รายงานผู้ป่วย

Cylindrical cell papilloma: a case report

Churaiorn Masmahisak* Somboon Keelawat*
Supang Maneesri* Saowanee Yenrudi*

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A case of cylindrical cell papilloma arising in the right maxillary sinus is reported. The diagnosis was made by light microscopic, histochemical and confirmed by electron microscopic studies. Upon follow-up 8 months postoperatively, there was no evidence of recurrence. A review of the literature reveals that the tumor is quite uncommon and is occasionally misdiagnosed as rhinosporidiosis and other benign or malignant papillary epithelial proliferation. Association with malignancy has been recognized.

Key words : *Oncocytic Schneiderian papilloma, Cylindrical cell papilloma, Schneiderian papilloma.*

Reprint request : Masmahisak C, Department of Pathology, Faculty of Medicine, Chulalongkorn University, Bangkok 10330, Thailand.

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*Department of Pathology, Faculty of Medicine, Chulalongkorn University

จุไรอร มาศมหิศักดิ์, สมบูรณ์ คีลาวัฒน์, ศุภางค์ มณีศรี, เสาวณีย์ เย็นฤดี. Cylindrical cell papilloma: **รายงานผู้ป่วย 1 ราย. จุฬาลงกรณ์เวชสาร** 2543 ก.พ; 44(2): 119 - 25

รายงานผู้ป่วยหนึ่งรายที่มี cylindrical cell papilloma เกิดขึ้นที่โพรงไซนัส maxillary ข้างขวา การวินิจฉัยโรคกระทำโดยการใช้กล้องจุลทรรศน์ การย้อมพิเศษและยืนยันโดยกล้องจุลทรรศน์อิเลคตรอน จากการติดตามผลการรักษาที่โรงพยาบาลหลังจากได้รับการผ่าตัดเอาเนื้องอกออกได้แปดเดือน ไม่พบว่ามีเนื้องอกกลับมาเป็นซ้ำอีก การทบทวนวรรณกรรมทางการแพทย์พบว่าเนื้องอกชนิดนี้พบน้อย และมีบางครั้งที่ได้รับการวินิจฉัยผิดเป็น rhinosporidiosis หรือเนื้องอกของเซลล์บุผิวที่มีการเจริญแบบ papillary ทั้งชนิดที่เป็นเนื้องอกธรรมดาและที่เป็นมะเร็ง พบว่าเนื้องอกชนิดนี้อาจกลายเป็นมะเร็งได้

Papillomas of the sinonasal tract have been described in the literature with different names including squamous papilloma, septal papilloma, Schneiderian papilloma, etc. Perhaps the most acceptable classification has been a histomorphologic one that was formulated by Hyams.[1] He divided these tumors into inverted, fungiform, and cylindrical cell types, with the latter being the rarest and most controversial.[1,2]

Fungiform and inverted papillomas, whether pure lesions or histologically mixed, account for about 95% of all sinonasal tract papillomas and cylindrical cell papillomas constitute about 5%.[3] Cylindrical cell papilloma (CCP) has occasionally been misdiagnosed as papillary adenocarcinoma or rhinosporidiosis. Although benign, CCP may have a locally aggressive behavior and, on rare occasion, it may be associated with malignancy.[4,5] The recurrence rate is variable from series to series[1,6,7] and it is strongly related to inadequate removal.[1]

We report herein a patient who had a CCP arising in the right maxillary sinus.

Case report

A 45-year-old man came to the Ear, Nose, and Throat Service at Chulalongkorn Hospital with a chief complaint of a few months of right nasal stuffiness. On examination, he was found to have polypoid lesions in the right maxillary sinus and right nasal cavity.

The lesions were removed via a Caldwell-Luc approach and submitted for histologic examination.

The surgical specimen consisted of an irregular gray-white polypoid mass from right maxillary sinus, which measured 3x2x0.5 cm, and multiple fragments of edematous polypoid masses from the right nasal cavity measuring up to 2.5x1x1cm.

Microscopic examination showed papillary lesions composed of multilayers of tall columnar epithelial cells. The cells possessed eosinophilic granular cytoplasm, having both inverted and exophytic growth patterns which surrounded loose fibrovascular cores (Figure 1). Within the epithelium there were numerous small microcysts (Figure 2). Mitotic activity was negligible. These light microscopic findings were compatible with CCP.

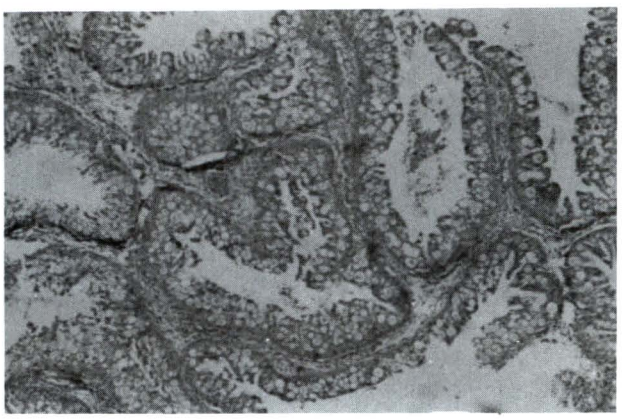

Figure 1. Low magnification of the papilloma showing papillary structures composed of epithelial lining which contains a lot of variable - sized cystic cavities and fibrovascular cores.(H&E)

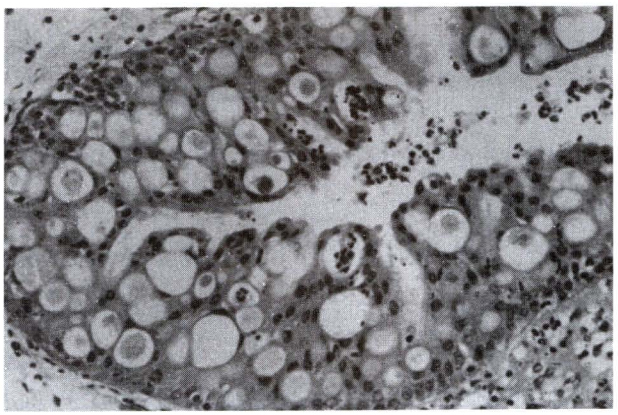

Figure 2. High magnification of the lining epithelium showing many cystic structures filled with mucin and neutrophils. (H&E)

The epithelial cells were positively stained with Phosphotungstic acid hematoxylin(PTAH). Electron microscopy of a portion of the paraffin-embedded tissue displayed intracytoplasmic lumina lined with microvilli (arrow, Figure 3A and B). Degenerated mitochondria were demonstrated (arrow, Figure 4).

The patient had no recurrence or malignant lesions when he came to the hospital for the last follow-up 8 months after the operation. He was subsequently last to follow-up.

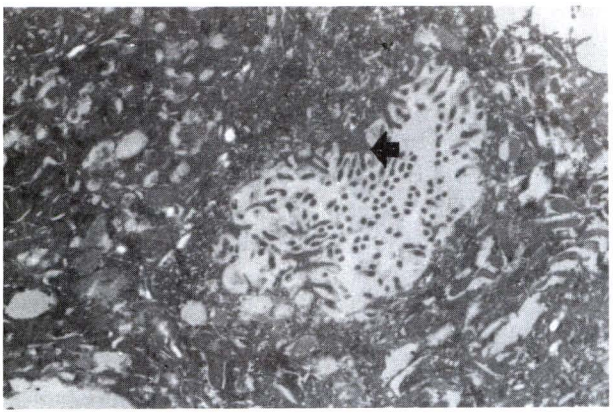

Figure 3A. Electron microscopy of cells in epithelium showing cystic structure lined with microvilli (E.M.,magnification x 13,600).

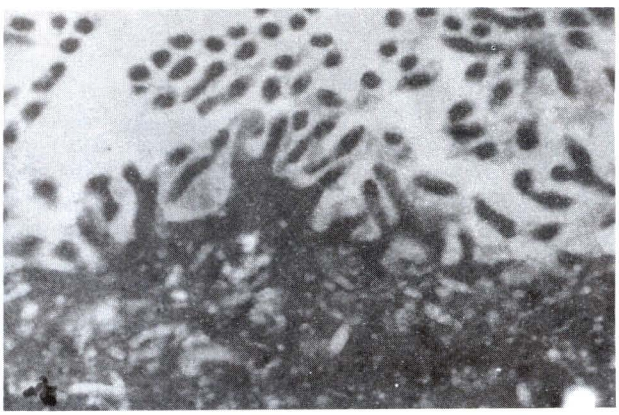

Figure 3B. High magnification of microvilli (E.M., magnification x 68,000).

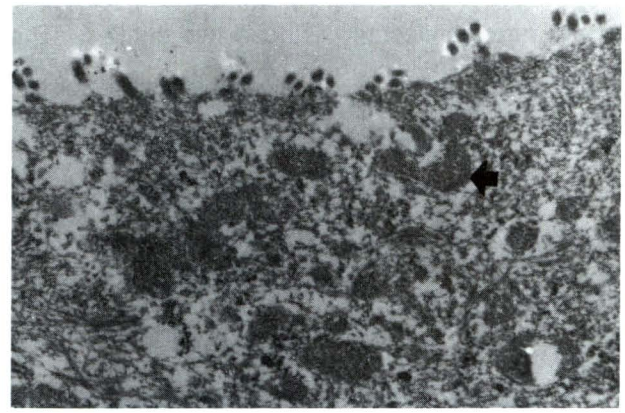

Figure 4. Electron microscopy of the cell - lining cyst showing microvilli and degenerated mitochondria (arrow) (E.M., Magnification x 25,500).

Discussion

The mucosa of the nasal cavity and paranasal sinuses originates from the ectodermal germ layer rather than from the endoderm as does the epithelium of the remaining respiratory tract. A common synonym for this ectodermally derived mucosa is the"Schneiderian membrane".[1] There are three morphologically distinct papillomas arising from the "Schneiderian mucosa" of the nose and paranasal sinuses whose names have been previously mentioned.[1,2] Fungiform and inverted papillomas, whether pure lesions or histologically mixed, comprise the majority of them while CCP is an unusual variant.[3] The respiratory mucosa of the endodermal origin in the remaining respiratory tract does give rise to papillomas but they generally correspond histologically to the typical exophytic squamous cell papilloma without the other forms of papillomas seen in lesions of the sinonasal area.[1]

Cylindrical cell papilloma is the rarest form of Schneiderian papilloma. [3] It has distinct light microscopic and ultrastructural morphological features. Microscopically, the classic features are identical to the current case. The epithelial cell layer typically has numerous small intraepithelial cystic structures filled with mucin and/or neutrophils.[7] Ultrastructurally, the cylindrical cell papilloma exhibits mitochondrial hyperplasia, mitochondrial hypertrophy, and intracytoplasmic lumina lined with microvilli and cilia.[2]

Barnes and Bedetti, [7] in their study, demonstrated that the epithelial cells of CCP were true oncocytes because they could find abundant mitochondria in the cytoplasms of these cells by using histochemical (Phosphotungstic acid hematoxylin and Luxol fast blue), immunocytochemical (Cytochrome c oxydase) and ultrastructural studies. These staining methods are staining for mitochondria. Therefore, they encouraged the term "Oncocytic Schneiderian papilloma".

Cylindrical cell papilloma appears in an age ranging from adolescence to the elderly, an average being around 50 years. No sex predilection is apparent.[8] Unilateral nasal obstruction is the most common presenting symptom. Intermittent epistaxis

and pain have also been described.[8] Rhinorrhea, sinusitis, and allergic symptoms are rare manifestations.[8] Duration of symptoms is usually months to years.[8] All cases have been unilateral.[8] CCP commonly arises in lateral nasal walls and sinuses but septal involvement has been recognized, although it is very rare.[9]

Classically, the tumors have gross shaggy or papillary configuration.[8] However, smooth, edematous, polypoid lesions are also common.[8] Color may vary among combination of red, brown, tan, pink, and gray.[8] Patient with CCP may have coexisting nasal polyps. This appears to be a coincidental finding, and is observed with fungiform and inverted papilloma as well.[8]

Sinus radiographs typically reveal abnormalities confined to the ipsilateral sinonasal passages. Sinus opacification associated with an intranasal soft tissue density are the predominant findings.[8] Bone destruction on plain films or tomography is suggestive of coexistent malignancy.[8]

Carcinoma has also been documented to arise from cylindrical cell papilloma. [4,5] It is most often of the squamous cell carcinoma, although mucoepidermoid carcinoma and adenocarcinoma may also occur.[4]

Since it is rare, and is unfamiliar to many pathologists, CCP is often confused with rhinosporidiosis and other benign or malignant papillary epithelial proliferation. Presence of mucus-containing microcysts in the epithelium may also be confused with the sporangia of rhinosporidiosis. In the latter, however, the sporangia are located in both the epithelium and the underlying stroma where they may evoke a conspicuous giant cell reaction.[4] Moreover, electron microscopy can easily distinguish between these two entities. The ultrastructural findings of rhinosporidiosis are distinctive. The structures formerly regarded as "sporangia" and "spores" are composed of fibrillar walls containing organized concentric lamellated bodies or amorphous electron dense materials depending on whether they are in their early or late stages.[10] Papillary carcinomas differ from CCP by the presence of nuclear pleomorphism and mitotic activity, a more complex pattern of proliferation, and invasion of the underlying stroma.[4]

Due to high rates of recurrence and/or persistence after inadequate surgical excision, and given the possibility of missing an associated carcinoma in an inadequate biopsy, a lateral rhinotomy with en bloc resection of the lateral nasal wall- followed by meticulous removal of all additional mucosa in the ipsilateral paranasal sinuses- is advocated for the management of CCP. The role of postoperative radiation and/or radical maxillectomy in patients in whom a carcinoma is discovered remains individualized at present, and depends upon the extent of disease in each patient.[8]

The causes of cylindrical cell papilloma and the other Schneiderian papillomas remains unknown. According to Hyams,[1] there is no convincing association with allergy, chronic infection, smoking, or other noxious environmental agents. Furthermore, no evidence of viral etiology has been discovered either by light microscopic, ultrastructural examination or tissue cultures.[7]

In the case reported here, age, presenting symptoms as well as microscopic , histochemical and ultrastructural findings were compatible with CCP. The patient was doing well on the last follow-up 8 months

postoperatively. That period, however, is too short because recurrence usually occurs approximately one year after the surgery.[1]

Summary

A case of a 45-year-old male with cylindrical cell papilloma of the right maxillary sinus was reported. The diagnosis was made by light and electron microscopy. At the follow - up, 8 months after removal of the lesions, the patient was still free from tumor recurrence.

Because of frequent confusion with other entities such as rhinosporidiosis or other benign and malignant papillary epithelial proliferation and the fact that cylindrical cell papilloma is associated with malignancy and recurrence , getting familiar with this unusual form of papilloma is crucial.

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