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Bilateral oncocytic-type Schneiderian papilloma in a pediatric patient☆,☆☆

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ABSTRACT

Oncocytic-type papilloma is the rarest histologic type of all sinonasal papillomas, and usually present with a unilateral mass in adults. Histologically, oncocytic Schneiderian papilloma (OSP) has an exophytic and endophytic growth pattern with oncocytic features. As in inverted papillomas, the potential of malignant transformation and the rate of recurrence are high in cases of oncocytic papilloma.

In this report, we present a 16 year old girl with OSP that originated from both maxillary sinuses, who underwent endoscopic transnasal bilateral medial maxillectomy.

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Introduction

Oncocytic Schneiderian papilloma (OSP), also known as cylindrical or columnar cell papilloma, is the rarest histologic type of all sinonasal papillomas and represents 1–19% of all sinonasal papillomas [1–4]. OSP originates from the Schneiderian membrane, and is histologically characterized by an endophytic and exophytic growth pattern of columnar cells with oncocytic features [2]. OSP usually occur unilaterally; the most common location is the maxillary sinus, and rarely, the ethmoid and sphenoid sinus [1,3].

We herein report a case of oncocytic Schneiderian papilloma occurred in a 16 year old girl bilaterally from the maxillary sinuses. To our knowledge this is the youngest patient reported in the literature with OSP occuring bilaterally.

Case report

A 16 year old girl presented to an otorhinolaryngologist with bilateral progressive nasal obstruction and rhinorrhea of 6 months and she underwent surgery with a preliminary diagnosis of nasal polyposis. However due to massive bleeding, the operation could not be performed and stopped after taking biopsy from both nasal cavities. The result of the biopsies from both nasal cavities was found to be OSP. The patient was then referred to our department for further evaluation.

Our examination by nasal rigid endoscopy showed pale and fleshy polypoid mass with purulent secretions in both nasal cavities. A computerized tomography (CT) scan revealed a soft tissue density occupying maxillary sinuses and nasal cavities bilaterally with bone erosion (Fig. 1).

In addition, two independent but similar masses that had typical cerebriform-columnar pattern with heterogeneously linear enhancement were detected bilaterally in the maxillary sinuses on T1 and T2-weighted magnetic resonance imaging (MRI) sections (Fig. 2).

Endoscopic endonasal bilateral medial maxillectomy was then performed and the tumor was completely removed bilaterally. Final histopathology was reported as bilateral OSP showing an exophytic and endophytic growth pattern. Microscopically the resected tumor from both sides was revealed to be multilayered columnar cells with finely granular oncocytic cytoplasms and small cysts filled by

☆ The informed consent form has been confirmed to the parents of the patient due to the age of the patient.

☆☆ This manuscript has not previously been presented any meeting.

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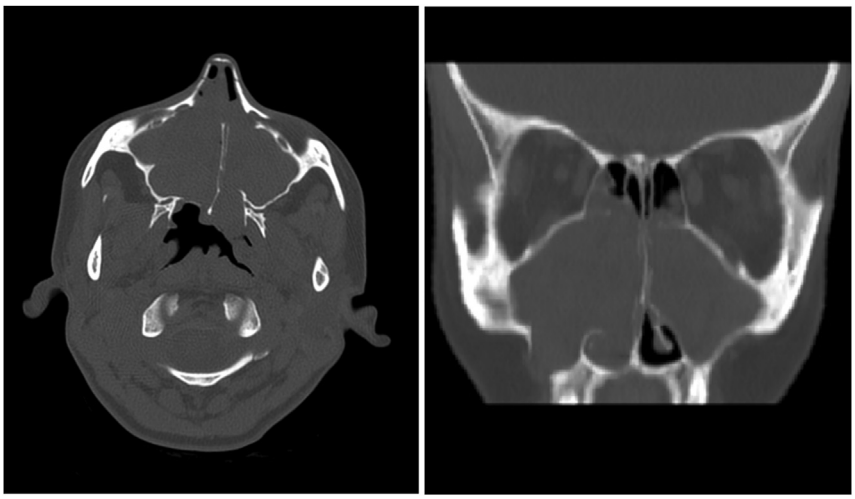


Fig. 1. Computerized tomography (CT) demonstrates a soft tissue density in both maxillary sinuses with bone erosion.

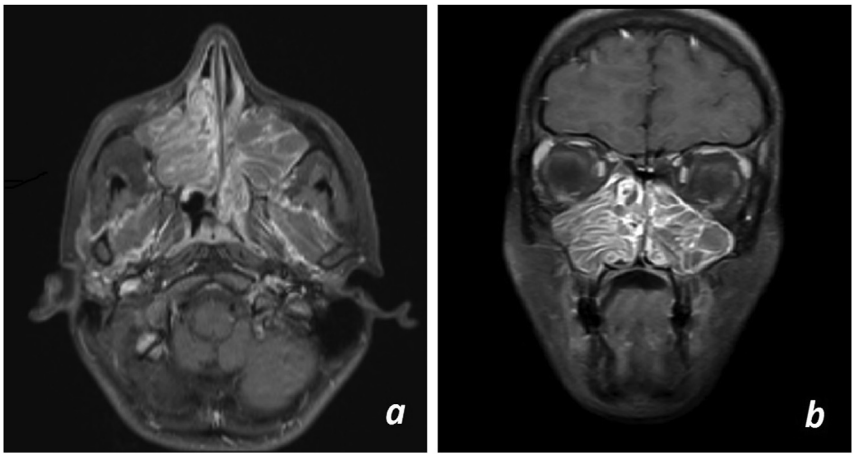


Fig. 2. Magnetic resonance imaging (MRI) findings of two independent oncocytic papillom show typical cerebriform pattern. *a. T1 axial section. b. T1 coronal section*.

neutrophils or mucin seen in the epithelium. The stroma was edematous and fibrotic, and contained inflammatory cells (Fig. 3). No complication was observed in the postoperative period and no recurrence was seen at the 1-year follow-up.

Discussion

Sinonasal papillomas were divided into three histologic groups by Hyams: inverted, fungiform, and oncocytic [1]. Oncocytic Schneiderian papilloma is the rarest subtype, and it is important because malignant transformation and recurrence are more frequent than inverted papilloma [2,3,5].

Oncocytic Schneiderian papilloma usually occurs in patients aged over 50 years [2,4,5]. Unlike other types of sinonasal papillomas, OSP has no sex dominance. To date, there are 3 cases of oncocytic papilloma diagnosed at a young age. A 26-year-old male patient reported by Olusina et al. had unilateral disease [6]. The second case was a 28-year-old patient with left-side OSP in the

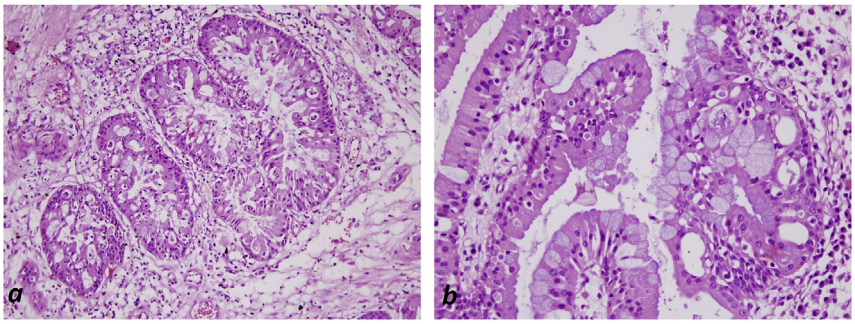


Fig. 3. Histopathologic examination shows *(a)* papillomatous lesion, (*H-E, x200*), and *(b)* stratified columnar epithelium with oncocytic features and microcysts, (*H-E, x400*).

presence of chronic rhinosinusitis, presented by Cheng et al. [7]. Also, the case reported by Levine et al. was a 33-year-old woman with unilateral disease [8]. In this context, our patient is the youngest to present with OSP to date. In addition, OSP almost always appears as a unilateral mass, and bilaterality is very rare unlike inverted papilloma, another type of Schneiderian papilloma. Only one case of bilateral OSP was reported in the study of Karligkiotis et al. with 33 patients (3.1%) [5]. Besides, there is no case of bilateral oncocytic papilloma published in the literature. Hence, this is a second distinctive characteristic of our case.

From the series published in the literature, oncocytic papilloma has been determined as mostly originating from the maxillary sinus, rather than the lateral nasal wall, predominantly as inverted papilloma [1–3]. In our case, in accordance with the literature, the tumors arose from the maxillary sinus on both sides.

In the study of Vorasubin et al., no malignancy was observed in any of the 9 patients with OSP, whereas the malignancy rate of patients with OSP in the larger series ranged from 3% to 17% [2–5,9]. In patients with oncocytic papillomas, squamous cell carcinoma is mostly seen, such as in inverted papilloma, and mucoepidermoid and sinonasal undifferentiated carcinomas have also been reported [2,3,9]. Although synchronous malignancy was not detected in our case, the patient should be followed closely because the rate of metachronous malignancy in oncocytic papilloma is not yet fully known.

Conclusion

Oncocytic Schneiderian papilloma is the rarest subtype of sinonasal papilloma, generally seen as a unilateral mass in patients aged over 50 years. The striking features of our case are its bilaterality, and the age of the patient; she is the youngest patient reported to date. In addition, in the event of excessive bleeding during nasal polyposis surgery, the surgeon should think about the possibility of a Schneiderian papilloma or malignancy. If required, the operation should be interrupted following a biopsy and definitive treatment should be planned according to the pathology result.

Conflict of interest

There is no conflict of interest among the authors.

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