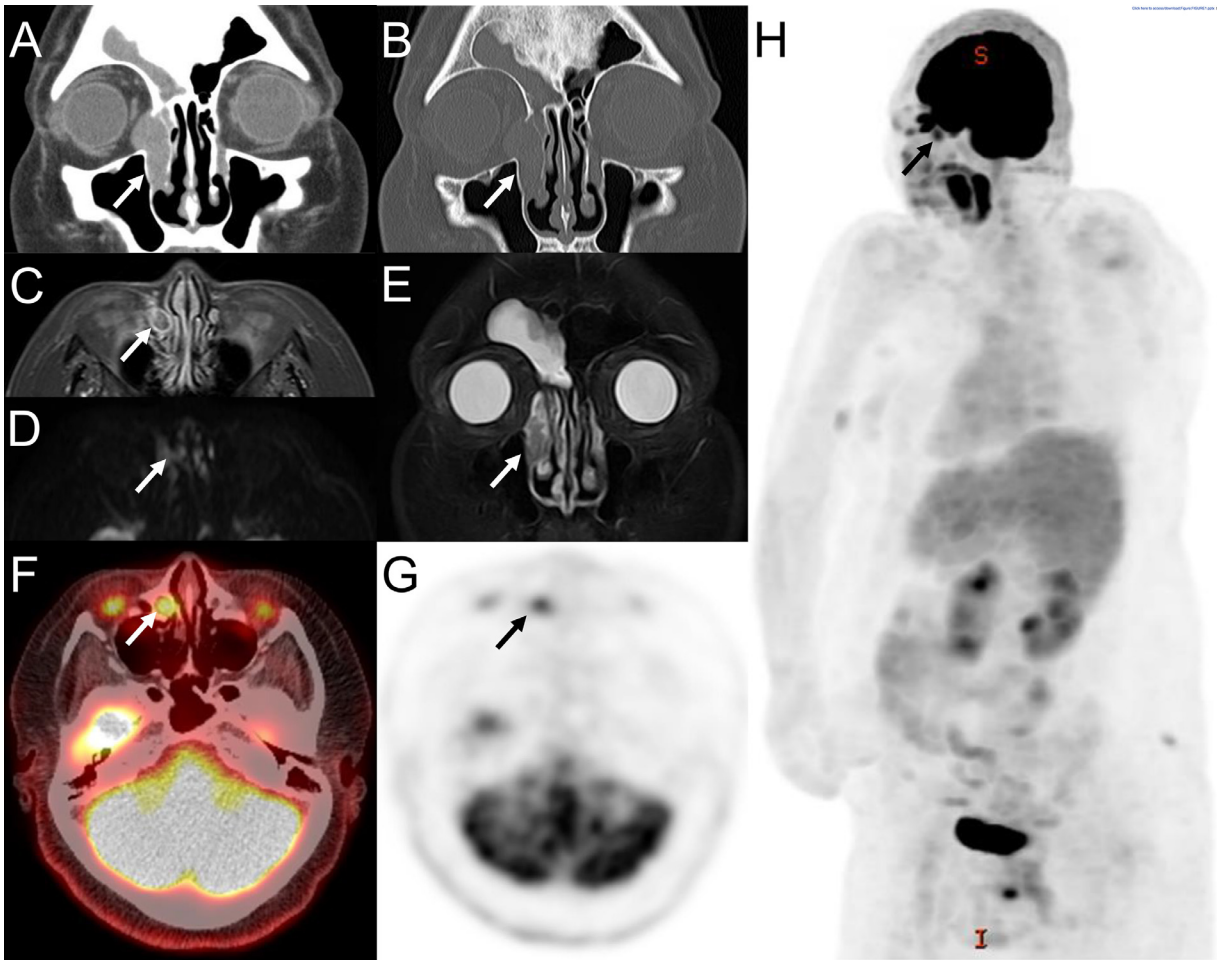

Fig. 1 – An attenuated mass of 3 cm in diameter in the right lacrimal sac and the nasolacrimal duct without bone destruction was observed on CT (A and B, arrow). MR imaging showed T1 iso-intensity, T2 hypo-intensity, minimal gadolinium enhancement (C, arrow) and no apparent restricted diffusion (D, arrow). STIR imaging demonstrated a convoluted cerebriform pattern (E, arrow). FDG-PET/CT revealed marked FDG accumulation with a SUVmax of 7.34 (F, fusion image; G, PET).

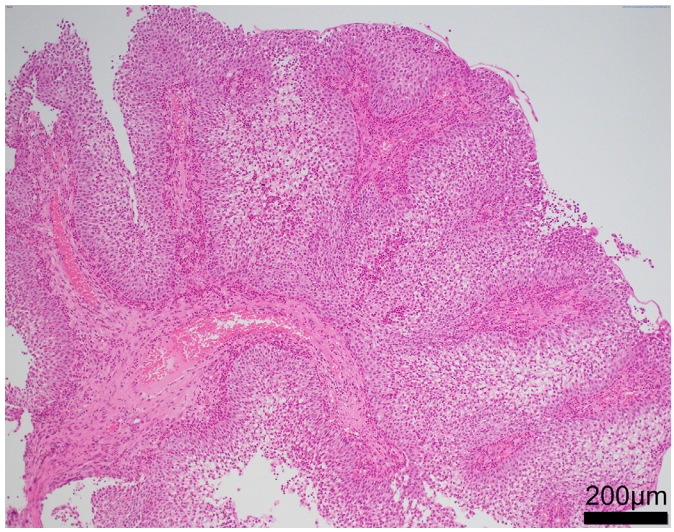

Fig. 2 – Histological investigation (hematoxylin-eosin staining, 10x) revealed hyperplasia of the epithelium with an endophytic growth pattern, which was correlated with the convoluted cerebriform pattern on MR imaging, confirming inverted papilloma with no malignant findings.

adenoma and cylindroma, malignant epithelial tumors, such as squamous cell carcinoma, transitional cell carcinoma, oncocytic adenocarcinoma, mucoepidermoid carcinoma and adenoid cystic carcinoma, and non-epithelial tumors such as lymphoma, melanocytic tumors, and mesenchymal tumors. In order to investigate histologically, endoscopic biopsy was performed and the mass was pathologically diagnosed as a papillomatous neoplasm without malignancy. MR imaging demonstrated T1 iso-intensity, T2 hypo-intensity, minimal gadolinium enhancement (Fig. 1C) and no apparent restricted diffusion (Fig. 1D). Moreover, a convoluted cerebriform pattern (Fig. 1E) often noted in sinonasal inverted papilloma was noted on STIR imaging. FDG-PET/CT revealed marked FDG accumulation (Fig. 1F, G, and H) with a maximum standardized uptake value of 7.34. No other significant FDG accumulation was detected (Fig. 1H). Taken together, a localized FDG-avid papillomatous neoplasm developed in a rare region of the lacrimal sac and the nasolacrimal duct without lymph node involvement or distant metastases.

Based on these imaging findings and biopsy, the mass was endoscopically excised with a clear margin and histology revealed hyperplasia of the epithelium with an endophytic growth pattern (hematoxylin-eosin staining, Fig. 2). No malignancy was observed. The final histological diagnosis was inverted papilloma. Although the resection margin was negative, local recurrence developed one year after the first surgery, requiring a second surgery. There was no recurrence thereafter.

Discussion

Approximately ten cases of inverted papilloma originating from the lacrimal sac and the nasolacrimal duct have been reported to date [1–10]. In previous reports, the inverted papilloma originating from the lacrimal sac and the nasolacrimal duct showed high attenuation on CT and T1 iso-intensity on MR imaging [1–8]. Our case exhibited CT and MR imaging features similar to those in previous reports, and a convoluted cerebriform pattern was depicted on MR imaging, which may be correlated with inverted or endophytic growth of the tumor. In addition, marked FDG accumulation in the inverted papilloma in this rare region was noted in our case.

Sinonasal inverted papillomas with marked FDG accumulation are often malignant [11–13]; however, benign cases may have also marked FDG accumulation [14]. Therefore, we paid attention when assessing malignancy in our case of inverted papilloma in the lacrimal sac and the nasolacrimal duct regardless of increased FDG accumulation. Moreover, FDG-PET/CT may accurately visualize tumor extension, and aid in the evaluation of lymph nodes and distant metastases in cases of FDG-avid inverted papilloma. In conclusion, our case of rare inverted papilloma originating from the lacrimal sac and the nasolacrimal duct had marked FDG accumulation on PET/CT, which enabled visualization of the localized tumor extension with no metastases.

Patient consent

Written and informed consent was received from the patient.

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