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Synchroniczny guz zatokowo-nosowy: podwójny kłopot?

Synchronous sinonasal tumour: double trouble?

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Streszczenie

Brodawczak Schneidera jest łagodnym guzem zatok przynosowych, który występuje w trzech wariantach morfologicznych: grzybiastym, odwróconym i onkocytarnym. Brodawczak odwrócony, będący najbardziej rozpowszechnionym wariantem, występuje trzykrotnie częściej u mężczyzn w 5.–7. dekadzie życia. Rak nosogardła jest nowotworem złośliwym wywodzącym się z nabłonka jamy nosowo-gardłowej. Rzadko występujący synchroniczny guz zatokowo-nosowy wciąż pozostaje problemem, zwłaszcza gdy dotyczy synchronicznego guza jamy nosowej o charakterze złośliwym i łagodnym. W pracy opisano przypadek skutecznie leczonego synchronicznego brodawczaka odwróconego zatok przynosowych współistniejącego z rakiem nosogardła u starszego mężczyzny. Autorzy zwracają uwagę na konieczność badania podejrzanych obustronnych guzów jamy nosowej ze względu na możliwość współistnienia dwóch różnych patologii, tak jak w opisywanym przypadku.

Słowa kluczowe: brodawczak odwrócony zatok przynosowych, rak nosogardła, nowotwór jamy nosowej

Abstract

Schneiderian papilloma is a benign sinonasal tumour, which has three morphological variants: fungiform, inverted and oncocytic. Inverted papilloma, being the most prevalent variant, is three times more common among males in their fifth to seventh decade of life. On the other hand, nasopharyngeal carcinoma is a malignant tumour arising from the nasopharyngeal epithelium. Albeit rare, synchronous sinonasal tumour has been reported and remains a quandary till date, especially when it involves malignant and benign synchronous nasal mass. In this paper, we reported a case of a successfully treated synchronous sinonasal inverted papilloma with nasopharyngeal carcinoma in an elderly male. We would like to highlight the need to investigate suspicious bilateral nasal masses due to the possible coexistence of two different pathologies, as in our case.

Keywords: sinonasal inverted papilloma, nasopharyngeal carcinoma, nasal tumour

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INTRODUCTION

Sinonasal inverted papilloma (IP), first described by Ward in 1854, is a type of benign Schneiderian papilloma, which derives from the Schneiderian membrane[1]. Schneiderian papilloma has 3 morphological variants, namely fungiform, inverted and oncocytic, with IP being the most common and presenting with unilateral nasal mass[1,2]. Despite possible associations with chronic inflammatory conditions, human papilloma virus infection and nicotine have been addressed as the main culprits[1]. IP constitutes 0.5–4% of all sinonasal masses with a tendency to malignant transformation[3]. On the contrary, co-occurrence of synchronous IP and other nasal mass has scarcely been reported in English literature more so when it is a malignant nasal tumour, such as nasopharyngeal carcinoma (NPC). NPC is a malignant tumour arising from the nasopharyngeal epithelium, with Epstein–Barr virus being the cardinal pathogenic factor. Synchronous carcinoma occurs in two-thirds of cases of IP, whereas some patients develop metachronous carcinoma after resection of IP. Squamous cell carcinoma (SCC) is the most common associated malignancy, predominantly among older-aged male population[3]. To our best knowledge, this is the first case report on synchronous IP and NPC.

CASE REPORT

A previously healthy 65-year-old male presented with a 2-month history of tinnitus and reduced hearing in addition to recurrent nasal symptoms: non-progressive nasal obstruction, rhinorrhoea and sneezing. The patient denied any epistaxis, anosmia, constitutional or B symptoms. There was also no neck swelling, dysphagia, or odynophagia. He had no family history of malignancy or history of childhood radiation.

Upon examination, he was comfortable and not in respiratory distress. There was no external nose deformity. Cold spatula test showed reduced misting over the right side. Anterior rhinoscopy revealed a pale mass over the left nasal cavity, which was not seen over the right side. Rigid nasoendoscopy revealed a polypoidal mass occupying the right nasal cavity, which bled on touch and was friable (Fig. 1 A), and there was a fungating mass occupying the left fossa of Rosenmüller (Fig. 1 B). Otoscopy revealed dull-looking tympanic membrane on the left with normal findings on the right. Besides that, intraoral, neck and other systemic examinations were unremarkable. All cranial nerves were intact. His vital signs were within a normal range. We decided to take biopsy from both nasal masses. To our surprise, the biopsy of the right nasal mass revealed endophytic papillomatous lesion composed of proliferating columnar epithelium which grew inversely into the underlying basement membrane (Fig. 2), suggesting an inverted papilloma, whereas the left nasal mass biopsy revealed non-keratinising stratified squamous epithelium suggestive of nasopharyngeal carcinoma (Fig. 3).

Computed tomography from brain to abdomen demonstrated enhancing soft tissue density occupying the right middle meatus and obliterating the right osteomeatal complex, measuring approximately 1.8 × 1.1 × 1.5 cm (AP × W × CC), while soft tissue density was noted over the left fossa of Rosenmüller measuring approximately 3.3 × 2.4 cm involving into the nasopharynx and left parapharyngeal space, prevertebral muscles and left pterygoid muscle with no evidence of cervical or distant metastasis. The patient was diagnosed as NPC T2N0M0 (stage II) with sinonasal inverted papilloma.

The patient successfully underwent concurrent chemoradiotherapy at the oncology unit and was planned for endoscopic right medial maxillectomy.

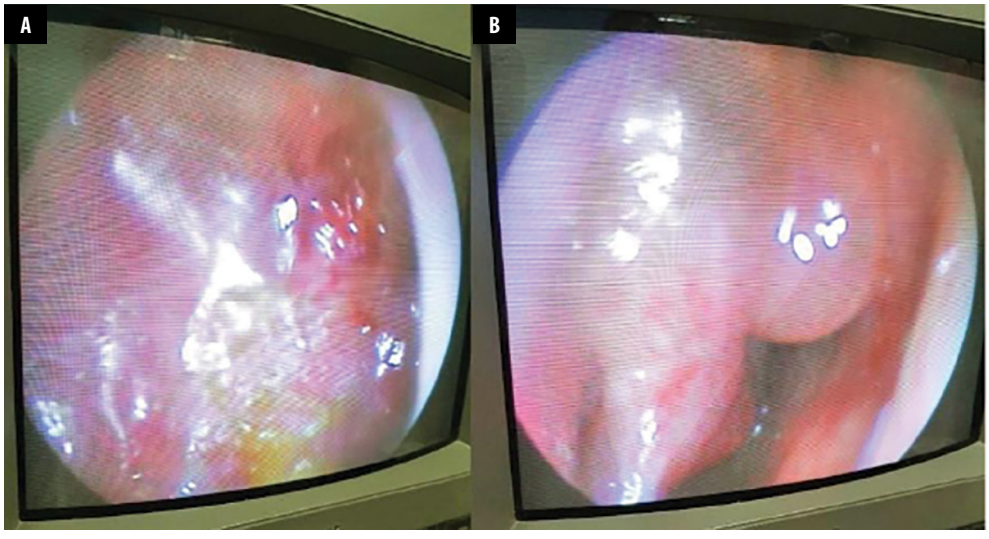

Fig. 1. **A.** *Left side fossa of Rosenmüller region post CCRT.* **B.** *Right pink and grey nasal cavity mass*

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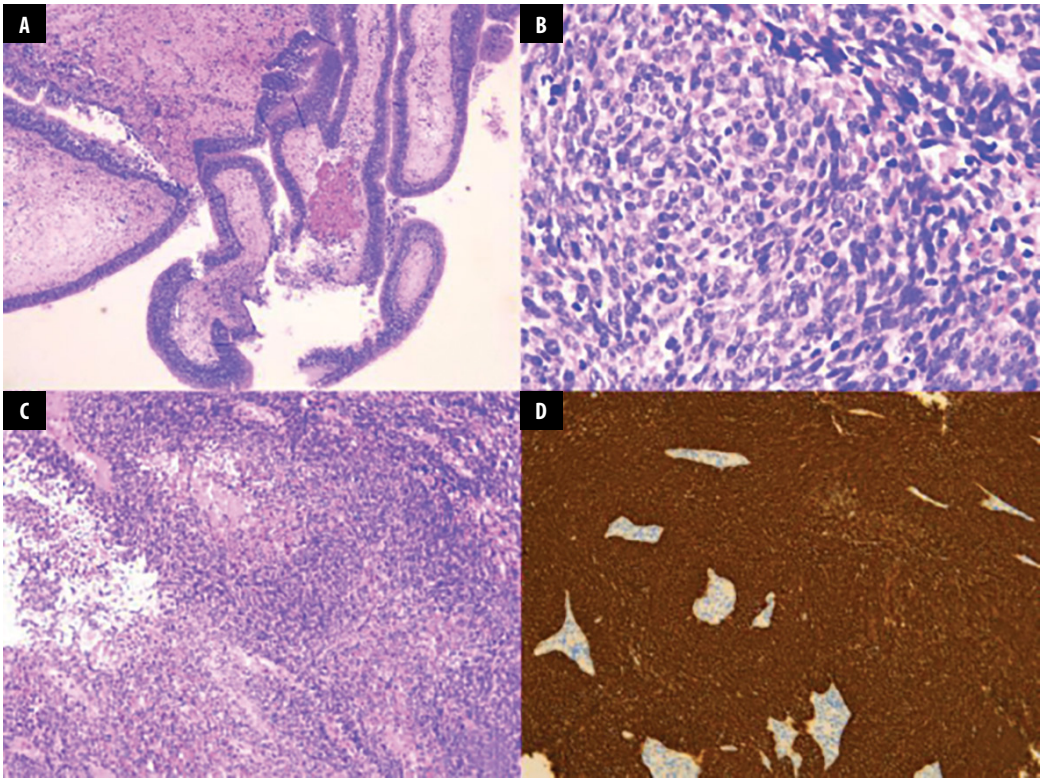

Fig. 2. Histopathology images. **A.** *Endophytic papillomatous lesion composed of proliferating columnar epithelium.* **B, C.** *Tumour composed of papillary fronds comprising delicate stromal core covered by stratified large tumour cells displaying indistinct cell border, poorly differentiated, vesicular nuclei with some mitosis. There is an enlarged nucleus: cytoplasmic ratio with scanty cytoplasm. No keratin pearls seen.* **D.** *Positive pan-cytokeratin staining indicating squamous cell carcinoma*

DISCUSSION

IP comprises 0.4–4.7% of all sinonasal tumours with male predominance (3.4:1) reported between 20 and 50 years of age. On the other hand, NPC remains the most common head and neck carcinoma originating from the nasopharyngeal epithelium. NPC accounts for 0.5–2.0 cases per 100,000 in the United States of America and Europe while higher prevalence of up to 25 per 100,000 is seen in endemic countries, such as southern China and Southeast Asia. In Malaysia, NPC is the fourth leading overall malignancy and the third most common malignancy among

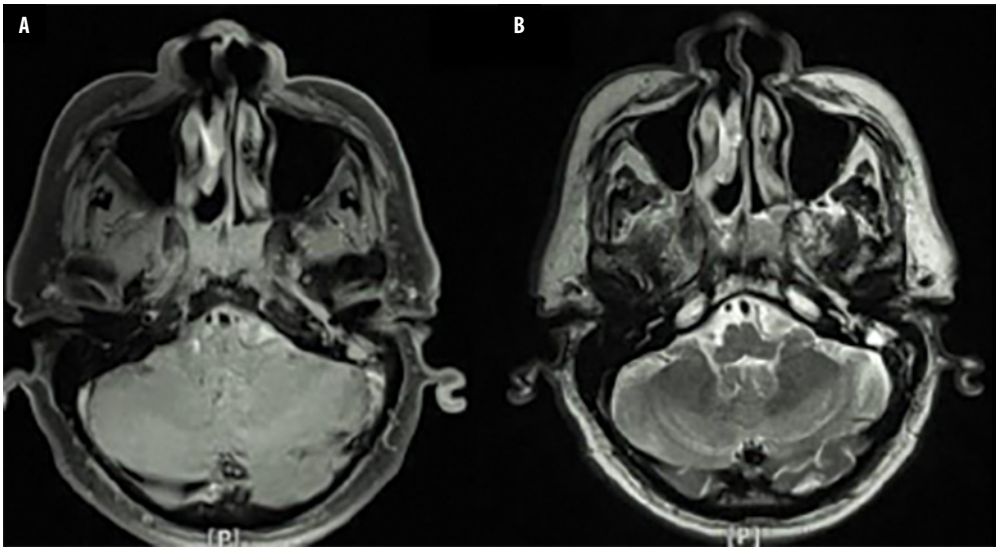

*Fig. 3. MRI T1 (***A***) iso-intense, and T2 (***B***) hyper-intense sections. There is heterogenous enhancement and thickening of the left fossa of Rosenmüller obliterating the left torus tuberous and soft tissue density at right middle meatus obliterating the right osteomeatal complex*

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male patients. As for ethnic distribution, NPC is more prevalent among the Chinese followed by Malay people and the least common among Indians. In our case, the patient was a Malay male in the 7th decade of life.

A patient with IP and NPC commonly presents with similar symptoms like unilateral nasal obstruction, nasal discharge, epistaxis, facial pain, and pressure[4,5]. Occupancy of the nasopharynx contributes to symptoms related to eustachian tube dysfunction, for example tinnitus, aural fullness, and reduced hearing. As the tumour progresses, patients may develop symptoms like headache and cranial nerve involvement in more advanced stages[6]. In this case, the patient initially presented with unilateral tinnitus and reduced hearing with vague non-alarming nasal symptoms. It is also noteworthy that NPC predominantly presents with neck swelling, which was also not present in our patient.

Nasal mass generally requires meticulous history taking, thorough physical examination along with rigid nasoendoscopy for early diagnosis and successful management. Rigid nasoendoscopy will unveil the characteristics of nasal mass. These include: the presence of translucent, polypoidal mass which does not bleed on touch represents nasal polyp, the presence of pink, grey or tan polypoidal growth with a convoluted, wrinkled surface or mulberry appearance is typical of IP[4]; ulcerated or fungating mass is suggestive of malignant tumours. IP arises mostly from the lateral nasal wall and subsequently extends into the maxillary, ethmoidal sinus, followed by sphenoid and frontal sinuses as the tumour progresses[4]. As for NPC, early lesion may appear to be subtle mass, bulge or asymmetry of fossa of Rosenmüller. Having said that, the occurrence of submucosal lesion in NPC is not uncommon.

Biopsy of the nasal mass is the gold standard in diagnosing this entity. The sinonasal tract is lined by ectodermally derived ciliated respiratory mucosa also known as the Schneiderian membrane, transitional cell papilloma and Ringerts cell papilloma. It gives rise to 3 histopathological subtypes, namely IP, exophytic (fungiform, septal and squamous papilloma) and oncocytic papilloma (cylindrical cell and columnar papilloma, 3–5%)[7,8]. NPC can be divided into 3 histological subtypes: keratinising squamous cell carcinoma, non-keratinising carcinoma, which is further divided into differentiated and undifferentiated subtypes, and basaloid squamous cell carcinoma. This subtyping in NPC enables us to determine the prognosis of the disease[9]. The undifferentiated type of NPC, although more aggressive, has been reported to exhibit better prognosis due to its higher radiosensitivity[6]. In the presented case, our patient was diagnosed with right sinonasal IP and left non-keratinising NPC.

Complete surgical resection is the definite treatment for IP. It should be borne in mind that surgery ought to be done promptly due to its aggressive nature, high recurrence rate as well as the possibility of malignant transformation. Hence, a long-term follow-up is often recommended[10]. Surgical approach depends on the extent of the tumour. On the contrary, NPC is a radio- and chemosensitive tumour. Most studies have reported that concurrent chemoradiotherapy (CCRT) has better outcome by improving the overall survival rate, which is about 62% with CCRT as compared to 56% with radiotherapy alone[6]. Our patient successfully underwent CCRT (35 Fr of 70 Gy radiotherapy at faciocervical region and weekly carboplatin AUC 2). The patient is planned for endoscopic medial maxillectomy at 3 months following CCRT.

CONCLUSIONS

This case highlights the first presentation of a synchronous sinonasal IP and NPC. Thus, clinicians should be alert of synchronous lesions and meticulous sampling is needed to provide adequate treatment to prevent recurrence and improve the survival rate.

Conflict of interest

All authors have no conflict of interest.

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