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# Bilateral Sinonasal Inverted Papilloma: Report of an Uncommon Case Involving Sinuses of the Face and Orbital Cavity

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Data Collection B  
Statistical Analysis C  
Data Interpretation D  
Manuscript Preparation E  
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**Conflict of interest:** None declared

**Patient:** Male, 60-year-old  
**Final Diagnosis:** Sinonasal inverted papilloma  
**Symptoms:** Facial asymmetry • nasal obstruction • right hemiface proptosis  
**Medication:** —  
**Clinical Procedure:** —  
**Specialty:** Dentistry • Pathology • Surgery

**Objective:** Rare disease

**Background:** Nasosinusal papilloma is a benign aggressive tumor. It usually occurs unilaterally in the nasal cavity and can extend to the sinuses. The diagnosis is made by the correlation of findings observed in tomographic and histopathological exams. The recommended treatment is surgical excision with clinical monitoring. Orbital involvement occurs in about 9% of cases of advanced SIP. However, there is no report of cases of a benign tumor that invaded the adjacent soft tissues. Therefore, our objective is to report an unusual case of SIP that bilaterally involved the nasal cavity and maxillary sinuses, and extended to involve the ethmoidal cells and sphenoid and frontal sinuses.


**Case Report:** In this article, we report an unusual presentation of sinonasal inverted papilloma (SIP) in a 60-year-old man. The tumor bilaterally involved the nasal cavity and maxillary sinuses and extended to involve the ethmoidal cells and the sphenoid and frontal sinuses, as well as the orbital cavity on the right side. An open surgical procedure was performed for complete removal of the lesion and follow-up with imaging exams.

**Conclusions:** The involvement of these structures is uncommon in SIP. This highlights the importance of this case report. Diagnosis and surgical treatment must be carefully planned. In this work, we describe all the steps that helped guide the choice of the best surgical technique to be performed and offer the best clinical follow-up.

**Keywords:** Case Reports • Diagnosis • Papilloma, Inverted • Paranasal Sinus Diseases • Pathology • Tomography, Spiral Computed

**Abbreviations:** SIP – sinonasal inverted papilloma; SP – sinonasal papilloma; CT – computed tomography

**Full-text PDF:** <https://www.amjcaserep.com/abstract/index/idArt/929910>

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## Background

A Sinonasal papilloma (SP) or nasal sinus papilloma is a benign neoplasm that constitutes 0.4-4.7% of all nasal cavity tumors. In the majority of cases, it originates from the lateral nasal wall [1-3]. SPs can be classified into 3 histopathological patterns [4,5]. The inverted pattern with endophytic growth constitutes between 47% and 79% of SPs and is localized almost exclusively to the side walls of the nasal cavities [6]. The exophytic or fungiform SP constitutes between 18% and 50% of papillomas, generally originates from the nasal septum, and rarely involves the paranasal sinuses [7]. The oncocytic or cylindrical SP constitutes between 3% and 8% of SPs and is considered by many authors to be a variant form of inverted papilloma [8].

Sinonasal inverted papilloma (SIP) usually affects men, with a male-to-female ratio of 4: 1, and normally occurs between the ages of 40 and 70 years; it is rarely observed during childhood and adolescence [9-11]. In descending order, the most commonly affected paranasal sinuses are the maxillary, ethmoidal, frontal, and sphenoidal; extension to the base of the skull is rare [12,13]. Clinically, the most common symptom that makes the patient seek medical treatment from an otolaryngologist is unilateral nasal obstruction [14,15]. Other frequently reported symptoms include epistaxis, rhinorrhoea, and epiphora [4,6,11]. Bilateral extension of the tumor is rare, and when it occurs, it is associated with destruction of the adjacent structures and a tendency for recurrence. In addition, the tumor can undergo malignant transformation [7,6,9].

Here, we report a rare case of bilateral SIP. The tumor was diagnosed using a computed tomography (CT) examination and confirmed via histopathological examination [2]. The tumor bilaterally involved the nasal cavity and maxillary sinuses, and extended to involve the ethmoidal cells and sphenoid and frontal sinuses as well as the orbital cavity of the right side and the adjacent soft tissue.

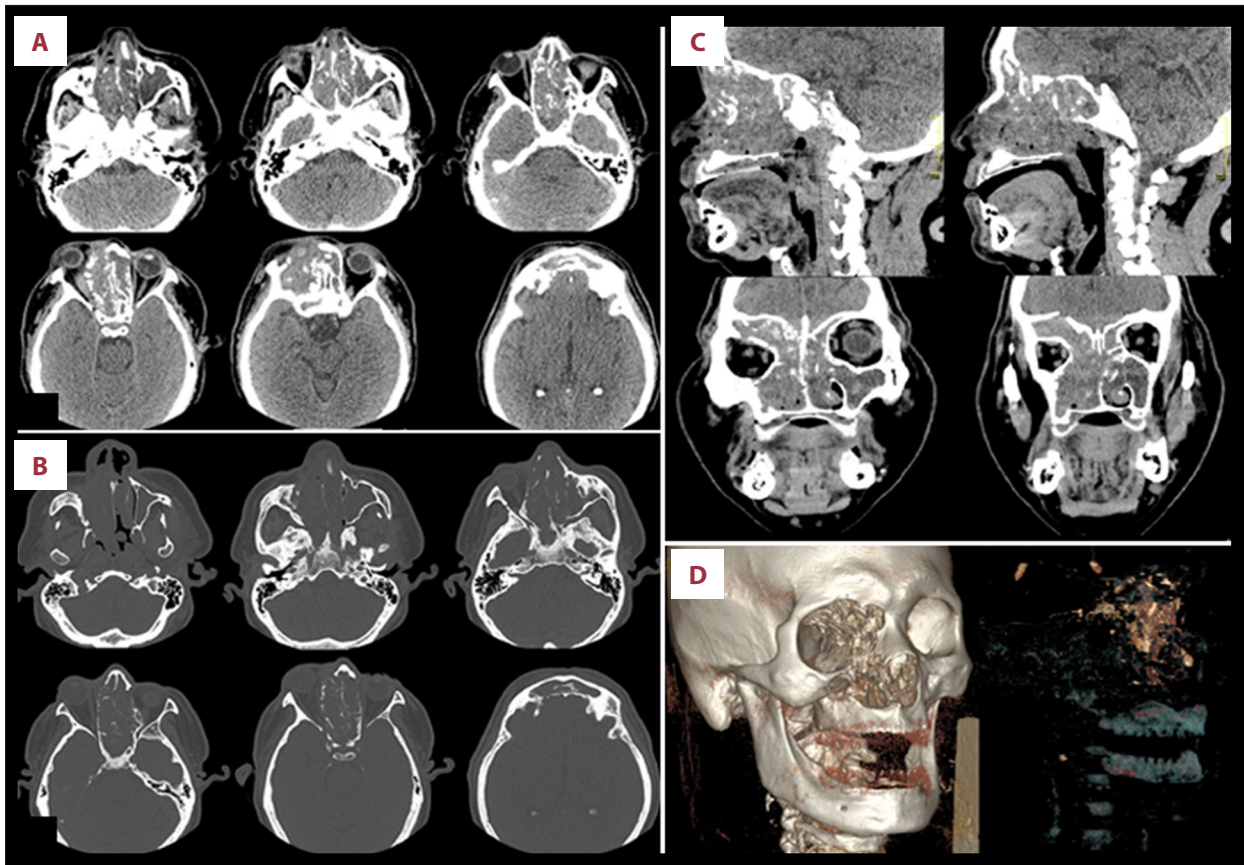
## Case Report

A 60-year-old man presented at the Ophir Loyola Hospital outpatient clinic in 2014 with facial asymmetry, right hemiface proptosis, and nasal obstruction (**Figure 1**). An extraoral clinical examination did not detect evidence of dental or systemic infection or palpable nodules. The patient denied smoking, alcoholism, previous radiotherapy, and family history of concomitant diseases. In the intraoral examination, there was an increase in volume in the left posterior palate, normal coloration, and absence of ulceration. Computed tomography was requested as an auxiliary imaging exam.

CT examination was performed, and it showed an expansive and infiltrative lesion with involvement of the nasal cavity, bilateral maxillary sinuses, ethmoidal cells, and frontal and sphenoidal sinuses, which were all obstructed. There was also a small extension of the lesion to the superficial tissues in the medial corner of the right orbit (**Figure 2**).



**Figure 1.** Clinical aspect before the surgical treatment. (A) frontal image showing facial asymmetry and proptosis on the right side. (B) image showing nasal obstruction.



**Figure 2.** Initial radiographic examinations. CT (axial cross-section). (A) Soft tissue window shows a discreet extension of the lesion to the superficial tissues in the medium bottom from the right orbit. (B) Bone window shows erosion of the bone structure and presence of a continuous solution in the walls of the maxillary sinus, ethmoidal intercellular septum, sphenoidal sinus wall and frontal sinus, notably, in the medium and top walls from the right orbit. (C) CT from sagittal and frontal cross-section: soft tissue window shows extension of the lesion to the paranasal sinuses, the nasal cavity bilaterally, and involvement of the right orbit cavity. (D) Volumetric reconstruction illustrating the tumor extension.

The differential diagnosis includes acute, chronic or fungal sinusitis, retention cyst, mucocele, benign, malignant and metastatic neoplasia, fibrous dysplasia, ossifying fibroma, foreign body, encephalocele, internal carotid artery aneurysm and sphenoidal polyp [14].

The anatomopathological diagnosis of SIP can be a challenge, as there is a tendency for this lesion to become malignant. The biological behavior of SIP encompasses a spectrum of activity ranging from limited nasal growth to aggressive and infiltrative lesions, with multiple recurrences and malignant transformation [15].

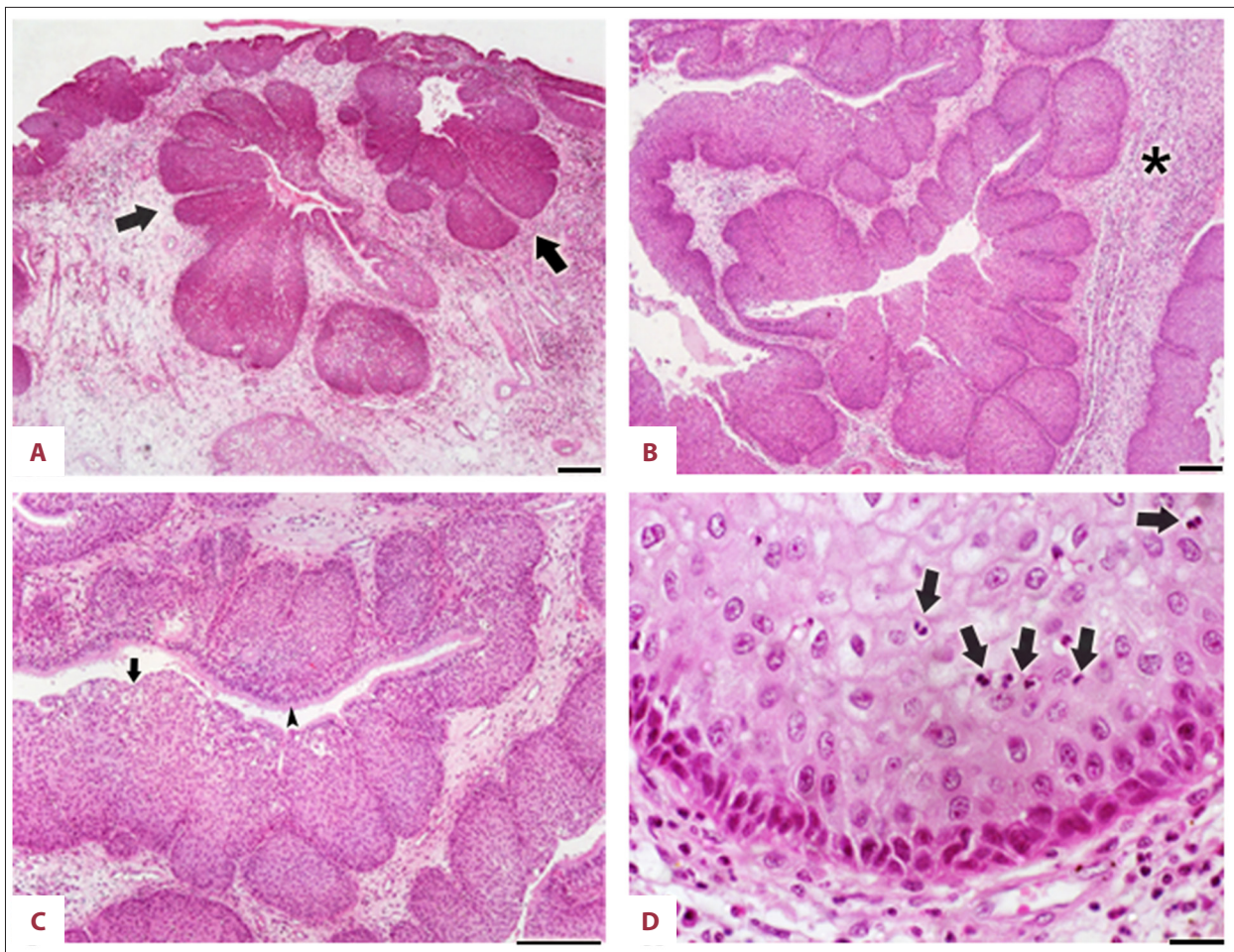
A transnasal biopsy was performed and histopathological slices showed fragments of mucous membrane that was involved by a stratified squamous keratinized epithelial tissue showing papillary projections of various sizes toward the adjacent tissue and koilocytes in the superficial layers. Hyperchromatic cells with large nucleus and scarce cytoplasm were observed

in focal areas of the basal and parabasal layers. In other areas, respiratory epithelium was observed.

In the lamina propria, which is made up of floppy connective tissue, an inflammatory lymphoplasmacytic infiltrate that permeated the congested blood vessels was observed. Areas of hemorrhage and hemosiderin pigmentation were also observed (Figure 3). Histopathological characteristics revealed a diagnosis of sinonasal inverted papilloma.

Due to the size of the lesion, in which all of the patient's paranasal sinuses were affected, a less invasive approach, such as nasosinus endoscopic surgery, was not possible. In view of this, the surgical intervention of choice was Le Fort I osteotomy, performed through the bicoronal route with a Weber-Ferguson incision (Figure 4). After removal of the lesion, which had an area of 13 cm<sup>2</sup>, the patient was referred for histopathological examination.





**Figure 3.** Histopathological exam. (A) Mucosis fragments covered with epithelial keratinized and non-keratinized tissue. Epithelial papillary projections through the adjacent stroma that confers an inverted architecture in relation to the neoplastic epithelium and the presence of continuous cells nests with the epithelial surface (arrows). Scale bar: 200  $\mu$ m. (B) Stratified epithelial tissue and conjunctive tissue loosely ordered with an inflammatory lymphoplasmacytic infiltrated (asterisk). Scale bar: 200  $\mu$ m. (C) Stratified squamous keratinized epithelial tissue showing papillary projections of many sizes (arrow). In other area, was observed respiratory epithelium (arrowhead). (D) Presence of hyperchromatic cells, with voluminous nucleus in the epithelium basal and parabasal and koilocytes in the superficial layers (arrows). Scale bar: 20  $\mu$ m.

The postoperative evolution was satisfactory and there were no adverse or unforeseen events. The control was carried out in 2 stages. One week after the operation, another CT scan exam (Figure 5) was performed for post-surgical evaluation. The patient recovered well and underwent clinical and imaging assessments in the last 6 months. After 5 years, control was performed and the patient no longer reported symptoms.

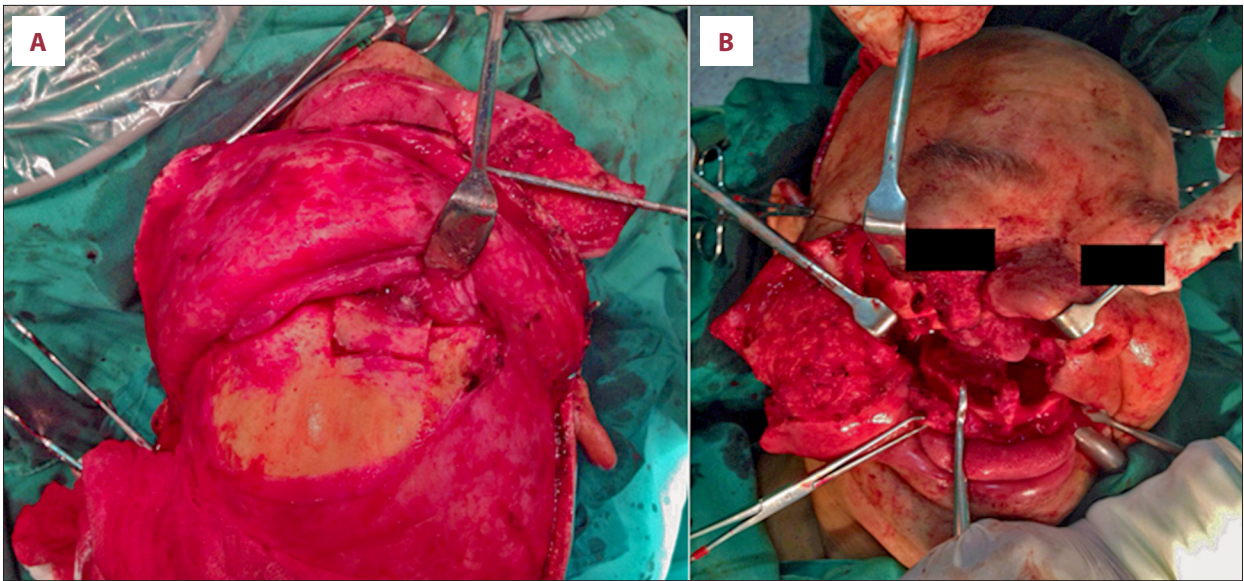
## Discussion

SIP is a benign tumor that has its origin in the Schneiderian epithelium of the lateral nasal wall (80%) [13]. Its etiology is not yet defined. This tumor is more commonly observed in men aged between 40 and 70 years [7], and its most important

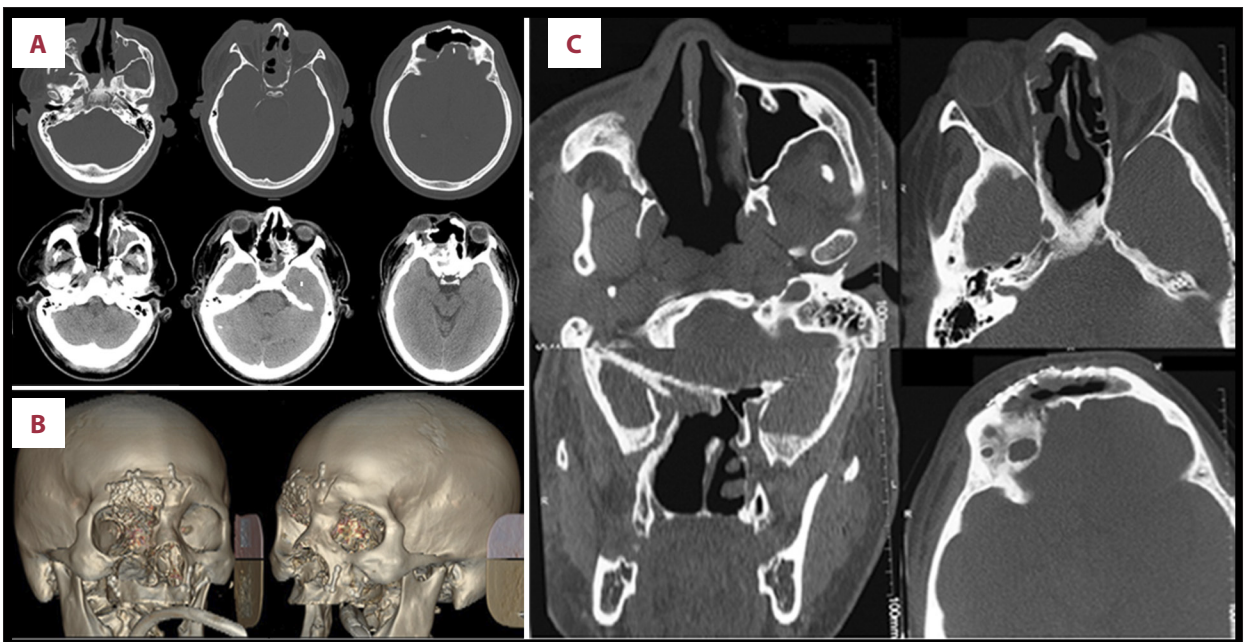
characteristics are the tendency of recurrence, local aggressiveness, and association with malignant transformation [12,13]. Consistent with the literature, the current case of SIP was a man who was 60 years old. The lesion originated in the right lateral nasal wall, and it was expansive and had an aggressive behavior; it did not undergo malignant transformation [17].

Nasal obstruction was the main symptom in this case, which is in agreement with the literature, which reports that this symptom is the main presentation in 78-100% of cases [17]. There are other symptoms associated with SIP, such as rhinorrhoea, epistaxis, increasing volume of the nasal area, and headache [3,4,6,8].





**Figure 4.** Intraoperative image. (A) Weber-Ferguson's access with Le Fort I osteotomy. (B) Bicoronal access.



**Figure 5.** Radiographic images of post-surgery and follow-up. (A) Axial cross-section showing surgical gaps. (B) Volumetric reconstruction showing titanium plates used for the bone fixation. (C) Control CT made 6 months after the surgery. Axial cross-section and front cross-section showing solution of continuity of the wall bone of the right antrum maxillary, ethmoidal cells, sphenoidal, and frontal sinuses, as well as plates and metal screws of osteosynthesis.

The tumor extended into the nasal cavity and maxillary sinuses bilaterally, ethmoid cells, and frontal and sphenoid sinuses. It also extended into the orbital floor, causing destruction and erosion of the bone. In addition, there was slight extension of the tumor to the superficial tissues in the right middle portion of the orbit. Only 3-5% of cases of SIP described in the literature have bilateral involvement of the nasal cavity and maxillary sinuses [9,3,4]. Orbital involvement occurs in

about 9% of cases of advanced SIP [8]. However, there are no reports of cases of benign tumors that invaded the adjacent soft tissues [10,11].

An advanced search was performed in the PubMed and Lilacs databases using the terms “sinonasal inverted papilloma”, “case report”, and “benign”. The search resulted in 66 studies, but none presented SIP with involvement in all the anatomical

structures that we describe in this report. To date, PIS with benign characteristics and simultaneous involvement of bilateral maxillary sinuses and nasal cavity, ethmoidal cells, frontal and sphenoid sinuses, orbit, and soft tissues have not been described.

In the present case, the extension of the lesion to the adjacent areas as demonstrated by imaging showed that this tumor had invasive characteristics, although there was no evidence of malignancy.

A histopathological examination confirms the diagnosis of SIP. This is important to exclude the presence of vascularized tumors (such as juvenile nasal angiofibroma) or lesions with extension to the central nervous system (such as meningocele and meningoencephalocele) [16]. The histopathological characteristics in this case allowed its classification as a benign tumor because there was no epithelial invasion of the stromal tissue.

The treatment that is recommended in the literature is open surgery and endoscopy, which ensures a satisfactory prognosis; however, each technique has its limitations depending on the behavior of the lesion. One possible limitation of the use of endoscopy as the only treatment is the difficulty in accessing the lesions localized in areas such as the frontal sinus or the bottom and posterior part of the maxillary sinus. In these areas, it is not feasible to use endoscopy alone [8]. Due to this limitation, many authors describe a combination of techniques as the better option for treatment [1,9,11].

The treatment option performed in this case were performed openly. Due to the expansive and destructive behavior of this lesion, a bicoronal approach was chosen with the Weber-Ferguson incision and Le Fort I osteotomy for total tumor removal.

Tumor recurrence usually occurs within the first 2 years, but in 17% of cases it occurs after 6 years of evolution, which justifies follow-up for at least this period [4,5].

The patient has been followed up for 5 years and 6 months by clinical and tomographic exams. The images obtained (CT) after surgery did not show the tumor, showing a better prognosis of the case.

## Conclusions

The importance of this case is related to the rarity and aggressiveness that the bilateral inverted nasosinus papilloma presents. Thus, due to its aggressive behavior, even though it is a benign lesion, the diagnosis and surgical treatment must be carefully planned; only then is it possible to offer a better quality of life to the patient. In this work, we report an unusual case of SIP involving the nasal cavity and maxillary sinuses bilaterally. The surgical approach was performed by Le Forte I, since, exceptionally, all the patient's paranasal sinuses were affected by the lesion, which did not allow a less invasive approach.

## Conflict of Interests

None.

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