

**Abstract**

**Objective:** Young's procedure, or closure of the nares, is an effective procedure for patients with treatment-refractory epistaxis secondary to hereditary hemorrhagic telangiectasia (HHT). Complications following this procedure are rare but may include persistent epistaxis, the development or worsening of obstructive sleep apnea, and patient intolerance of the lack of a nasal airway. Herein we present a patient who developed a symptomatic intranasal fungus ball one year after nasal closure.

**Methods:** Case report and review of the literature.

**Results:** A 44 year old female underwent bilateral nasal closure (Young's procedure) and one year later developed persistent foul smelling postnasal drainage, nasal fullness, and nearly daily nausea and retching. Maxillofacial CT revealed heterogenous material in the nasal cavity and ethmoid sinus mucosal thickening. Due to worsening symptoms, the patient underwent a sublabbial approach to the nasal cavity for examination and debridement, which revealed amorphous brown material occupying roughly half of the nasal cavities bilaterally and extending into the choanae. Frozen section and permanent pathologic and microbiologic analysis revealed hyphae with cultures positive for *Aspergillus fumigatus* and *Klebsiella pneumoniae*, likely representing bacterial colonization or superinfection. One month postoperatively the patient noted no malodor, decreased nasal fullness, and a marked reduction in nausea and retching.

**Conclusions:** This represents the first documented case of fungus ball complicating nasal closure (Young's procedure) for hereditary hemorrhagic telangiectasia. The sublabbial approach to entering the nasal cavity at the level of the piriform aperture is a safe, effective method for endoscopic nasal or sinus surgery after nasal closure that does not necessitate reversal of the procedure.

**Case Presentation**

- A 44 year old female who underwent a modified Young's procedure for refractory HHT.
  - Young's procedure technique
    - 1) KTP laser ablation of intranasal telangiectasias followed by antibiotic irrigation
    - 2) Trilobed flap with circumferential incision at the mucocutaneous junction bilaterally<sup>1-3</sup>.
- Clinical course
  - 13 months post-Young's procedure: development of sensation of thick postnasal drainage with nausea, vomiting, and intermittent epistaxis
  - Initial maxillofacial CT suspicious for retained intranasal blood products
  - 15 months post-Young's procedure: progression of opacification intranasally (Figure 1)
- Operative examination of nasal cavity and paranasal sinuses
  - Operative technique (see below)
  - Intraoperative findings: extensive fungal debris and inflammatory response throughout bilateral nasal cavities
- Postoperatively
  - Reduction in post-nasal drainage symptoms, foul smell, nausea, and retching
  - Cultures demonstrated *Aspergillus fumigatus* and *Klebsiella pneumoniae*

**Figure 1: Preoperative CT**

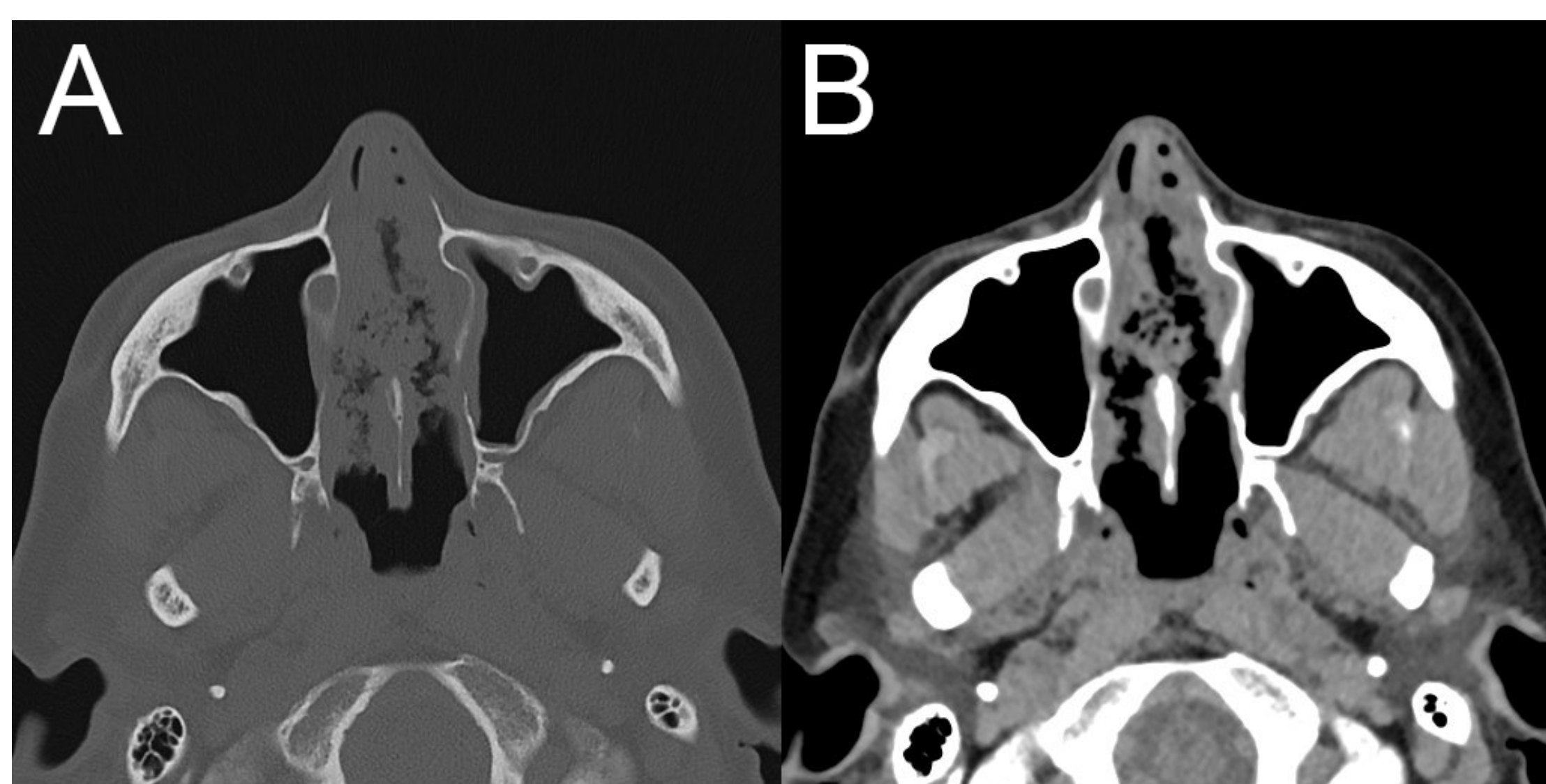


Figure 1: Preoperative bone windowed (A) and soft tissue windowed (B) axial CTs demonstrating heterogeneous material occupying bilateral nasal cavities. No hyperattenuation suggestive of fungal elements was seen.

**Figure 2: Sublabbial Approach**

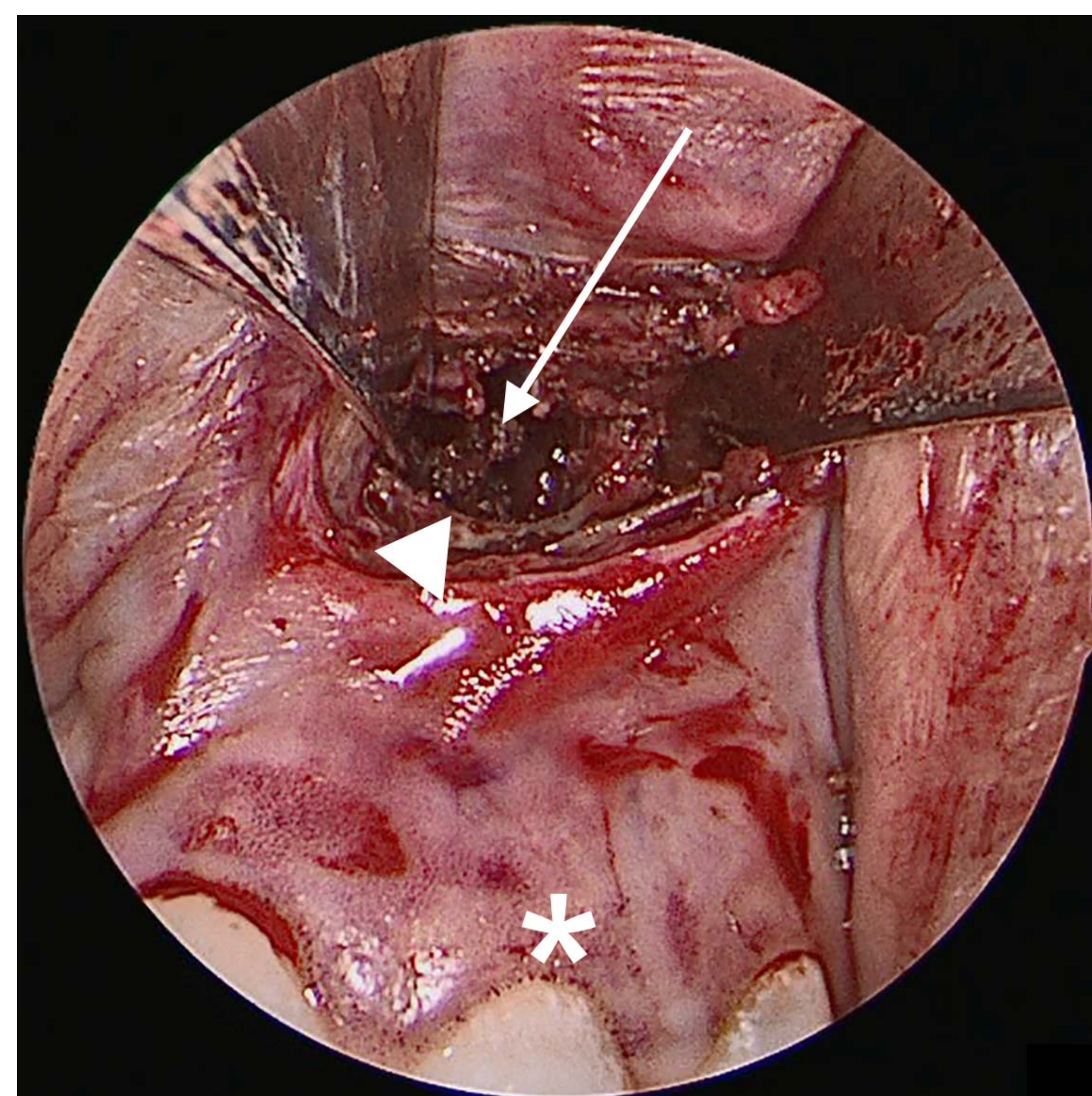


Figure 2: Intraoperative photograph demonstrating sublabbial approach. Under general anesthesia, an incision was made in the upper midline gingivolabial sulcus at the level of the piriform aperture. The mucosa of the floor of the nose was incised and the nasal cavity was subsequently entered. A retractor used for transsphenoidal surgery was placed and rigid 0, 30, and 45 degree endoscopes were utilized to examine the nasal cavities and paranasal sinuses.

Legend:

\* = tooth #9

White arrowhead = piriform aperture

White arrow = amorphous fungal debris in the nasal cavity

**Discussion**

- Complications following nasal closure (Young's procedure) are uncommon, and include epistaxis, development of sleep apnea (or worsening of pre-morbid sleep apnea), untoward speech changes, and intolerance of obligate oral breathing.
- In-office examination of the nasal cavity is impossible using anterior rhinoscopy and endoscopy
- Differential diagnosis is unclear for intranasal pathology following nasal closure
- The "aerogenic" pathophysiologic mechanism is suspected in this patient, whereby airborne fungal spores become sequestered in the nasal cavity<sup>6</sup>.
- Poor mucociliary clearance in this patient may have facilitated lack of clearance (nasal septal perforation, multiple surgeries)
- Sublabbial incision commonly used for correction of congenital piriform aperture stenosis, transseptal pituitary approaches

**Figure 3: Identification and Removal of Fungus Ball**

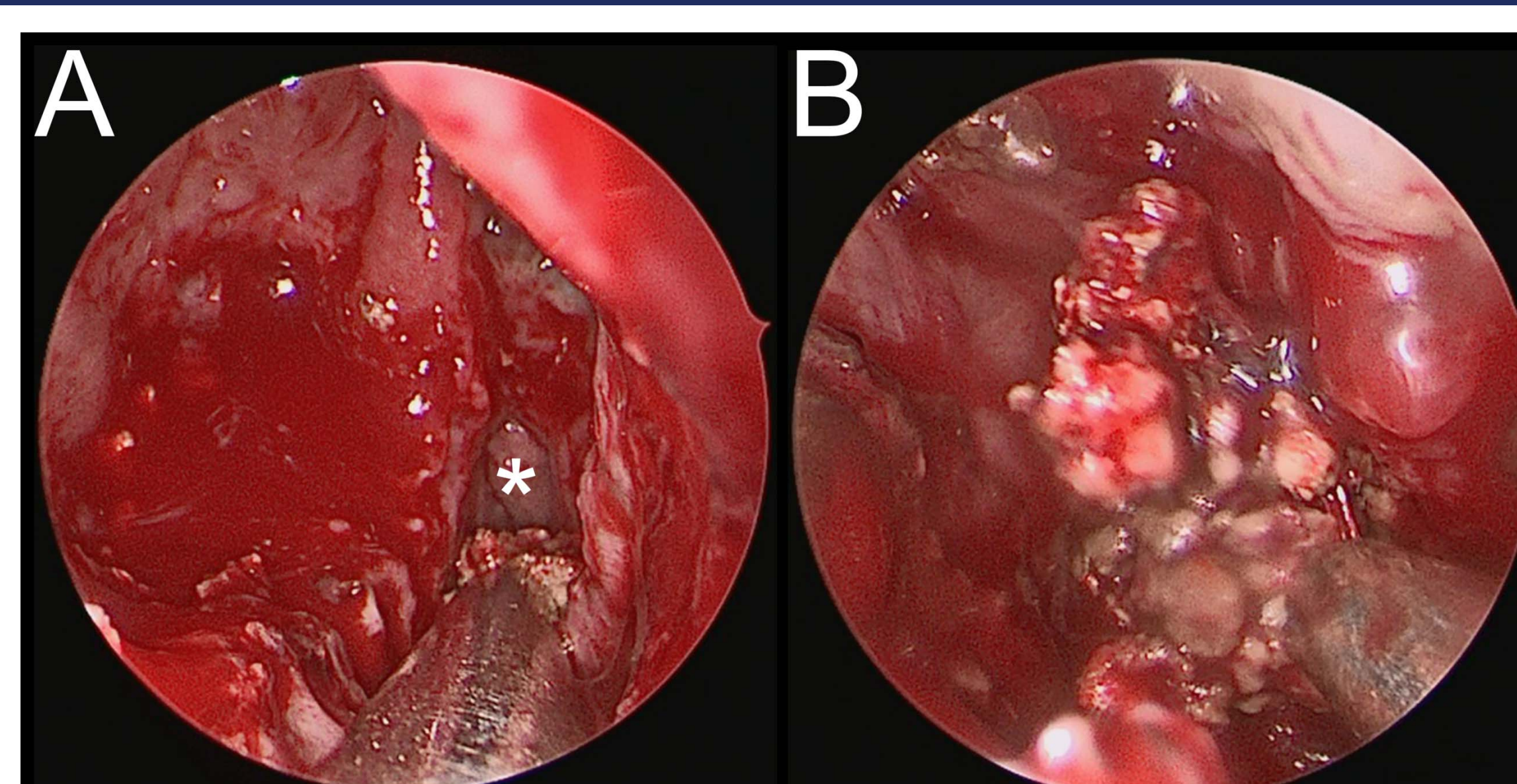


Figure 3: Intranasal fungal elements extending from choana (A, asterisk) to anterior third of the nasal cavity (B). Note the hyperemic surrounding tissue and inflammation from fungus ball and superimposed bacterial infection.

**Conclusions**

- To our knowledge, this is the first description of an intranasal fungus ball following nasal closure (Young's procedure) for treatment-refractory HHT.
- Given the inability to perform endoscopic examinations and treatment of intranasal pathology without reversal of the nasal closure, we utilized a sublabbial approach through the piriform aperture with endoscopic assistance.
- This resulted in a safe, efficient approach to the nasal cavity that may be used for breakthrough epistaxis, paranasal sinus, or skull base surgery in patients who have undergone Young's procedure.

**References**

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