

Localized Maxillary Sinus Papilloma: Management of Incidental Lesion

Motoki SEKINE, Fumiyuki GOTO, Kosuke SAITO, Shoji KANEDA,
Hikaru YAMAMOTO, Tomoaki MURAKAMI and Kenji OKAMI

Department of Otolaryngology, Tokai University School of Medicine

(Received October 13, 2020; Accepted December 2, 2020)

Most maxillary sinus papillomas are confirmed when they have extended beyond the nasal cavity and are rarely found while localized in the maxillary sinus. We experienced two cases of localized papilloma in the maxillary sinus. Case 1 was a 69-year-old man with a localized left maxillary sinus lesion detected during a routine imaging examination. As the lesion was likely to be papilloma, we recommended that the patient undergo diagnostic surgery, which he refused. He experienced bloody rhinorrhea 1 year and 9 months after the first visit, and computed tomography (CT) showed increased lesions and bone destruction. Histological examinations revealed squamous cell carcinoma ex inverted papilloma. He died 5 years after the first visit. Case 2 was a 46-year-old woman in whom positron emission tomography/CT showed a localized right maxillary sinus lesion. Tissue biopsy results indicated oncocytic papilloma. Endoscopic resection was performed later. On an imaging examination, sinonasal papilloma was determined accidentally to be a localized lesion of the maxillary sinus. A detailed interpretation of the CT scan was useful in estimating sinonasal papilloma. Tissue biopsy or diagnostic surgery should be performed when sinonasal papilloma is suspected during appropriate image evaluation.

Key words: sinonasal papilloma, localized lesion, incidental lesion, malignant transformation, case report

INTRODUCTION

Sinonasal papilloma (SP; synonym: Schneiderian papilloma) is locally aggressive, has a high recurrence rate postoperatively, and is associated with malignant lesions [1, 2]. Most maxillary sinus papillomas are confirmed when they have extended to the nasal cavity [3] and are rarely found when they are localized in the maxillary sinus. No previous reports have discussed a localized maxillary sinus papilloma. In addition, there is no consensus on the diagnosis and management of SP in this condition. Here, we report two representative and educational cases of localized papilloma in the maxillary sinus, in which detailed interpretations of computed tomography (CT) scans were useful in estimating SP.

CASE REPORTS

Case 1 was a 69-year-old man with a history of cervical malignant lymphoma. A sinus lesion was found accidentally during a routine imaging examination. The CT scan showed an irregular marginal mass shadow in the left maxillary sinus and hyperostosis on the upper wall, suspected to be the tumor attachment site (Fig. 1). Nasal endoscopy revealed no lesion exposure to the nasal cavity. The lesion size had not changed as compared with the CT image taken 1 1/2 years previously. As the lesion was likely to be SP, we recommended that he undergo diagnostic surgery, which he refused. He experienced bloody rhinorrhea 1 year and 9 months after the first visit, and CT revealed increased lesions and bone destruction (Fig. 2A). Tissue

biopsy of the nasal mass showed inverted papilloma (Fig. 2B), and a biopsy of the maxillary sinus tumor by external incision revealed squamous cell carcinoma ex inverted papilloma (Fig. 2C, D). The diagnosis was maxillary SCC (T4bN0M0, UICC 8th edition) and indicated concurrent chemoradiotherapy based on the judgment of inoperable lesion. A complete response was obtained with concurrent chemoradiotherapy with CDDP 80 mg/m², repeated three times every 21 days, and 66 Gy radiotherapy in 33 fractions. However, the patient had local recurrence and cervical spine metastasis and ultimately died 5 years after the first visit.

Case 2 was a 46-year-old woman in whom positron emission tomography/CT incidentally showed FDG accumulation in the cervical lymph nodes and right maxillary sinus on the evaluation of an ovarian lesion (Fig. 3A). A CT scan revealed an irregular marginal mass shadow in the right maxillary sinus (Fig. 3B). Slight hyperostosis was observed on the posteroinferior wall (Fig. 3C). Nasal endoscopy revealed no abnormalities in the right middle nasal meatus. Fine-needle aspiration cytology of the cervical lymph node yielded a result of class 2. We considered the lesion to be either SP or metastatic lesions or maxillary sinus cancer with cervical lymph node metastasis. To obtain a final diagnosis, we performed a tissue biopsy of the maxillary sinus lesion under local anesthesia. The maxillary ostium was enlarged via the middle nasal meatus and was sufficient to view the tumor localized at the posteroinferior wall. The lesion was partially resected for biopsy. Pathological diagnosis was oncocytic papilloma. It was later completely resected by endoscopic sinus surgery

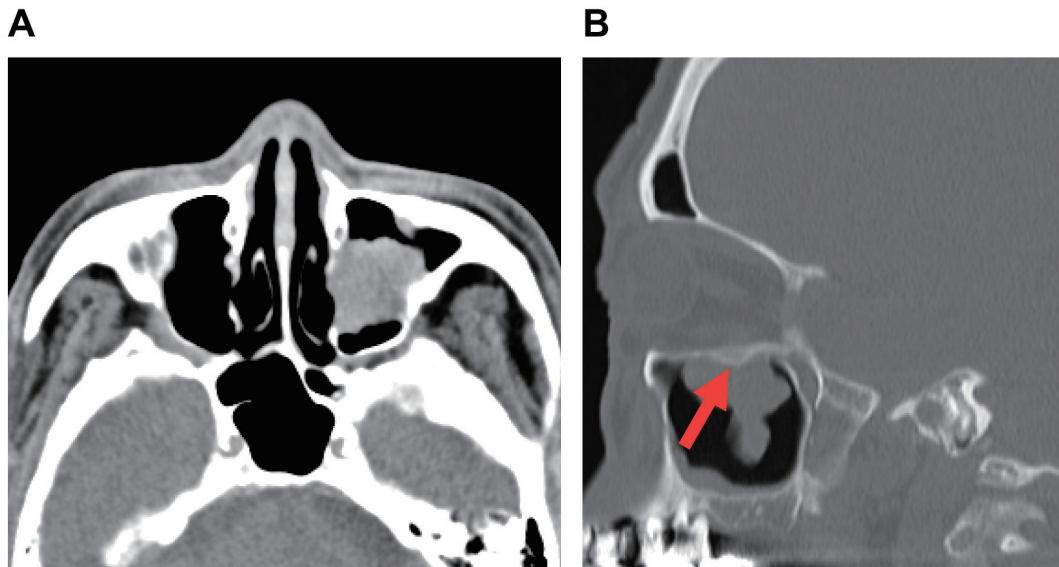


Fig. 1 Case 1
A: A horizontal CT scan showing irregular marginal mass shadows in the left maxillary sinus.
B: A coronal CT scan showing hyperostosis on the upper wall in contact with the mass lesion (arrow).

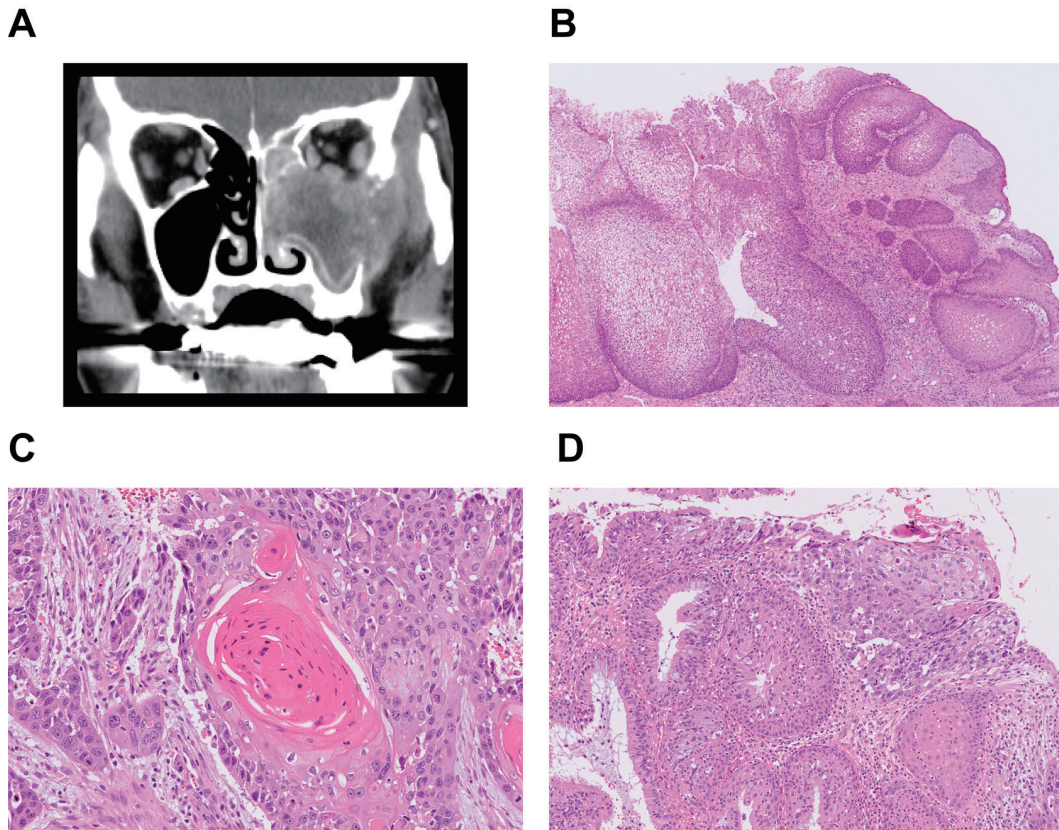


Fig. 2 Case 1
A: A coronal CT scan 1 year and 9 months after the initial examination. The lesions are increasing and extending to the orbit and pterygopalatine fossa, with bone destruction.
(B-D): Microscopic findings (hematoxylin and eosin stain): B, nasal biopsy specimen; C, D, maxillary sinus specimens by external incision.
B: Nasal biopsy specimen showed inverted proliferation of the squamous epithelium without atypia, which was diagnosed as inverted papilloma.
C: Maxillary sinus specimen showed sheet-like or trabecular proliferation of atypical cells with keratinization, which was diagnosed alongside differentiated squamous cell carcinoma.
D: Most specimens were composed of squamous cell carcinomas, but inverted papilloma components were focally observed (lower field).

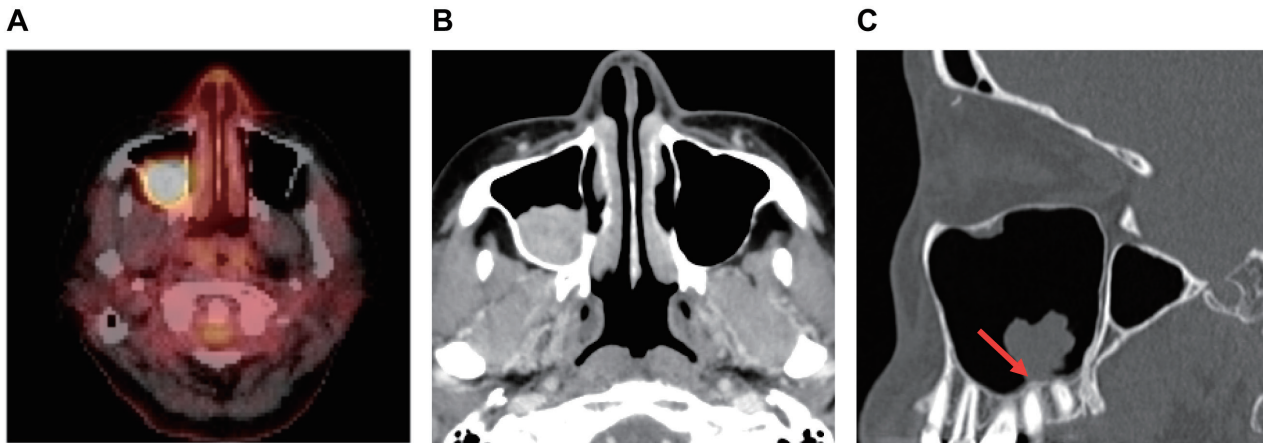


Fig. 3 Case 2 (A: PET/CT; B, C: CT)

A: FDG accumulation is observed in the right maxillary sinus (SUV max: 10.7).

B: An axial CT scan showing an irregular marginal mass shadow in the right maxillary sinus.

C: A sagittal CT scan showing slight hyperostosis on the posteroinferior wall in contact with the mass lesion (arrow).

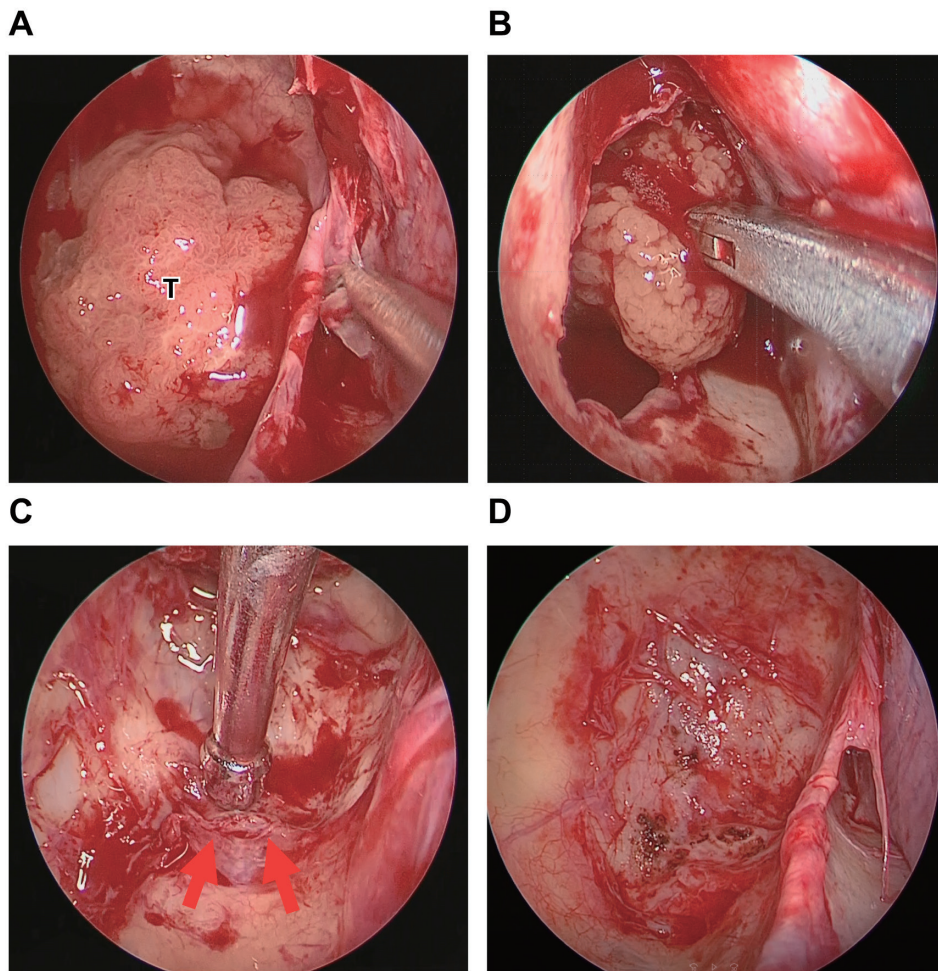


Fig. 4 Case 2 surgical findings

A: A tumor localized in the posteroinferior wall of the maxillary sinus.

B: The tumor was resected en bloc with the mucosa at the tumor attachment.

C: Slight hyperostosis was observed at the tumor attachment site (arrow).

D: Thickened bone was removed, and marginal mucosa was resected.

T: Tumor

for radical treatment (Fig. 4). The patient has no signs of recurrence after 2 years and 4 months of follow-up.

DISCUSSION

The course of these patients resulted in two important clinical suggestions: (1) SP can be detected acci-

dentally as a localized lesion of the maxillary sinus on imaging examination and (2) a detailed interpretation of CT scans is useful in estimating papillomas of this condition.

First, SP can be detected accidentally as a localized lesion of the maxillary sinus on imaging examination.

No previous reports have discussed localized maxillary sinus papillomas. We experienced two cases of accidentally detected localized papillomas in the maxillary sinus. Many maxillary sinus papillomas are identified after they have extended into the nasal cavity and the patient presents with nasal complaints. A study of 82 cases of SP reported that 38 of 40 cases with maxillary sinus lesions also had lesions in the middle nasal meatus [3]. If a lesion is found in the nasal cavity, it can be differentiated from inflammatory disease by endoscopic inspection; histological examination is easy, and a diagnosis can be obtained easily. On the other hand, most lesions localized in the maxillary sinus are not directly visible by endoscopic inspection, except in cases with a large accessory ostium of the maxillary sinus, and judgment is based on imaging findings only. When it is difficult to distinguish a maxillary sinus lesion from an inflammatory lesion, it is unclear how much examination and treatment should be considered. Clinicians must be aware that papilloma may be present in localized lesions of the maxillary sinus that are found accidentally on imaging.

Second, a detailed interpretation of CT scans was useful in estimating this papilloma. For patients without lesions in the nasal cavity, the detection of characteristic findings of SP using imaging examination is important. In SP, CT often shows osteitic bone changes at the tumor attachment site [4–7]. Therefore, in cases of suspected SP, it is important to carefully confirm on CT the presence of osteitic bone changes on the wall that contacts the mass lesion. In these two cases, hyperostosis was confirmed by CT, which was the key to strongly suspecting SP. In addition, the air-space area was recognized around the tumor shadow, and an irregular marginal mass shadow was confirmed. If SP is suspected based on imaging findings, performing tissue biopsy, or diagnostic surgery is advisable.

Previous studies have reported that SP is associated with malignancy (synchronous or metachronous) in 4–11% of cases [3, 8–11]. SP is classified into three major histological types [12]: inverted, oncocytic, and exophytic, and all types have been reported to be associated with malignant lesions [13, 14]. In case 1, 1 year and 9 months after the first visit, the tumor underwent malignant transformation. CT findings of SP associated with malignancy include rapid growth, bone erosion/destruction, and invasion of the adjacent structure [15–16]. By comparing the CT scan taken at the first visit with those taken 1 1/2 years before presentation and 1 year after, no change was seen in lesion size, so we considered it highly likely that it had become malignant after the first visit. In such cases, it is highly possible that the prognosis can be improved by treating the lesion at the benign stage. Describing the SP as a “benign tumor” may result in the patient underestimating the disease. Patients should be informed that SP is a “benign tumor,” but that it is “not a good” tumor and may have coexisting malignant lesions and malignant transformation, and the disease severity should be communicated fully.

In conclusion, SP can be detected accidentally as a localized lesion of the maxillary sinus during imaging examinations. Detailed interpretations of CT scans are useful in estimating SP in this condition. Clinicians must be aware that SP may be present in localized

lesions of the maxillary sinus and may be found accidentally during imaging. With the increasing opportunity of imaging examinations, such localized lesions will more likely be detected accidentally. Lesions should be estimated by the appropriate image evaluation, and if SP is suspected, biopsy via the middle nasal meatus or diagnostic surgery should be performed. The early detection and treatment of localized SP associated with malignancy might improve the prognosis. Further reports should be accumulated to determine whether there is a greater frequency of localized maxillary sinus papilloma.

ACKNOWLEDGMENTS

I am deeply grateful to Kenichi Hirabayashi from the Department of Pathology at Tokai University School of Medicine. The authors would like to thank Enago (www.enago.jp) for the English language review.

DATA AVAILABILITY

Primary data about the patient were obtained from the electronic medical records of the Tokai University School of Medicine. Cited manuscripts were found on PubMed.

FUNDING STATEMENT

None.

ETHICAL STATEMENT

The present study was approved by the Institutional Review Board for Clinical Research of the Tokai University School of Medicine (20R-177). The study was carried out in accordance with the Code of Ethics of the World Medical Association (Helsinki Declaration). The institutional review board takes responsibility for the anonymization of the patient and the informed consent has been waived.

REFERENCES

- 1) Lisan Q, Antoine Moya-Plana A, Bonfils P. Association of Krouse classification for sinonasal inverted papilloma with recurrence: A systematic review and meta-analysis. *JAMA Otolaryngol Head Neck Surg* 2017; 143: 1104–10.
- 2) Lisan Q, Laccourreye O, Bonfils P. Sinonasal inverted papilloma: From diagnosis to treatment. *Eur Ann Otorhinolaryngol Head Neck Dis* 2016; 133: 337–41.
- 3) Buchwald C, Franzmann MB, Tos M. Sinonasal papillomas: A report of 82 cases in Copenhagen County, including a longitudinal epidemiological and clinical study. *Laryngoscope* 1995; 105: 72–9.
- 4) Chiu AG, Jackman AH, Antunes MB, Feldman MD, Palmer JN. Radiographic and histologic analysis of the bone underlying inverted papillomas. *Laryngoscope* 2006; 116: 1617–20.
- 5) Yousuf K, Wright ED. Site of attachment of inverted papilloma predicted by CT findings of osteitis. *Am J Rhinol* 2007; 21: 32–6.
- 6) Lee DK, Chung SK, Dhong HJ, Kim HY, Bok KH. Focal hyperostosis on CT of sinonasal inverted papilloma as a predictor of tumor origin. *Am J Neuroradiol* 2007; 28: 618–21.
- 7) Bhalla RK, Wright ED. Predicting the site of attachment of sinonasal inverted papilloma. *Rhinology* 2009; 47: 345–8.
- 8) Mirza S, Bradley PJ, Acharya A, Stacey M, Jones NS. Sinonasal inverted papillomas: recurrence, and synchronous and metachronous malignancy. *J Laryngol Otol* 2007; 121: 857–64.
- 9) Krouse JH. Endoscopic treatment of inverted papilloma: Safety and efficacy. *Am J Otolaryngol* 2001; 22: 87–99.
- 10) Kim DY, Hong SL, Lee CH, Jin HR, Kang JM, Lee BJ, *et al.* Inverted papilloma of the nasal cavity and paranasal sinuses: a

- Korean multicenter study. *Laryngoscope* 2012; 122: 487-94.
- 11) Re M, Gioacchini FM, Bajraktari A, Tomasetti M, Kaleci S, Rubini C, *et al.* Malignant transformation of sinonasal inverted papilloma and related genetic alterations: a systematic review. *Eur Arch Otorhinolaryngol* 2017; 274: 2991-3000.
 - 12) Hyams VJ. Papillomas of the nasal cavity and paranasal sinuses. A clinicopathological study of 315 cases. *Ann Otol Rhinol Laryngol* 1971; 80: 192-206.
 - 13) Kapadia SB, Barnes L, Pelzman K, Mirani N, Heffner DK, Bedetti C. Carcinoma ex oncocytic Schneiderian (cylindrical cell) papilloma. *Am J Otolaryngol* 1993; 14: 332-8.
 - 14) Nudell J, Chiose S, Thompson LD. Carcinoma ex-Schneiderian papilloma (malignant transformation): a clinicopathologic and immunophenotypic study of 20 cases combined with a comprehensive review of the literature. *Head Neck Pathol* 2014; 8: 269-86.
 - 15) Miyazaki T, Haku Y, Yoshizawa A, Iwanaga K, Fujiwara T, Mizuta M, *et al.* Clinical features of nasal and sinonasal inverted papilloma associated with malignancy. *Auris Nasus Larynx* 2018; 45: 1014-19.
 - 16) Yan CH, Tong CCL, Penta M, Patel VS, Palmer JN, Adappa ND, *et al.* Imaging predictors for malignant transformation of inverted papilloma. *Laryngoscope* 2019; 129: 777-82.