



## CASE REPORT

# Unusual presentation and histopathology of bilateral nasal polypi; cavernous hemangioma (right) and inverted papilloma (left)

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## Abstract

Hemangiomas are common lesions in medical practice, but those arising in the nasal cavity and/or paranasal sinuses are rare. Inverted papilloma is a rare benign tumor with a high incidence rate in both the nasal cavity and sinuses. The presence of both lesions in the same patient is even rarer. Here we present a case of a male patient with an unusual presentation and dual pathologies of cavernous hemangioma and inverted papilloma of the sinonasal tract that underwent endoscopic surgery and showed no recurrence of disease after a 2-year follow-up. The association of nasal cavernous hemangioma at one side and inverted papilloma at the other side is a very rare occasion that requires further studies and histopathology is the only diagnostic tool.

## KEYWORDS

hemangioma, histopathology, inverted papilloma, nasal polyp

## 1 | INTRODUCTION

Nasal polyps are usually presented by nasal obstruction, which is a very common symptom and is caused by a wide range of etiologies. Inflammatory polyps are the most common type of nasal mass, and neoplastic lesions are rare.

Intranasal tumors may be seen as a typical proliferative lesion with tissue destruction or may mimic an inflammatory type in the early stages, showing polyps on endoscopy.<sup>1,2</sup> Some of these polyps are rare and unusual and require special attention in diagnostic and therapeutic management.<sup>3</sup>

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Hemangioma is the most common vascular lesion in the head and neck region and is usually seen on the scalp, face, orbit, or oral cavity. Involvement of the nasal cavity and paranasal sinuses is a very rare presentation. However, the majority of the sinonasal hemangiomas are of the cavernous type; they are usually seen in childhood in the septum and vestibule, and a few are encountered in adults with a female preponderance.<sup>4</sup> Here we report a large cavernous hemangioma in the nasal cavity of a male adult patient.

The left-side polyp in our patient was diagnosed as an inverted papilloma. Sinonasal inverted papillomas (SIP) are benign tumors of the nasal cavity and paranasal sinuses, representing approximately 0.5%–4% of all primary nasal tumors, and are found more frequently in males in the fifth and sixth decades of life.<sup>5</sup> Due to the rarity of both diseases and the extremely rare occurrence of both in the same patient at the same time, as well as limited information on the etiology, pathology, and treatment outcomes, There is limited information in the literature regarding the etiology, pathology, and treatment outcomes due to the rarity of both diseases and the extremely rare occurrence of both in the same patient at the same time. To the best of our knowledge, it is the first case in Saudi Arabia to describe this association of bilateral nasal polypi.

## 2 | CASE PRESENTATION

A 35-year-old man presented with a history of progressive right-sided nasal obstruction for about 1 year with associated mild nasal discharge that was occasionally blood-stained. There were no eye or ear symptoms. On clinical examination, he was conscious, oriented, alert, and hemodynamically stable.

On nasal endoscopy, there was a large polypoidal mass arising from the right inferior turbinate, involving about one-third of its posterior part. It was irregular and granular in appearance and easily bled on touch. The right middle meatus was clear, the left middle meatus was obliterated with mucosal swelling, and a small polypoidal mass was also seen. The latter was firm and granular, unlike the usual inflammatory polyps. The rest of the ENT examinations were unremarkable.

The patient's laboratory investigations, including hematology and chemistry, were within normal limits.

A CT scan was requested and showed a polypoidal mass at the posterior part of the right inferior turbinate, measuring 4.5 × 2.5 cm, extending into the right nasopharynx, while the left side showed obstruction of the osteomeatal complex with polypoidal opacity and complete heterogeneous opacity of the left maxillary sinus. No bone erosion

was noted. The rest of the paranasal sinuses were found to be clear. On MRI, the right inferior turbinate mass was hyperintense on T2W images without any erosion of the surrounding soft tissues (Figure 1).

Endoscopic sinus surgery was performed, and polypoidal masses were completely removed, with more bleeding on the right side. The excised lesions were sent for histopathology examination. The postoperative course was uneventful, and the patient was discharged on the second postoperative day (Figure 2).

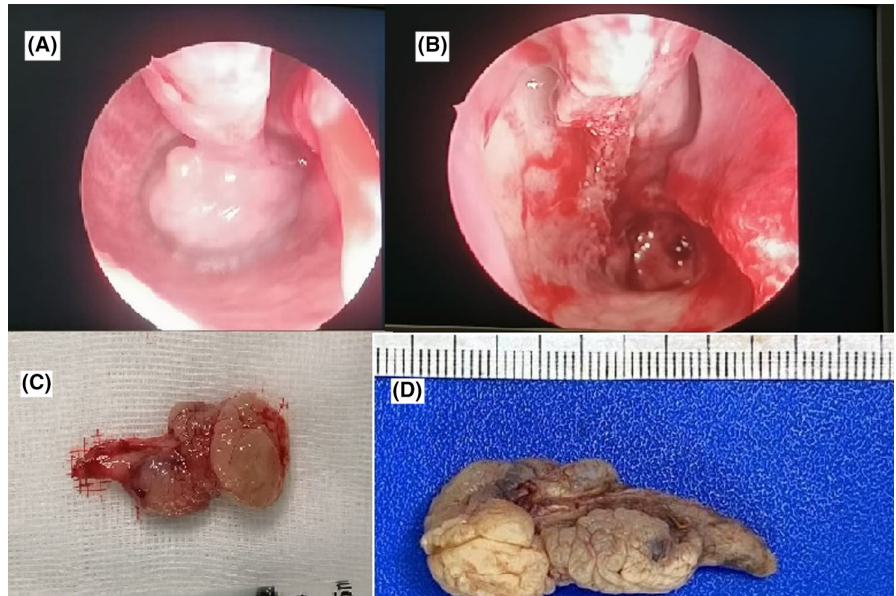
On gross examination, a right polypoidal mass measuring 4.3 × 2.2 × 2 cm with a soft to rubbery and grayish pink cut section was seen, and a left side biopsy was received as a fragmented grayish-white tissue piece measuring 2.3 × 2 cm.

Microscopic examination revealed a right-side polyp with respiratory epithelial covering and underlying tissue formed of loose connective tissue with proliferated and dilated blood vessels lined by bland-looking endothelial cells with no communication or atypia (Figure 3). Left-sided slide examination showed hyperplastic (5–10 layers) epithelium of respiratory and transitional types exhibiting downward endophytic growth and round nests with smooth outer contours and occasional mitotic figures (Figure 4). The final diagnosis was issued as right-side cavernous hemangioma and left-side inverted papilloma.

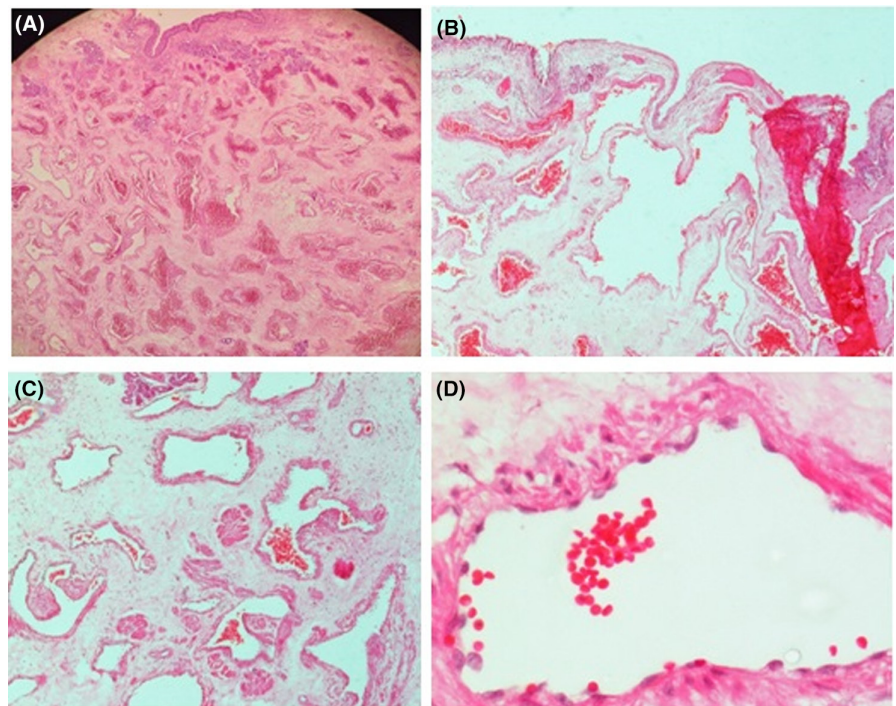


FIGURE 1 Preoperative T2 MRI picture showing right and left nasal masses.

**FIGURE 2** Cavernous hemangiomas. (A) Preoperative endoscopic view. (B) Postoperative endoscopic view. (C and D) Naked eye picture.



**FIGURE 3** Histopathology picture of the cavernous hemangioma showing nasal mucosal covering with underlying vascular lesion composed of mainly dilated capillaries filled by blood (A and B); bland looking endothelial cells (C and D) H&E stain  $\times 40, 100, 200$  &  $400$ .



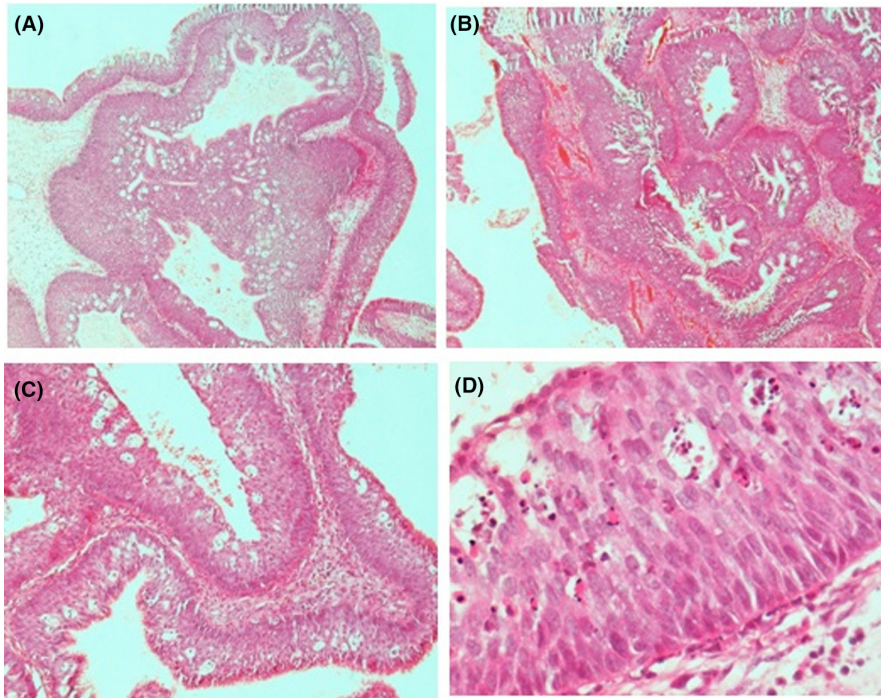
### 3 | DISCUSSION

Although hemangiomas are frequently seen in the medical practice without the exception of the head and neck region, those arising in the nasal cavity and/or paranasal sinuses are rare.<sup>6</sup>

There are multiple known classification systems for hemangiomas in scholarly literature, with histological subtyping being the most accepted and widely used classification, according to the microscopic features, where the hemangiomas are classified into three categories based on the predominant vascular size: capillary, cavernous, and

mixed types.<sup>7</sup> Histologically, cavernous hemangiomas consist of vascular spaces lined with flat endothelial cells containing red blood cells (RBCs). When hemangioma arises in the nasal cavity, it is predominantly capillary and originates from the nasal septum (about 80% of the cases), namely in the Little area or the Kiesselbach triangle.<sup>8</sup>

Here, we report a case of cavernous variety in the right inferior turbinate. Few previously reported cases originated from the inferior turbinate,<sup>9–12</sup> but Caylakli et al reported cavernous hemangioma in the middle turbinate,<sup>13</sup> Nakahira et al reported in vomer,<sup>14</sup> Graumüller et al demonstrated a case in lamina perpendicularis ossi



**FIGURE 4** Inverted growth, stratified epithelium, and intraepithelial microabscess in inverted Papilloma (A)  $\times 40$ , (B)  $\times 100$ , (C)  $\times 200$  & (D) 400 H&E stains.

ethmoidalis,<sup>15</sup> Engels et al reported a case in the sinus maxillaris,<sup>16</sup> and recently Yeon et al published a case report on the lateral wall of the inferior nasal meatus.<sup>17</sup> Extremely rare conditions are seen in patients with concomitant malformation or benign or malignant lesions in the sinonasal sinus anatomical site. Our patient had a history of progressive right and left-sided nasal obstruction without a history of excessive bleeding and experienced endoscopic sinus surgery removing polypoidal masses from both sides with quite excessive intraoperative bleeding from the right side that has been proven to be a cavernous hemangioma, whereas the left side polyp was proven to be an inverted papilloma.

Nasal **inverted papilloma** is a also rare **benign tumor** with a high incidence rate in both the **nasal cavity** and sinuses. It has a probability of easy prolapse and **malignant transformation**, so radical surgery is the first line of **treatment**.<sup>18</sup>

Pathology is the gold standard for diagnosis and differential diagnosis of the inverted papilloma, as it may coexist with the inflammatory process of the nose, showing inflammatory polyps.<sup>19</sup> Recently, **morphometric** differences in the head and neck tumors have aided in roughly differentiating the challenging neoplastic lesion, including the papilloma.<sup>20,21</sup> However, most of these methods are invasive, **endoscopic** images can also be informative, and the **deep learning** network processing these images has recently demonstrated its validity.<sup>21</sup> Inverted papilloma may coexist with the inflammatory process of the nose, exhibiting inflammatory polyps.<sup>19</sup>

Recent morphometric differences in head and neck tumors, including papillomas, have aided in distinguishing the difficult neoplastic lesion.<sup>20,21</sup> However, most of these methods are invasive, endoscopic images can also be informative, and the deep learning network processing these images has recently demonstrated its validity.<sup>21</sup>

EGFR mutations might be the early event initiating this tumor's development by enhancing epithelial cell proliferation, and numerous factors, including viral infection (such as HPV), smoking, and **chronic inflammation**, can contribute to nasal inverted papilloma, however, the pathogenesis of the inverted papilloma is still unclear. Recently, some authors demonstrated the distinct inflammatory pattern of the inverted papilloma, implying the oncogenic role of the acute inflammatory cells, neutrophils, in the pathogenesis of the inverted papilloma.<sup>22</sup>

As the incidence of malignant transformation has been reported variably, ranging from 5% to 80%, so it is mandatory to continue follow-up with endoscopic surveillance for early detection of recurrence. The patient with this reported condition had no evidence of recurrence or suspicious lesions after the complete excision of both lesions during a 2-years follow-up period.

#### 4 | CONCLUSION

Two rare nasal lesions in one patient are reported, who experienced complete removal of the right nasal cavernous

hemangioma without complications and simultaneously complete excision of left sinonasal inverted papilloma with free 2-year follow-up; however, the final diagnosis of this patient came with the routine histopathology examination.

## AUTHOR CONTRIBUTIONS

**Abdulkarim Hasan:** Conceptualization; data curation; project administration; formal analysis; investigation; methodology; software; visualization; writing – original draft; writing – review and editing. **Muhammad Yousaf Mairaj:** Conceptualization; data curation; investigation; methodology; resources; writing – original draft. **Ahmed Elbadawy:** Conceptualization; data curation; methodology; resources; validation; visualization; writing – review and editing. **Sameh Zayed:** Conceptualization; investigation; software; visualization; writing – review and editing. **Abdulkhaliq Saeed Alghamdi:** Conceptualization; investigation; software; writing – review and editing. **Saeed Awad Alrashdi:** Conceptualization; formal analysis; investigation; methodology; software; writing – review and editing. **Khalid Nafie:** Conceptualization; data curation; formal analysis; investigation; methodology; supervision; validation; visualization; writing – original draft; writing – review and editing.

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## CONFLICT OF INTEREST STATEMENT

The authors declare no conflict of interest.

## DATA AVAILABILITY STATEMENT

The data that support the findings of this study are available from the corresponding author upon reasonable request.

## CONSENT

Written informed consent was obtained from the patient to publish this report in accordance with the journal's patient consent policy.

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