



# Isolated Sphenoid Lesions

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## Abstract

**Background:** Isolated Sphenoid Sinus Lesions (ISSL) are rare. They occur in 1% to 3% of all PNS lesions. Diagnosis is made by clinical evaluation, endoscopic examination and with imaging. Aim of this study to describe the different clinical presentations of isolated sphenoid lesions, the differential diagnosis, radiological findings and the endonasal endoscopic navigation assisted management.

**Method:** A retrospective study of the minimal invasive navigation-assisted endoscopic approach to different pathologies, affecting the sphenoid sinus at King Fahad Specialist Hospital, Dammam (2009 to 2019).

**Results:** Out of 50 cases found, there were 12 cases of bacterial sphenoid sinusitis (24%), 4 cases of allergic fungal sinusitis (8%), 4 cases of fungal ball (8%), 2 cases of invasive fungal sinusitis (4%), 3 cases of pediatric (both allergic fungal and sphenoid sinusitis) (6%), 1 case of mucocele (2%), 1 case of mycopyocele (2%), 1 case of inverted papilloma (2%), 2 cases of Lymphoma (4%), 2 cases of SCC (4%), 1 case of adenoidcystic carcinoma (2%), 2 cases of metastatic carcinoma (4%), 2 cases of fibrous dysplasia (4%), 2 cases of osteoma (4%), 7 cases of CSF leak (14%), 1 case of meningocele (2%) and 3 case of asymptomatic (6%).

**Conclusion:** Isolated sphenoid sinus disease has a variable prognostic and pathological range yet has a common presentation.

Most commonly headache will be the presenting symptom and to confirm the diagnosis, CT is the imaging of choice.

Pathologically it varies, from bacterial sinusitis to a carcinoma invading the cavernous sinus.

**Keywords:** Sphenoid sinus; Sinusitis; Isolated sphenoid lesions; Sphenoid sinus surgery

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Received Date: 28 Apr 2021

Accepted Date: 18 May 2021

Published Date: 21 May 2021

### Citation:

AlMomen A, Alali M, Alkhalidi SK, Alsheef H, Aldhuraish A, Alawami B, et al. Isolated Sphenoid Lesions. *Am J Otolaryngol Head Neck Surg.* 2021; 4(4): 1134.

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## Introduction

Isolated Sphenoid Sinus Lesions (ISSL) are considered to be rare. They occur in 1% to 3% of all PNS lesions [1]. Typically, symptoms if the disease do not appear in the early stages or are non-specific, thus making it difficult to diagnose. It is most likely due to the few numbers of studies reporting cases of isolated sphenoid sinus disease. Diagnosis is made clinically, endoscopically, and with the help of current imaging techniques such as CT scan or MRI. All the three previously mentioned modalities, especially endoscopic examination and imaging techniques increased the likelihood of diagnosing such a disease.

Computed Tomographic scan (CT) is used as first choice of investigation [2].

According to the literature, the most common cause of sphenoid sinus lesions is inflammatory in up to 65% to 72% of cases, followed secondly by neoplasms which accounts for 18% (in benign) and 10.9% (in malignant) of the cases respectively [3]. Adjacent to the sphenoid sinus, there are many vital and crucial structure, including optic chiasma and nerve, cranial nerves (3<sup>rd</sup>, 4<sup>th</sup>, 5<sup>th</sup> 1&2, and 6<sup>th</sup>), middle cranial fossa, and cavernous sinus. Thus, advancement of the sphenoid sinus disease may lead to life-threatening complications. Common presentations in these patients include headache (33% to 81%), visual disturbance (24% to 50%) [3], nasal obstruction and discharge, ptosis and asymptomatic. The goal of this study to describe the different clinical presentations of isolated sphenoid lesions, the differential diagnosis, radiological findings and the endonasal endoscopic navigation assisted management cases.

## Methods

A retrospective study of the minimal invasive navigation-assisted endoscopic approach

to different pathologies, affecting the sphenoid sinus at KFSH, Dammam in the Otorhinolaryngology Department (2008 to 2017). KFSH, Dammam, Eastern Province, SA. A tertiary, with capacity of 640 beds, and equipped with latest equipment's.

The study is a review study of the medical records, including observation charts, clinical data, and radiological findings of 50 patients with isolated sphenoid sinus. This study includes asymptomatic patients that were diagnosed incidentally on an imaging study. All the cases were carefully analyzed in order to point out the leading symptoms of the disease, and to finally determine for how long the symptoms of sphenoid sinus had been there before each patient was referred for medical treatment. This study includes the management with the endoscopic endonasal approach only.

### Results

Out of 50 cases found, there were 12 cases of bacterial sphenoid sinusitis (24%), 4 cases of fungal ball (8%) 4 cases of allergic fungal sinusitis (8%), 2 cases of invasive fungal sinusitis (4%), 3 cases of pediatric ( both allergic fungal and sphenoid sinusitis) (6%), 1 case of mucocele (2%), 1 case of mycopyocele (2%), 1 case of inverted papilloma (2%), 2 cases of Lymphoma (4%), 2 cases of SCC (4%), 1 case of adenoidcystic carcinoma (2%), 2 cases of metastatic carcinoma (4%), 2 cases of fibrous dysplasia (4%), 2 cases of osteoma (4%), 7

cases of CSF leak (14%), 1 case of meningocele (2%) and 3 case of asymptomatic (6%) (Figure 1, Table 1).

### Discussion

Isolated sphenoid sinus pathology is diagnosed more frequently than in the past due to increased availability and accuracy of radiological studies. A large series study was first to report in the medical literature (O. Cakmak, M. Shohet and Eugene Kern, Mayo Clinic). A retrospective study and chart review of 182 cases ISSL (1935 to 1998) found ages of patients were between 8 to 91 years (56 to 57 years as a mean), followed up from a period ranging from 3 months and up to 26 years. Isolated sphenoid lesions are usually insidious in onset; the most common presentation of these patients is headache, which can be frontal, retro-orbital, occipital, and vertex and diffuses [4].

The second common presentation is visual disturbance and diplopia. Others include nasal obstruction with nasal discharge, and cranial nerve palsies which are more common to be found in patients with neoplastic and less common in patients with inflammatory disease. The diagnosis of isolated lesions of the sphenoid sinus depends on the clinical, followed by radiologic investigations. CT scan imagings have improved the diagnosis chance of such a disease preoperatively. CT is considered the best modality of diagnosis needed to evaluate diseases of the sinus. However, MRI is essential in selected cases of the sphenoid sinus lesions to give details of soft tissues, if intracranial lesions are suspected and in fungal infections. Angiography can be used if suspected vascular lesion such as JNA.

In our series the pathology spectrum was wide and include 27 inflammatory cases, 12 tumors involved benign and malignant tumors, 8 CSF leaks and meningocele, 3 asymptomatic (Figure 2). According to the literature, inflammatory causes are the most common. Yet, in infectious causes, the patients presented with retro-orbital pain and frontal headache, nasal endoscopy showed inflammatory sings with mucosal edema and mucopurulent discharge, CT scan showing

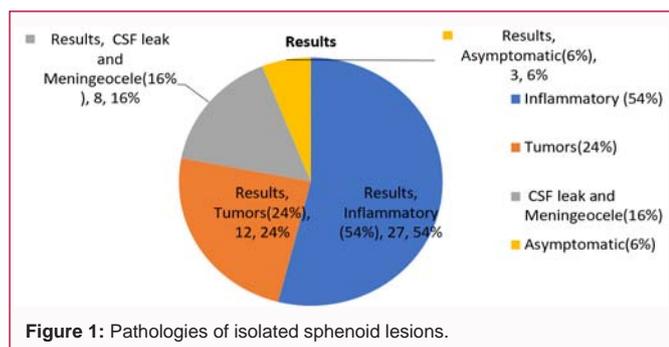


Figure 1: Pathologies of isolated sphenoid lesions.

Table 1: Pathologies of isolated sphenoid lesions.

| Pathologies of Isolated Sphenoid Lesions |  | Number of Cases |
|--|--|-----------------|
| Inflammatory                             | Bact. Sphenoid sinusitis (acute, chronic)  | 12              |
|  | Allergic fungal sinusitis                  | 4               |
|  | Fungal ball                                | 4               |
|  | Invasive fungal sinusitis                  | 2               |
|  | Pediatric: 1 AFS, and 2 sphenoid sinusitis | 3               |
|  | Mucocele, mucopyocele                      | 2               |
| Tumors                                   | Inverted papilloma                         | 1               |
|  | Lymphoma                                   | 2               |
|  | SCC  | 2               |
|  | Adenoidcystic carcinoma                    | 1               |
|  | Metastatic carcinoma                       | 2               |
|  | Osteomas                                   | 2               |
|  | Fibrous dysplasia                          | 2               |
| CSF and Meningocele                      | CSF leaks (7)                              | 8               |
|  | Meningocele (1)                            |                 |
|  | Asymptomatic                               | 3               |
|  | Total                                      | 50              |

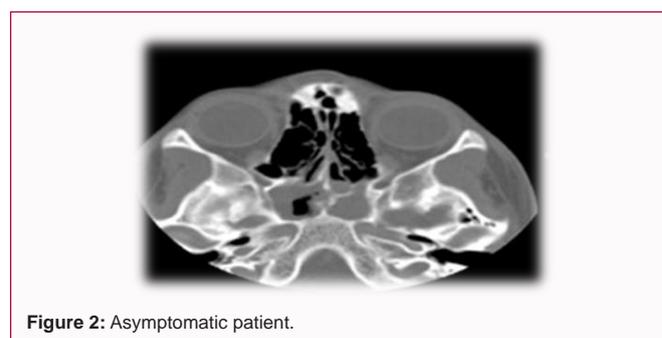


Figure 2: Asymptomatic patient.

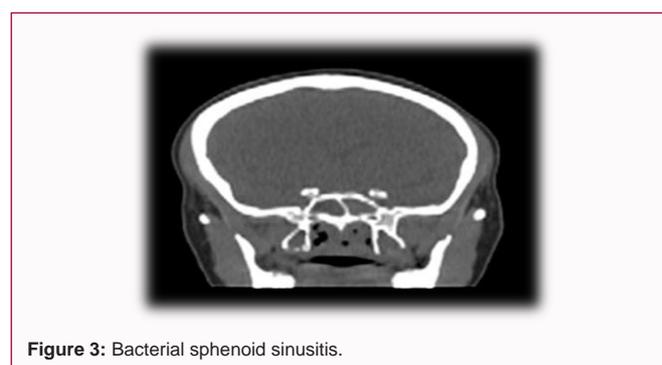


Figure 3: Bacterial sphenoid sinusitis.



Figure 4: Invasive sphenoid sinusitis.



Figure 6: MRI mucocoele.



Figure 5: CT scan mucocoele.

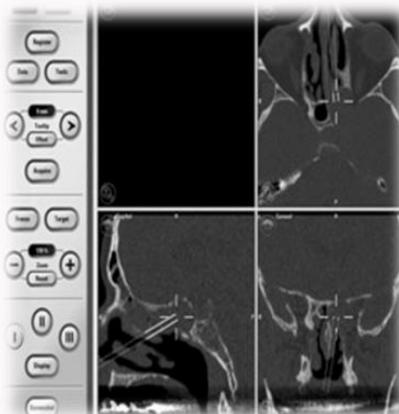


Figure 7: Navigation assisted surgery.

bilateral hypodensity of sphenoid sinuses without involvement of other sinuses (Figure 3). The first line of treatment of uncomplicated sphenoid sinusitis is medical [5]. If there is no improvement after the medical treatment or the patient presented with complications of sphenoid sinusitis endoscopic drainage and widening of sphenoid, ostium is required.

Fungal infections of the sphenoid sinus are the least common of the inflammatory lesions [2]. Generally, fungal sinusitis is classified into two main types, invasive and non-invasive types. Invasive type could be acute (fulminant), chronic or granulomatous (indolent). On the other hand, the non-invasive type includes allergic fungal sinusitis (AFS) and fungus ball [6,7]; with allergic fungal sinusitis being the most common subtype of non-invasive fungal sinusitis [7]. Non-invasive fungal infections with formation of fungal balls are uncommon (we found only 4 cases in our series). Therefore, it is not a surprise that the exact mechanism of spread of fungal infection only to sphenoid sinus is still unknown. However, some hypothesis suggest that the ostial closure creates an anaerobic environment suitable for the growth of *Aspergillus* species, or that chronic sinusitis might predispose the development of fungal balls [8,9]. Hormonal and environmental factors may be involved in the pathogenesis of Sphenoid Sinus Fungal Balls (SSFb) [8,10-13]. The main treatment modality is surgery, where removal of fungal debris from the affected sinus takes place, with re-establishing its proper ventilation and drainage. Allergic fungal sinusitis is present in immunocompetent patients as in the case of our study. The CT scan showed a hyperattenuating allergic mucin within the lumen of the paranasal sinus, with expansion of involved sinuses. Treatment

requires combined surgical and medical treatment with steroid. Acute invasive fungal sinusitis present in immunocompromised patients, in one of our cases of Invasive Sphenoidal Sinusitis (Figure 4), the patient was immunocompromised, on immunosuppressant therapy, treated with endoscopic debridement and antifungal medications.

Regarding mucocoele, sphenoid sinuses are the least common site of a mucocoele to appear in, representing only 1% to 2% of all cases among all age groups. Mucocoeles are benign, encapsulated, expansile, locally invasive masses within a paranasal sinus lined by epithelium and filled with mucus. Mucocoeles are assumed to be due to the obstruction of the insulated sinus. However, there are other hypothesis about the etiology, including cystic dilatation of the glandular structures, and cystic development of the embryonic epithelial residues [14]. Compression of the adjacent structures is what primarily causes the presenting symptoms. Therefore, decline in vision is considered a common presenting symptom too, as the disease may compress structures of the orbit. Acute visual loss is considered the main indication for imaging in most cases of sphenoidal mucocoeles. Another serious complication, yet potentially uncommon is superadded infection followed by brain abscess [15]. The recommended management of a mucocoele of the sphenoid sinus is endonasal sphenoidotomy with sufficient removal of the anterior and inferior wall of the sinus along with drainage of the mucocoele [16]. In some of our cases CT scan and MRI done. CT scan showed expansion of the left sphenoid sinus (Figure 5). MRI showed a high signal intensity of the left sphenoid sinus with enhancement at the

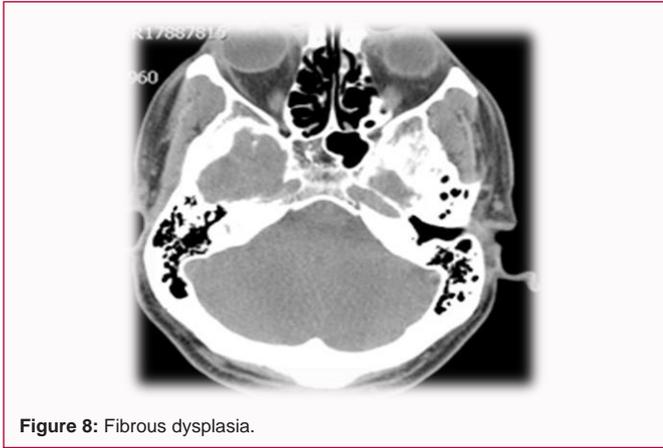


Figure 8: Fibrous dysplasia.

periphery (Figure 6). This case was managed by endoscopic transnasal navigation assisted marsupialization of sphenoid mucocele, (Figure 7).

Fibrous dysplasia is an uncommon, non-neoplastic, primary disorder of bone with unknown etiology. The pathophysiology of the disease takes place in replacing the normal medullary bone with that of a variable amount of abnormal and structurally weak osseous and fibrous tissue. In a study that was reported by the author Lawrence R Lustig et al., the authors reported 21 cases of fibrous dysplasia of facial bones, and surprisingly found an involvement of sphenoid in 43% patients [7]. However, in all these cases there wasn't a single case reporting isolated sphenoid sinus insult, and that what makes our study special. Unlike AFS, in fibrous dysplasia, CT scan is considered the initial diagnostic modality and can give a differentiation of fibrous dysplasia from other osteodystrophies of the skull base, including otosclerosis, osteopetrosis, Paget' disease of the bones, and osteogenesis imperfecta. MRI as another diagnostic modality in this case can also help distinguish fibrous dysplasia from osteoma, meningioma or mucocele. Spontaneous transformation of fibrous dysplasia into a malignancy has been only reported in 0.5% of patients [18]. Regarding malignancies in fibrous dysplasia, osteosarcoma is found to be the most common malignancy, Fibrous dysplasia of sphenoid sinus presented with frontal and retro-orbital headache, in one of our cases CT showed areas of thickening and sclerosis occupying the right sphenoid sinus (Figure 8). The patient underwent an endoscopic sphenoid removal of the tumor through a transethmoidal approach.

In the cases of sphenoid diseases presenting with unilateral rhinorrhea, a CSF leak is mostly the cause (Figure 9), one of the patients is 20 y/o female with hydrocephalus with VP shunt. MRI showed CSF leak of right sphenoid (Figure 10), other patient gave history of recurrent meningitis (Figure11) and the third showed traumatic meningocele with bone fragment (Figure 12). Intraoperative intrathecal fluorescein (Figure 13) and intraoperative topical fluorescein (Figure 14) used to confirm diagnosis and to accurately localize the site of the defects. Navigation assisted nasal endoscopy for repairing of the leaks with different grafting materials (Figure 15).

Out of all cancers, neoplasms of the paranasal sinuses account for only 0.2% to 0.8% and out of head and neck cancers it accounts for 2% to 3%. Among all paranasal sinus tumors, only 1% to 2% reported to have a primary involvement of the sphenoid sinus [19]. Tumors of the sphenoid, benign or malignant, primary

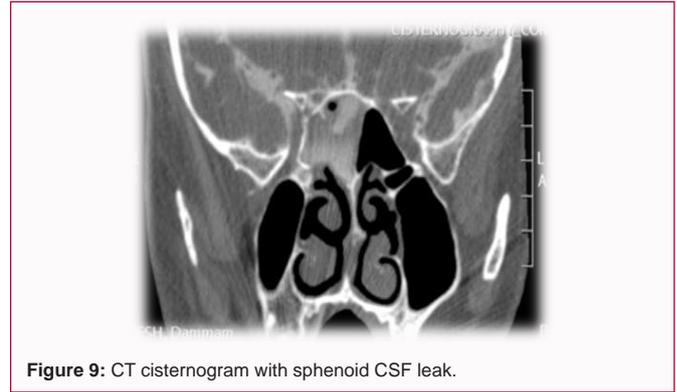


Figure 9: CT cisternogram with sphenoid CSF leak.



Figure 10: MRI with vp shunt and sphenoid csf leak.



Figure 11: CSF leak Lateral sphenoid defect.



Figure 12: Traumatic sphenoid meningocele with bone fragments.

or metastatic. However, metastatic disease is quite uncommon, with renal metastasis, followed by lung and prostate reported in literature. Endonasal endoscopic biopsy with histological study will certainly allow the diagnosis of such a disease. According to the literature, there are several histological types which include:



Figure 13: Intrathecal fluorescein dye in left sphenoid sinus.



Figure 14: Topical fluorescein dye in right lateral sphenoid wall defect.

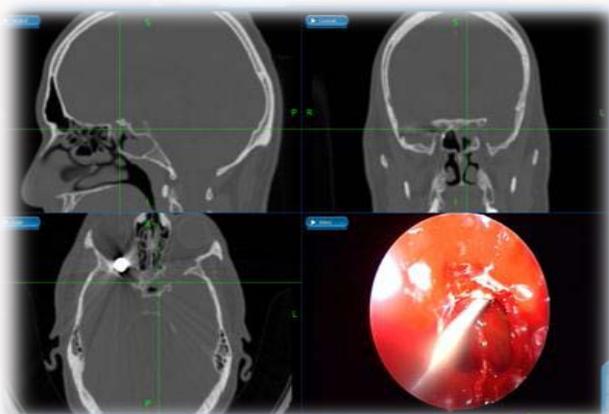


Figure 15: Repair of CSF leak using navigation assisted nasal endoscopy.

Squamous cell carcinoma, undifferentiated carcinoma, adenoid cystic carcinomas, adenocarcinomas, and non-Hodgkin lymphoma (Figure 16), lymphosarcoma, lymphoepithelioma papillary carcinoma, leiomyosarcoma, osteosarcoma, malignant adamantinomas, and finally undifferentiated sarcomas [20-21]. Also, other rare primary malignant tumors of the sphenoid sinuses are also described in the medical literature including esthesioneuroblastoma [22], melanoma [23], and finally myxofibrosarcoma [24]. The management approach to the malignant tumors of the sphenoid sinus is mainly based on the combination of a debulking surgery and radiotherapy [25]. Depending on the stage of the disease upon diagnosis, histopathological type, tumor size, and finally aggressiveness of the disease; the prognosis could be determined [26]. In a case of patient with lymphoma of sphenoid, biopsy was taken from the sphenoid sinus by endoscopic nasal surgery and histopathology confirms the diagnosis.

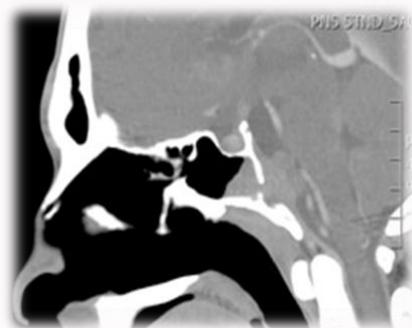


Figure 16: Sphenoid Lymphoma.

Approaches to Sphenoid Sinus nowadays is by endoscopic technique which is considered to be the gold standard for sphenoid sinus surgery, which could be by transnasal approach, transeptal approach, transthmoid approach, or transpterygoid approach, and it is considered a safe technique with excellent visualization of the whole sphenoid sinus. Factors affecting the choice of surgical approach are: Preoperative diagnosis, extent of the sphenoid sinus pneumatization, anatomical location of the disease process, available surgical equipment, level of experience in endoscopic sinus surgery, and extent of the surgery.

Since the inception of the endoscopic approach in the mid 1980s, Navigation Assisted Surgery (NAS) is considered the most significant advances in endoscopic sinus surgery. NAS is a type of technology which provides clear anatomical dissection of the sinuses for the surgeon to follow in a real time manner by monitoring through a computer in the operation room settings. Therefore, difficult anatomical relationships can be easily seen and carefully treated with the comfortability that the all the adjacent critical landmarks are secured. Moreover, a recent meta-analysis declared that both major and total complications are less likely to be encountered with the use of NAS system [27]. In a nation-wide questionnaire survey that was conducted in the US back in the year of 2006, the authors found that revision surgery, followed by frontal and sphenoid sinus surgery are the most considered the most commonly acceptable indication for imaging guided sphenoidotomy [28]. The success rates were higher than 90% when imaging guided sphenoidotomy was done. However, the success rate dropped to 68.6% when revision FESS was performed without imaging guided sphenoidotomy [29]. Moreover, it is more difficult to confirm without imaging guided sphenoidotomy whether the sphenoid sinus is successfully penetrated or not, as there is a possibility to mistake the opening of Onodi cell with the penetrated sphenoid sinus. Therefore, the actual success rate for the sphenoid sinus penetration might be lower than 68.6% in non- imaging guided sphenoidotomy group [29].

## Conclusion

Isolated sphenoid sinus disease has a variable prognostic and pathological range yet has a common presentation. After adequate topical decongestion and anesthesia, endoscopy should always be conducted at the time of evaluating an isolated sphenoid sinus. CT scans and if needed, MRIs are not valuable in ruling-out or evaluating sphenoid sinus disease. In contrast, when every other cause of fever is ruled out, the otorhinolaryngologist physician is mandated to take a fluid sample and/or of tissue for microbiology.

Headache is declared to be what most of the patient present with

and CT scan is the modality to confirm the diagnosis.

The pathology ranges from bacterial sphenoidal sinusitis to an invasive carcinoma to the cavernous sinus. Endoscopic Navigation-Assisted wide sphenoidotomy is ideal for diagnosis and management.

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