

Oncocytic Schneiderian Papilloma Found in A Recurrent Chronic Paranasal Sinusitis

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Oncocytic schneiderian papilloma (OSP), also known as cylindrical cell papilloma, is a rare benign sinonasal neoplasm. Due to its rare incidence, this disease is not well-understood by clinicians. The probability of malignancies arising in OSP is not low but they are rarely encountered in clinical practice. To the best of our knowledge, there is no documentation of OSP associated with recurrent chronic paranasal sinusitis (CPS). We hereby report an unusual case of OSP in a 28 year-old man which was first diagnosed and treated as recurrent chronic paranasal sinusitis. After three surgeries, OSP was revealed in the pathological findings and the tumor was completely resected endoscopically in the fourth operation. There was no sign of recurrence after two years of follow-up. A thorough review of histopathological slides from the first two surgeries showed no sign of OSP in these tissue sections. We present this case to remind clinicians that OSP can be found with inflammatory polyps in CPS, and it is easily overlooked clinically. Nevertheless, sufficient alertness from both the surgeon and pathologist is the cornerstone for the proper diagnosis and appropriate treatment of this rare nasal neoplasm. (*Chang Gung Med J 2006;29:336-41*)

Key words: oncocytic schneiderian papilloma, inverted papilloma, recurrent chronic paranasal sinusitis, endoscopy.

The Schneiderian epithelium, an ectodermally derived respiratory mucosa lining the nasal cavity and paranasal sinuses, can give rise to 3 histologically distinct papillomas, fungiform (exophytic, septal, squamous), inverted, and cylindrical cell papilloma.⁽¹⁻⁵⁾ Inverted papillomas account for 47% of all sinonasal papillomas and are the most commonly seen among the three benign neoplasms in rhinological practice.^(1,2,3,5) On the other hand, cylindrical cell papilloma (CCP), which was renamed oncocytic schneiderian papilloma (OSP) by Barnes and Bedetti,⁽³⁾ is the rarest subtype, accounting for only 3% to 5% of all sinonasal papillomas.^(1,2,5) It is easily overlooked on physical examination and often misdiagnosed as an inverted papilloma. A comparison of

the clinical characteristics of these three kinds of papilloma is shown in Table 1. Patients with OSP are older than 50 years in most cases and there is no sex predilection.⁽⁴⁾ Initial symptoms usually include unilateral nasal obstruction, epistaxis, and pain. The duration of symptoms ranges from months to years. Physical examination generally shows a ragged, polypoid, red or pink intranasal mass, which can be confused with inflammatory polyp.⁽⁴⁾ Malignancies arising in OSP are rare, with invasive squamous cell carcinoma being the most frequently seen.⁽¹⁾ Herein we present an unusual case of OSP. This is the first case of OSP incidentally found in a patient with CPS reported in the English literature. We present this case to remind clinicians that OSP can be found with

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Table 1. Clinical Characteristics of the Three Kinds of Papilloma

	Location	Percentage of all sinonasal papilloma	Malignant change potential	Clinical symptoms	Age of prevalence	Treatment
FP	nasal septum, vestibule	45%~50%	0%	unilateral nasal obstruction	N/A	endoscopic excision, laser vaporization
IP	the lateral wall of the nose and the paranasal sinuses	45%~50% (slightly higher than fungiform papilloma)	5%~10%	unilateral nasal obstruction, epistaxis	50~60 year-old	medial maxillectomy, endoscopic excision, Caldwell-Luc approach
OSP	the lateral wall of the nose and the paranasal sinuses	3%~5%	14%~19%	unilateral nasal obstruction, epistaxis, pain	30~80 year-old most often > 50 year-old	a lateral rhinotomy with en-bloc resection of the lateral nasal wall, Caldwell-Luc approach

Abbreviations: FP: fungiform papilloma; IP: inverted papilloma; OSP: oncocytic schneiderian papilloma

*Table 1 summarized from references 1, 2, 3, 4, 5, and 6

inflammatory polyps in chronic paranasal sinusitis, and it is easily overlooked clinically.

CASE REPORT

A twenty-eight year-old man visited our clinic with a 3-month history of severe nasal obstruction and left nasal purulent discharge with a foul odor. Left nasal polyposis was noted on physical examination. Left chronic paranasal sinusitis (CPS), chronic hypertrophic rhinitis, and nasal septal deviation were our tentative diagnoses. Computed tomography also revealed a heterogenous soft tissue density over the left lateral nasal cavity, osteomeatal complex, and maxillary sinus. (Fig. 1) Subsequently, left functional endoscopic sinus surgery (FESS) with a septomucosal resection (SMR) was performed. A postoperative surgical specimen confirmed the preoperative diagnosis. The patient was discharged uneventfully and received regular follow-up in our clinic. He still suffered from left nasal obstruction and three months later returned to our clinic. Left nasal polyposis was diagnosed again. Under the impression of left recurrent CPS, he underwent revised FESS three months later. The postoperative surgical specimen still revealed left-side CPS. One year later, he suffered from left retro-orbital fullness with headache. A left-side nasal polyp with mucopurulent discharge was seen again on physical examination in our outpatient department (OPD). At that time, left-side recurrent CPS was suspected. Seven months later, he had a third operation, a left-side revised FESS. This time, the postoperative pathology revealed OSP as well as



Fig. 1 Computed tomography (coronal view) demonstrates a heterogenous soft tissue density over the left lateral nasal cavity, osteomeatal complex, and maxillary sinus.

inflammatory polyp of CPS. One year later, a left-side nasal recurrent tumor was observed on endoscopy in our OPD. Eight months later, under the impression of recurrence of OSP, radical endoscopic sinus surgery was done. The surgical specimen confirmed the diagnosis of OSP. The patient was discharged uneventfully and received regular follow-up in our clinic. The two-year follow-up showed no evidence of recurrence.

Under histological examination, nasal polyps composed of polypoid paranasal mucosa with edema

and mild chronic inflammatory cell infiltration were seen in specimens from the first two surgeries. (Fig. 2A, B) The pathology of the specimens obtained from the subsequent 2 surgeries demonstrated oncocytic epithelium composed of multiple layers of columnar cells with eosinophilic and granular cytoplasm, inspissated mucin, and intraepithelial collections of neutrophils. The growth pattern of both papillomas was predominantly exophytic and papillary, although a focal mildly inverted growth pattern

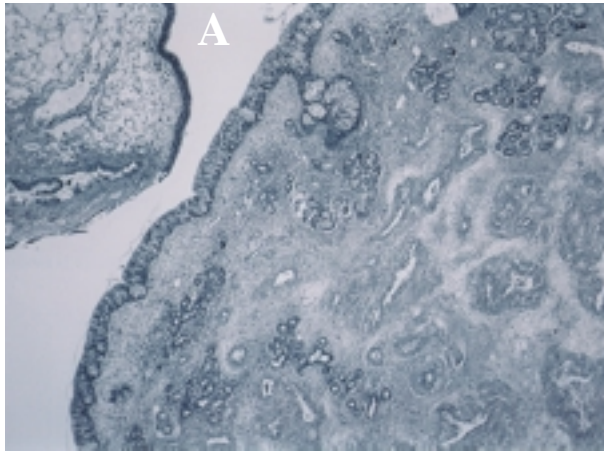


Fig. 2A H&E 40X Nasal polyp: two specimens of polypoid tissue lined by pseudostratified ciliated columnar epithelium. The tissue on the left is lined by epithelium with abundant mucous cells. There are seromucinous glands in the stroma. The stroma of both specimens of paranasal tissue is edematous.

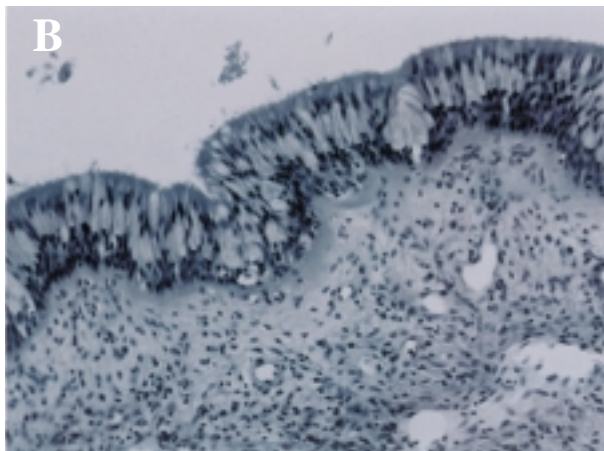


Fig. 2B H&E 200X Nasal polyps are characterized by a pseudostratified ciliated columnar epithelium and thickening of the epithelial basement membrane. Chronic inflammatory cells are seen in the subepithelial stroma.

was also observed. These findings indicate the diagnosis of typical cylindrical (oncocytic) papilloma. (Fig. 3A, B) After a thorough review of the histopathological slides of tissue sections from the first two surgeries, no OSP could be found.

DISCUSSION

Fungiform papilloma, inverted papilloma, and OSP are three morphologically distinct lesions aris-

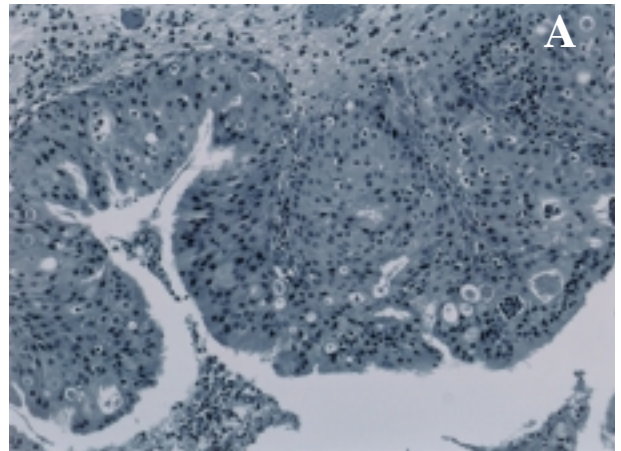


Fig. 3A H&E 40X Oncocytic schneiderian papilloma: papillary pattern of growth.

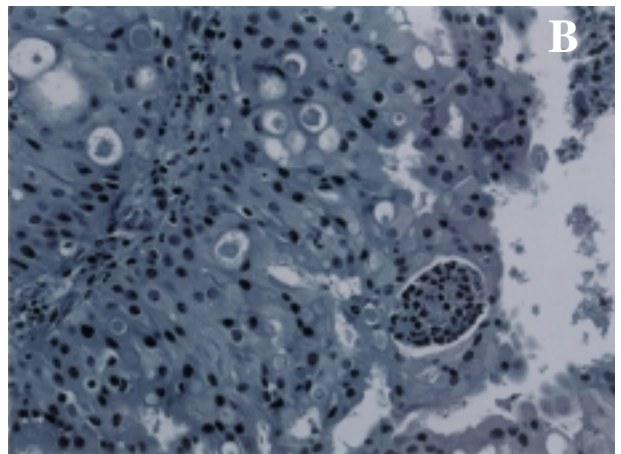


Fig. 3B H&E 400X Oncocytic schneiderian papilloma: high power view of the papillae. The epithelium is composed of several layers of columnar cells with abundant eosinophilic and granular cytoplasm. The nuclei are uniform and round, sometimes staining darkly in the superficial layers. Intraepithelial cysts containing mucin and large collections of neutrophils are seen here.

ing from the Schneiderian membrane.⁽¹⁻⁸⁾ However, there is still some controversy in the classification of sinonasal papillomas. Previously some researchers believed that all papillomas in the sinonasal tract were inverted papilloma; others used various synonyms such as Schneiderian papilloma, transitional (cell) papilloma, inverted or inverting papilloma, and endophytic papilloma.⁽⁷⁾ The histomorphologically based classification formulated by Hyams, in which papillomas of the sinonasal tract are classified as inverted papilloma, septal papilloma, and cylindrical cell papilloma, is probably the most accepted one.⁽⁴⁾ Fungiform papilloma, which is usually located on the nasal septum, has a risk of recurrence but is not associated with malignant potential.^(2,8) By contrast, both OSP and inverted papillomas involve the lateral wall of the nose and the paranasal sinuses, and have a potential for local invasion, and high frequency of recurrence if incompletely excised, and an increased risk of malignancy.^(1,2,4,5,8) Inverted papilloma has a definite association with malignancy. However, cylindrical cell papilloma (OSP) has been distinguished from other papillomas by its histological as well as cytologic features.

The oncocyctic features of OSP are due to mitochondrial hyperplasia, as they are in oncocytes of various tissues, such as oncocytoma, Hurthle cell, and Warthin's tumor.⁽²⁾ OSP occurs in patients from 33 to 83 years old; most patients are over 50 years of age.^(2,5) There is no sex predilection, in contrast to the male predominance in inverted and fungiform papillomas. Although it has features similar to inverted papilloma, it appears to have a higher frequency of association with malignancy (10% to 17%) than inverted papilloma (5% to 10%).⁽²⁾ However, the incidence of OSP is much lower than that of inverted papilloma.

Unilateral nasal obstruction is the most common presenting symptom for OSP. Intermittent epistaxis and pain are also reported. Rhinorrhea, sinusitis and allergic symptoms are rarely described. The duration of symptoms is usually months to years. All cases known to date have been unilateral. A gross shaggy or papillary configuration is most typical. However, smooth, edematous, polypoid lesions are not uncommon.⁽⁵⁾ Routine sinus radiographs reveal abnormalities confined to the ipsilateral sinonasal passages. Sinus opacification associated with an intranasal soft tissue density is the predominant finding. Bony

destruction on plain radiographs and tomography could be more suggestive of coexistent malignant disease.^(1,2,5,7,8) However, does recurrent CPS lead to transformation to inverted papilloma or OSP? Previous reports have demonstrated the potential malignant transformation of OSP, but none have mentioned the possible transformation of CPS into OSP. In this case, there was no obvious transformation zone of CPS to OSP under thorough microscopic histological examination of the third and fourth surgical specimens, because the specimens obtained were too fragmented microscopically. Therefore, we cannot assume that the OSP of this patient arose from the inflammatory epithelium of CPS, or that recurrent CPS could predispose the Schneiderian epithelium to transform into OSP. However, there is no debate that the OSP in this case was found with inflammatory polyp in CPS. Since the prognosis and the clinical characteristics of OSP are very different from that of CPS, the differentiation of OSP from CPS has a vital impact on the surgical approach for the management of this rare sinonasal neoplasm. We have summarized the clinical characteristics of OSP and CPS in Table 2. Clinicians should be cautious when examining a patient with recurrent CPS, and pathologists must be meticulous in searching for malignancy or OSP on excised specimens from patients with recurrent CPS in order to avoid misdiagnoses and mistreatment. Furthermore, there is no definite proof that any systemic or local factors can contribute to the transformation of the OSP. A further series study to elucidate more evidence supporting systemic predisposition is necessary.

Yoon et al. reported that inflammatory polyps were associated with inverted papilloma in 21.9% of cases.⁽⁹⁾ However, this is the first case of OSP incidentally found in a patient with CPS reported in the English literature. We present this case to remind clinicians that OSP can be found with inflammatory polyps of chronic paranasal sinusitis, and it is easily overlooked clinically. Sufficient alertness from both the surgeon and pathologist is the cornerstone for the proper diagnosis and appropriate treatment of this rare sinonasal neoplasm.

REFERENCE

1. Maitra A, Baskin LB, Lee EL. Malignancies arising in oncocytic schneiderian papillomas. a report of 2 cases and

Table 2. Comparison of Clinical Characteristics of Oncocytic Schneiderian Papilloma with Chronic Paranasal Sinusitis

	Clinical symptoms	Gross findings	Sinus CT findings	Treatment	Prognosis
OSP	unilateral nasal obstruction, epistaxis, pain	a ragged, polypoid, red or pink intranasal mass, diffuse mucosal thickening with a finely granular surface	sinus opacification with intranasal soft tissue density, and erosion of the bony sinus wall with or without bony destruction	a lateral rhinotomy with en-bloc resection of the lateral nasal wall, Caldwell-Luc approach	high recurrence rate (25%~35%), local aggressiveness, malignant change potential
CPS	nasal obstruction, anosmia, colorless stringy or purulent secretions, postnasal catarrh, headache	with or without pale, grey smooth swelling of the nasal mucosa	sinus opacification, soft tissue mass filling the sinus	intranasal antrostomy endoscopic sinus surgery, Caldwell-Luc approach	variant recurrence, rate, no or low local aggressiveness, no malignant change potential

Abbreviations: CT: computed tomography; OSP: oncocytic schneiderian papilloma; CPS: chronic paranasal sinusitis

* Table 2 summarized from references 1, 2, 3, 4, 5, and 6

- review of the literature. *Arch Pathol Lab Med* 2001;125:1365-7.
- Yang YJ, Abraham JL. Undifferentiated carcinoma arising in oncocytic schneiderian (cylindrical cell) papilloma. *J Oral Maxillofac Surg* 1997;55:289-94.
 - Barnes L, Bedetti C. Oncocytic schneiderian papilloma: a reappraisal of cylindrical cell papilloma of the sinonasal tract. *Hum Pathol* 1984;15:344-51.
 - Hyams VJ. Papillomas of the nasal cavity and paranasal sinuses: a clinicopathological study of 315 cases. *Ann Otol Rhinol Laryngol* 1971;80:192-207.
 - Frantz TD, Rasgon BM, Rumore GJ. Pathologic quiz case 2. oncocytic schneiderian papilloma. *Arch Otolaryngol Head Neck Surg* 1994;120:102-3;106.
 - William L, Matthew RK, Hugh FB. Treatment outcomes in the management of inverted papilloma: an analysis of 160 cases. *Laryngoscope* 2003;113:1548-56.
 - Michaels L, Young M. Histogenesis of papillomas of the nose and paranasal sinuses. *Arch Pathol Lab Med* 1995;119:821-6.
 - Ward BE, Fechner RE, Mills SE. Carcinoma arising in oncocytic schneiderian papilloma. *Am J Surg Pathol* 1990;14:364-9.
 - Yoon JH, Kim CH, Choi EC. Treatment outcomes of primary and recurrent inverted papilloma: an analysis of 96 cases. *J Laryngol Otol* 2002;116:699-702.

發現於復發性慢性鼻竇炎中之腫瘤性司奈德氏乳突瘤

鄭宗岳 翁世樺¹ 陳盈霖 張凱評 陳聰明

一位 28 歲的男性病人，左側鼻塞 3 個月，合併有臭味，亦被發現左側鼻有鼻息肉，被診斷是慢性鼻竇炎。於民國 86 年在本院接受第一次功能性內視鏡鼻竇手術，之後因為鼻竇炎復發，又接受第二次鼻竇內視鏡手術，在第三次手術後之病理報告顯示為腫瘤性司奈德氏乳突瘤。術後門診追蹤又復發，病人又接受了第四次鼻竇內視鏡手術，術後的病理報告亦顯示為腫瘤性司奈德氏乳突瘤，門診追蹤兩年至今尚未復發。據我們所知，目前文獻上並無報告復發性慢性鼻竇炎與腫瘤性司奈德氏乳突瘤同時存在同一個病人身上，本病例是第一個報告的病例，也還沒有文獻報告有明顯證據指出復發性慢性鼻竇炎會使鼻黏膜有轉化為乳突瘤的可能性，日後繼續地研究探討此可能性是有必要的。我們希望能藉此一病例報告，提醒耳鼻喉科醫師及病理科醫師，應提高臨床上的警覺性。在慢性鼻竇炎病人鼻腔的鼻息肉內，可能會藏有此少見的腫瘤，而手術切除下來的慢性鼻竇炎組織，在顯微鏡下必須要更仔細的檢查，尤其是對復發性慢性鼻竇炎病患，以期能正確診斷出混合在鼻息肉中的腫瘤性司奈德氏乳突瘤。(長庚醫誌 2006;29:336-41)

關鍵字：腫瘤性司奈德氏乳突瘤，倒生性乳突瘤，復發性慢性鼻竇炎，內視鏡。

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