© Pediatr Med Rodz 2023, 19 (1), p. 55–59
DOI: 10.15557/PiMR.2023.0009

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Otrzymano: 03.08.2022
Zaakceptowano: 14.11.2022
Opublikowano: 30.06.2023

Brodawczak odwrócony o obrazie przypominającym naczyniakowłókniaka młodzieńczego nosogardła u młodego mężczyzny

Inverted papilloma masquerading as juvenile nasopharyngeal angiofibroma in a young adolescent male

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Streszczenie

Brodawczak odwrócony to łagodny nowotwór wywodzący się z nabłonka błony śluzowej zatok przynosowych. Występuje głównie w piątej i szóstej dekadzie życia, częściej u mężczyzn. Częstość występowania nowotworu w populacji młodych osób dorosłych jest odpowiednio niższa. Obraz kliniczny i radiologiczny schorzenia może naśladować inne łagodne typy nowotworów rozwijających się w obrębie zatok przynosowych, zatem ostateczne rozpoznanie wymaga badania histopatologicznego. W związku z powyższymi uwarunkowaniami niezbędne jest precyzyjne planowanie postępowania diagnostycznego i terapeutycznego. W pracy przedstawiono przypadek guza umiejscowionego jednostronnie w obrębie zatok przynosowych u młodego mężczyzny, u którego stwierdzono kliniczne i radiologiczne cechy naczyniakowłókniaka młodzieńczego nosogardła, m.in. poszerzenie otworu klinowo-podniebiennego, tylny rozrost guza w kierunku nosogardzieli oraz obecność pustych przestrzeni sugerującą jego wysoce unaczyniony charakter. Pomimo tych charakterystycznych cech na podstawie pooperacyjnego badania histopatologicznego rozpoznano brodawczaka odwróconego.

Słowa kluczowe: brodawczak odwrócony, naczyniakowłókniak, młode osoby dorosłe, nowotwór zatok przynosowych

Abstract

Inverted papilloma is a benign sinonasal epithelial tumour that mainly occurs in the fifth to sixth decades of life, with male predominance. The incidence in the young adolescent population is comparatively lower. The clinical and radiological findings may mimic other benign sinonasal tumours, so the diagnosis can only be confirmed by histopathological examination. Consequently, precise diagnostic and therapeutic planning is recommended to achieve the optimal result. We describe a case of unilateral sinonasal tumour in a young adolescent male who showed clinical and radiological features of juvenile nasopharyngeal angiofibroma, such as widening of the sphenopalatine foramen, extension of the mass posteriorly towards the nasopharynx, and presence of flow voids that suggested highly vascular nature of the mass. However, a postoperative histopathological examination showed a diagnosis of inverted papilloma.

Keywords: inverted papilloma, angiofibroma, adolescent, sinonasal neoplasm

INTRODUCTION

A sinonasal tumour is a growth that arises from the nasal cavity or paranasal sinuses. Sinonasal tumours may occur among all age groups, but they are mainly present in the fourth decade of life. For patients who are within the age group of 11 to 20 years, more commonly associated pathologies include inflammatory lesions and benign neoplasms, as compared to malignancy. A considerable number of differential diagnoses are available, such as a nasal polyp, Schneiderian papilloma, haemangioma, and juvenile nasopharyngeal angiofibroma (JNA)[1]. In most situations, the diagnosis can be determined by obtaining a tumour biopsy in the clinic setting. However, not every case can be subjected to a similar procedure. For instance, in cases of JNA, tumour biopsy is largely avoided in view of the risk of severe haemorrhage. In such patients, the diagnosis is based solely on clinical and radiological findings[2].

CASE REPORT

A 19-year-old male patient presented with a persistent right-sided nasal obstruction which progressively worsened for a one-year duration. It was associated with blood-stained nasal discharge and hyposmia. There was no history of facial swelling or numbness, tinnitus or reduced hearing, double vision or blurring of vision, headaches or vomiting. He did report any loss of weight, loss of appetite or neck swelling. Upon examination, a firm, fleshy and pinkish mass was found occupying the entire right nasal cavity. Through the left nasal cavity, the mass was seen extending to the nasopharynx. The nasal bridge was broadened, with obliteration of the right nasomaxillary fold (Fig. 1). Oral cavity examination showed no signs of trismus or intraoral mass. There were no palpable cervical nodes. The cranial nerves, otoscopic and ophthalmic examinations were within normal limits.

Contrast-enhanced computed tomography (CECT) of the paranasal sinuses revealed an enhancing lesion in the right nasal cavity, and in the right frontal and ethmoid sinuses, extending posteriorly to the nasopharynx and inferiorly to the oropharynx, displacing the soft palate. The right sphenopalatine foramen appeared to be widened as compared to the contralateral side (Fig. 2). On magnetic resonance imaging (MRI) of the brain and paranasal sinuses, the lesion showed intermediate signal on T1, intermediate to high signal on T2, and heterogeneous enhancement post-gadolinium injection. Flow voids were noted especially at the oropharyngeal part (Fig. 3). There was no orbital or intracranial extension. The tumour also demonstrated a convoluted cerebriform pattern. In view of the CT scan showing widening of sphenopalatine foramen, which is a finding typically associated with JNA, a provisional diagnosis of JNA was made.

The patient was scheduled for preoperative embolisation prior to definitive surgical resection. Angiography showed tumour blush with a supply from the right internal maxillary artery. After performing embolisation of the right internal maxillary artery, no more tumour blush was seen (Fig. 4). Subsequently, the patient underwent transnasal endoscopic excision of the tumour. Intraoperatively, the mass was found arising from the right anterior and posterior ethmoid, with the involvement of the right superior and middle turbinates. The tumour was dissected along the pseudocapsule, as recommended in JNA treatment (Fig. 5).

Unexpectedly, the histopathological examination (HPE) of the tumour later returned the diagnosis of inverted papilloma. A prominent papillomatous configuration with evidence of endophytic growth was noticed in the specimen

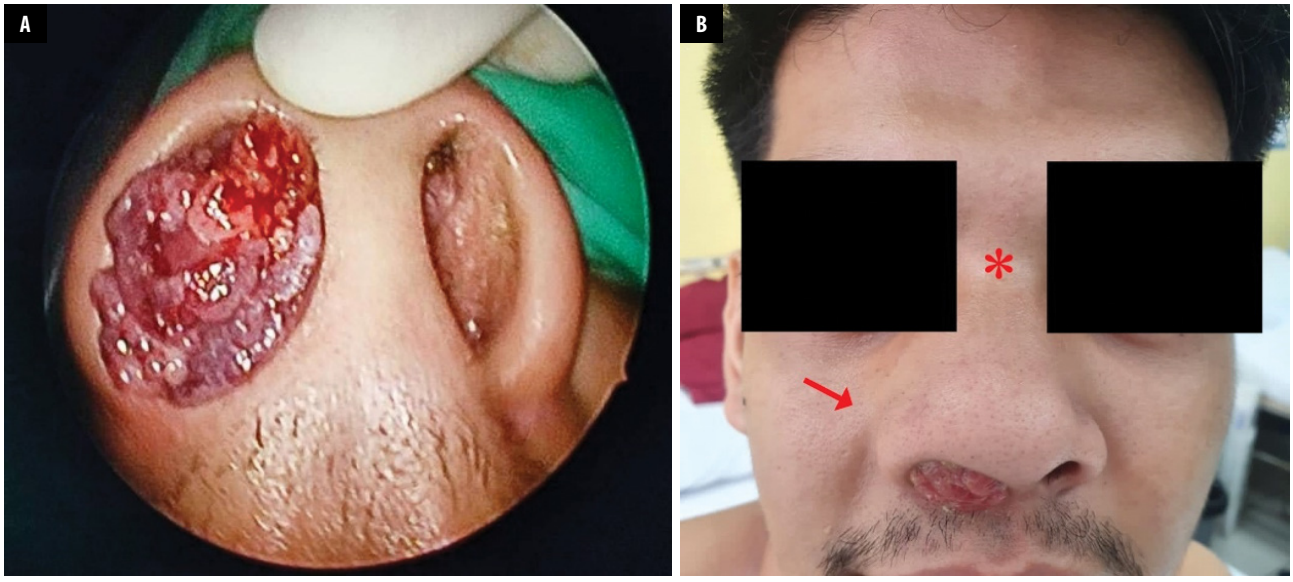

*Fig. 1. Mass filling up the right nasal cavity (**A**), with typical facial features (asterisk: broadening of nasal bridge, arrow: obliteration of right nasomaxillary fold) (**B**)*

DOI: 10.15557/PiMR.2023.0009

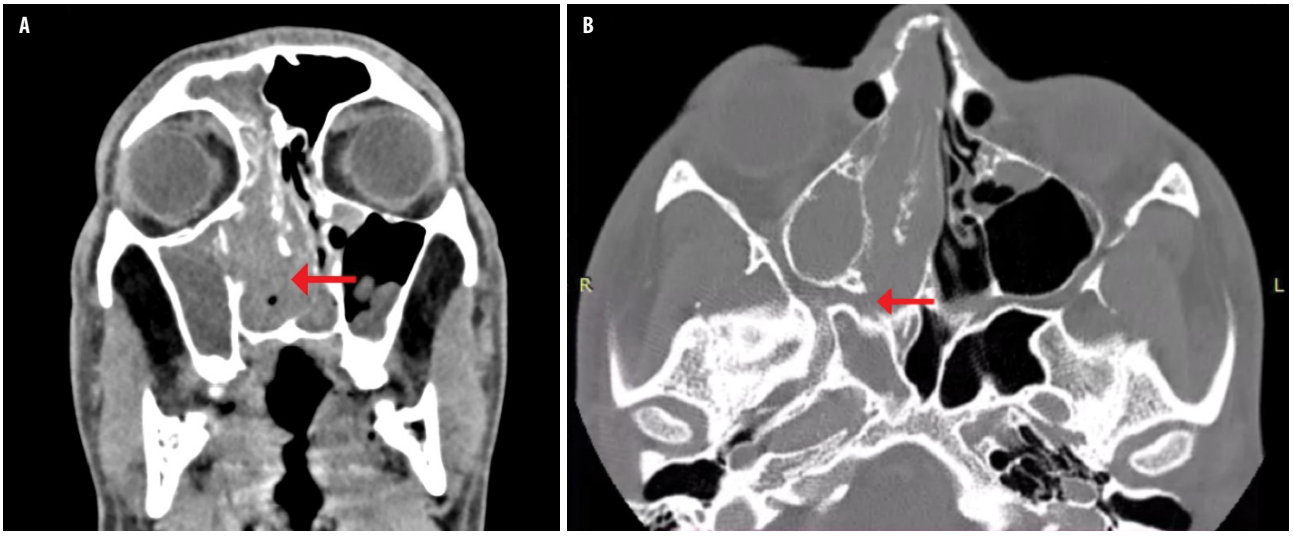

*Fig. 2. CT scan demonstrated the mass at right nasal cavity, frontal and ethmoid sinuses (**A**), with widening of right sphenopalatine foramen (**B**)*

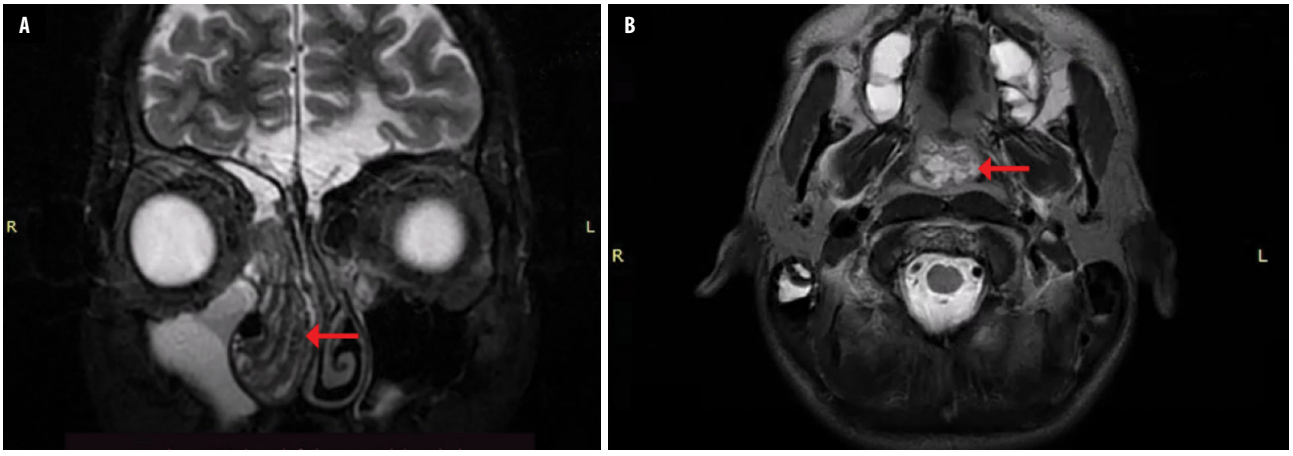

*Fig. 3. MRI scan findings were consistent with an intermediate-to-high signal with convoluted cerebriform pattern on T2-weighted image (**A**) with the presence of flow voids (**B**)*

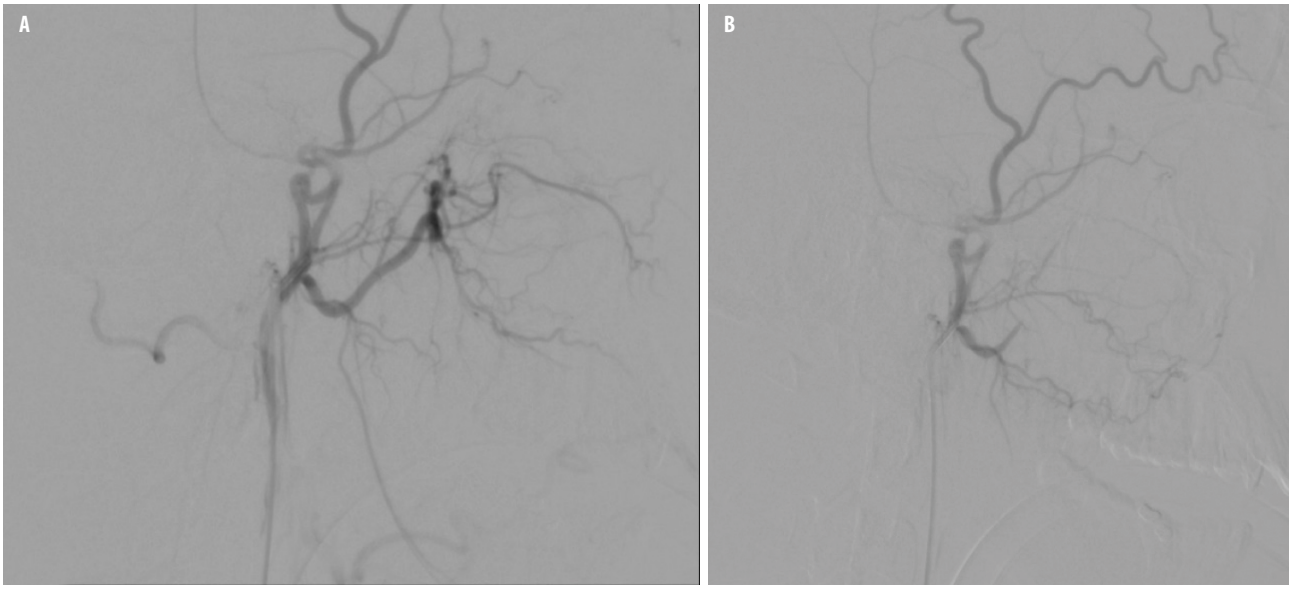

*Fig. 4. Angiography findings showing the tumour blush pre-embolisation (**A**) and significantly reduced post-embolisation (**B**)*

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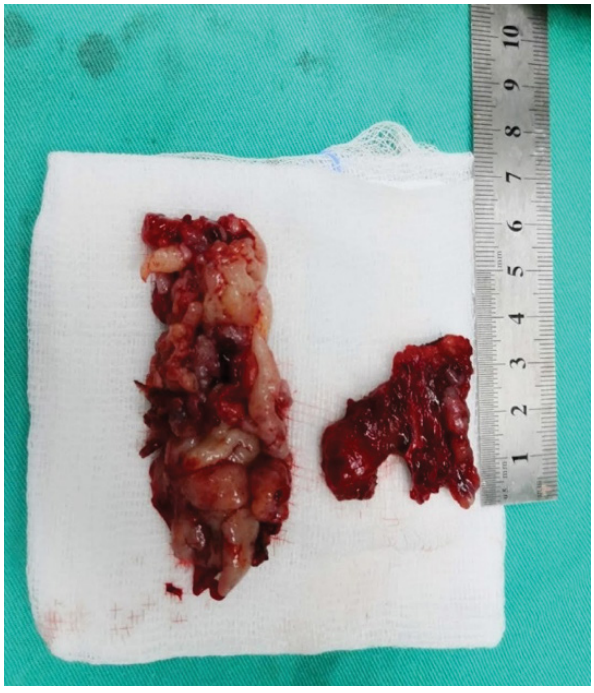

Fig. 5. Tumour after resection – mixed pale yellowish and fleshy polypoidal mass

(Fig. 6). Four months later, the patient was found to have tumour recurrence and was scheduled for a revision surgery.

DISCUSSION

Inverted papilloma (IP) is a benign sinonasal tumour that affects predominantly adults during the fifth to sixth decades of life. Although it is classified as a benign tumour, it is closely associated with the risk of recurrence and malignant transformation. The symptoms are non-specific and include nasal obstruction, rhinorrhoea, epistaxis, hyposmia or anosmia, and headache. The tumour appears on endoscopy as a reddish-grey lobulated mass, firm in nature, with a characteristic raspberry appearance. It is usually friable, and bleeds on contact[3]. Most IPs originates from the lateral nasal wall, in the middle meatus, and secondarily involves the maxillary and ethmoid sinuses[4].

JNA, on the other hand, is a benign vascular tumour that occurs almost exclusively in young male patients between the ages of 9 to 19. Symptoms include nasal obstruction, rhinorrhoea, and recurrent epistaxis. The tumour usually presents as a fleshy mass posterior to the middle turbinate. It is considered to arise either from the sphenopalatine foramen or pterygopalatine fossa, with the tendency to spread medially to the nasopharynx as well as towards the natural foramina, which offers the least resistance[5]. The manifestation of our patient generated a suspicion towards JNA, as he was an adolescent and reported with the typical complaint of unilateral epistaxis.

It is difficult to establish the diagnosis by just judging on the appearance of the mass because both IP and JNA may present as a fleshy mass with contact bleeding, similarly to other haemangiomatous lesions. The huge mass presentation in this case also makes the assessment of the origin problematic. Nevertheless, the nature of the mass, extending towards the nasopharynx and eventually to the contralateral side, is more consistent with JNA.

Imaging examinations, such as CT and MRI scans, are necessary to further evaluate the sinonasal tumour. On CT scans, IP shows as an isodense homogeneous lesion generally centred on the middle meatus. Bony erosions are common, and intralesional calcifications are present in 20% of cases. There may be an area of focal hyperostosis, which is highly suggestive of the tumour implantation site. MRI scan shows low signal on T1-weighted images and iso- or hypointense on T2-weighted images. The convoluted cerebriform pattern of the tumour strongly orientates the diagnosis[3].

In contrast, JNA appears on CT scans as a heterogeneous mass centred within the sphenopalatine foramen and enhancing after contrast administration. Typical radiological features include anterior bowing of the posterior maxillary wall (Holman–Miller sign) and widening of the sphenopalatine foramen. On MRI scans, JNA shows low signal intensity on T1-weighted images and intermediate signal on

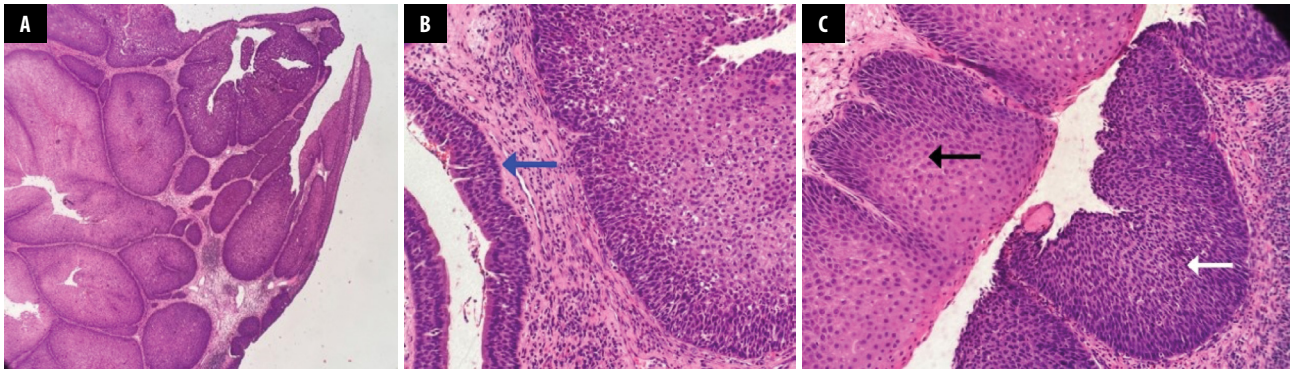

Fig. 6. HPE findings. **A.** *Prominent endophytic growth of proliferating epithelial cells which invaginate the underlying stroma (HPE ×40).* **B, C.** *Papillomatous lesion lined by respiratory epithelium (blue arrow), transitional epithelium (white arrow), and stratified squamous epithelium (black arrow) (HPE ×200)*

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T2-weighted images. Prominent flow voids are observed due to vascularity[6]. The tumour in our patient showed features of high vascularity, as it was associated with flow voids and enhancement post-contrast administration.

The reason why there was widening of the sphenopalatine foramen in our reported case is not known, and can probably be attributed to anatomical variation, as no enhancing mass was found within. A convoluted cerebriform pattern is considered to be a reliable diagnostic feature of IP, but it does not always guarantee the correct diagnosis[7]. Furthermore, IP generally appears to be avascular when angiography is performed, but in our case it was otherwise[8]. The combined findings led us to the diagnosis of JNA, even though the patient did not have the pathognomonic Holman–Miller sign.

We faced a dilemma in establishing the correct diagnosis in this case in view of the atypical presentation and unavailability of tumour biopsy. A similar experience was reported by Jayakody et al. They provisionally diagnosed an 11-year-old boy with JNA based on his young age, imaging finding of a mass extending posteriorly to the nasopharynx, and the presence of irregularity along the posterolateral wall of the maxillary sinus. However, intraoperative frozen section revealed papillomatous and squamous features suggestive of IP[9]. It is important to differentiate between IP and JNA, as these are two separate entities requiring different surgical approaches. IP is well known for its recurrence, so proper resection must be planned. The bone at the base of the tumour should be managed by drilling, removal or cauterisation in order to reduce the risk of recurrence[10].

CONCLUSION

Despite being uncommon, IP should be considered a close differential diagnosis of a unilateral sinonasal mass in the population of young adolescents. Failure to detect the disease may lead to inadequate treatment and risk of recurrence. When in doubt or dilemma, as in our case, with the patient reporting with an atypical presentation, an intraoperative frozen section is a wise option to achieve the diagnosis prior to the surgical excision. Changes in the consent taking should be considered to include the treatment of IP such as medial maxillectomy, but ultimately the type of surgery would depend on the site and extent of the tumour, which should be assessed on a case-by-case basis.

Remark

This case was submitted as an E-poster presentation during the 14th Malaysian International ORL-HNS Congress 2022.

Conflict of interest

The authors do not report any financial or personal connections with other persons or organisations that could adversely affect the publication's content and claim the right to this publication.

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