

# Aspergillomas of the sphenoid sinus: A series of 10 cases treated by endoscopic sinus surgery

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## SUMMARY

*We report our experience with 10 cases of sphenoidal aspergillomas treated by endoscopic sinus surgery (ESS). Chronic symptoms such as cough, post-nasal discharge, dysphonia and even facial pain can be encountered in the history. Computerised tomography and, occasionally, magnetic resonance imaging are of great help in the assessment of this disease, especially when extensive skull base involvement is present. The radiological presentation can vary from an heterogeneous to homogeneous opacity with or without bone lysis to a frank pseudo-tumoural appearance. Four diagnostic tools have been evaluated to confirm the diagnosis: histology, direct smear, fungal cultures, and serology for Aspergillus. ESS has been successfully carried out without morbidity in all cases. No recurrence of the disease is seen after a mean follow-up of 27 months.*

*Key words: Aspergillosis, sphenoid, sinusitis, functional endoscopic sinus surgery, CT findings*

## INTRODUCTION

Aspergilloma is a non-invasive form of *Aspergillus* infection that may develop in the paranasal sinuses. The disease is being encountered more often during the last decade with the wider use of computerised tomography (CT) and endoscopic examination. Sinus aspergillosis was first described in immunosuppressed patients who presented with an invasive infection associated with mucosal invasion (Rowe-Jones et al., 1994). A review of the literature in 1979 reported only 103 cases (Jahrsdoerfer et al., 1979).

The increment in the number of diagnosed cases may be explained by the systematic analysis of all material taken from the sinuses during surgery. The fungus has a characteristic presentation at histological examination with numerous septate hyphae and dichotomous 45°-angle branchings (Brandtwein, 1993). We now have a 10-year experience of this disorder. *Aspergillus fumigatus* is the only cultured fungus in the present study. Four types of paranasal aspergillosis have been described (Hartwick and Batsakis, 1991). The fulminant and the indolent presentations show a mucosal invasion. This characteristic is not found in the other two forms, allergic aspergillosis and aspergilloma or "fungus ball".

We report 10 cases of sphenoidal aspergillomas managed by endoscopic sinus surgery (ESS), which to the best of our knowledge is the largest reported series in the literature.

## MATERIAL AND METHODS

A total of 10 patients with sphenoidal aspergillomas were treated by ESS in the last seven years. No case of immunosuppression was encountered. All patients had a diagnostic nasal endoscopy and CT scan. All specimens taken during surgery were analysed by four diagnostic tools: histological examination, direct smear, fungal culture, and serological studies for *Aspergillus*.

Patients were all closely followed post-operatively with serial endoscopic examinations and CT.

## CASE REPORTS

### *Cases 1-4: Heterogeneous presentations*

These patients (Table 1) had a pre-operative CT that showed a complete heterogeneous opacification of a sphenoid sinus with or without micro-calcifications. They presented chronic post-nasal drainage with nocturnal cough. Endoscopy revealed some oedema in the sphenoidal recess. An endoscopic sphenoidotomy allowed a complete removal of all fungal material

Table 1. Characteristics of 10 patients with sphenoidal aspergilloma.

sex/age (M-F/year)	localisation (paranasal)	endoscopic results in spheno-ethmoidal recess	CT-results	follow-up (months)
F/58	right, sphenoidal	oedema	heterogeneous, microcalcifications	35
M/69	right, sphenoidal	purulent discharge, oedema	heterogeneous, microcalcifications	28
F/44	right, sphenoidal	purulent discharge, oedema	heterogeneous, microcalcifications	35
M/47	left, sphenoidal	oedema	heterogeneous	14
F/39	left, spheno-ethmoidal	polyps, oedema, purulent discharge	homogeneous, pseudotumoural	12
F/53	right, sphenoidal	purulent discharge, oedema	homogeneous, pseudotumoural	44
F/37	bilateral, sphenoidal	oedema	homogeneous, pseudotumoural	22
F/25	left, pansinusal	polyps, oedema	heterogeneous, pseudotumoural	33
M/41	left, sphenoidal	purulent discharge, oedema	homogeneous,	40
F/48	left, sphenoidal	purulent discharge, oedema	homogeneous, bone condensation	10

within the sinus. After a minimum of 14 months of follow-up, all patients remained asymptomatic and no abnormality was seen at endoscopy.

#### Cases 5-8: Pseudotumoral presentations

A CT scan demonstrated in this group of patients an heterogeneous or homogeneous opacity associated with some bone lysis. Patients consulted for chronic posterior nasal discharge. The spheno-ethmoidal recess was hidden by few polyps or oedema. The CT findings first suggested the presence of a



Figure 1. Pre-operative CT in axial view showing a homogeneous opacification of the left sphenoid and posterior ethmoid associated with an extensive bone erosion (F/39). This CT presentation suggests the presence of a mucocele.



Figure 2. A pre-operative T<sub>2</sub>-weighted MRI in axial plane shows in the same patient (F/39) a hypo-intense signal within a hyper-intense zone, suggesting an aspergilloma in the left spheno-ethmoidal region.

oedema. The CT findings first suggested the presence of a mucocele (Figure 1). In contrast, magnetic resonance imaging (MRI) was more accurate and demonstrated the presence of an aspergilloma (Figure 2). A complete unilateral opacification of all paranasal sinuses was even observed in one case (Figure 3). A sphenoidotomy associated with a posterior ethmoidectomy,



Figure 3. Pre-operative axial CT (F/25) with a complete heterogeneous opacity of left paranasal sinuses. A diffuse bone lysis and architectural deformity are obvious.

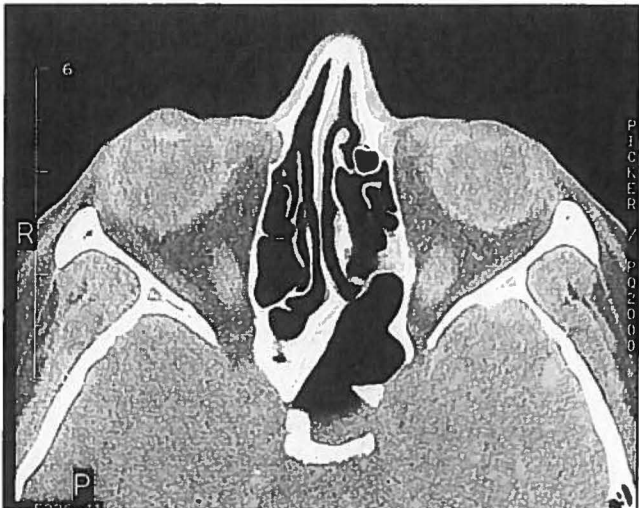


Figure 4. Axial view on CT performed 8 months after ESS for an aspergilloma of the left sphenoid-ethmoidal region (F/39). A clean and regular post-operative cavity is obtained.

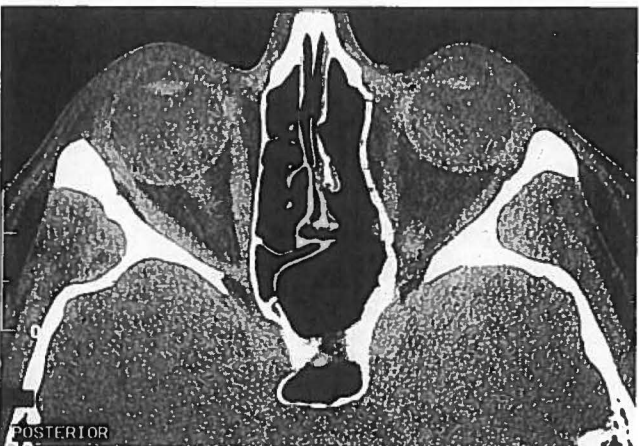


Figure 5. Post-operative CT in axial view (F/25). A clean and regular paranasal cavity is associated with some bone reconstruction.

or complete ethmoidectomy and middle antrostomy, was performed in the two cases with an extension outside the sphenoid sinus. An absence of recurrence was confirmed in all cases by both CT (Figures 4-5) and endoscopy during follow-up.

#### Cases 9-10: Homogeneous presentations

One patient consulted for chronic unilateral periorbital headaches associated with a left nasal discharge. Another presented an unexplained long-term dysphonia. Nasal endoscopy revealed in both patients an oedematous stenosis of the sphenoidal ostium with some muco-pus. CT confirmed the presence of an

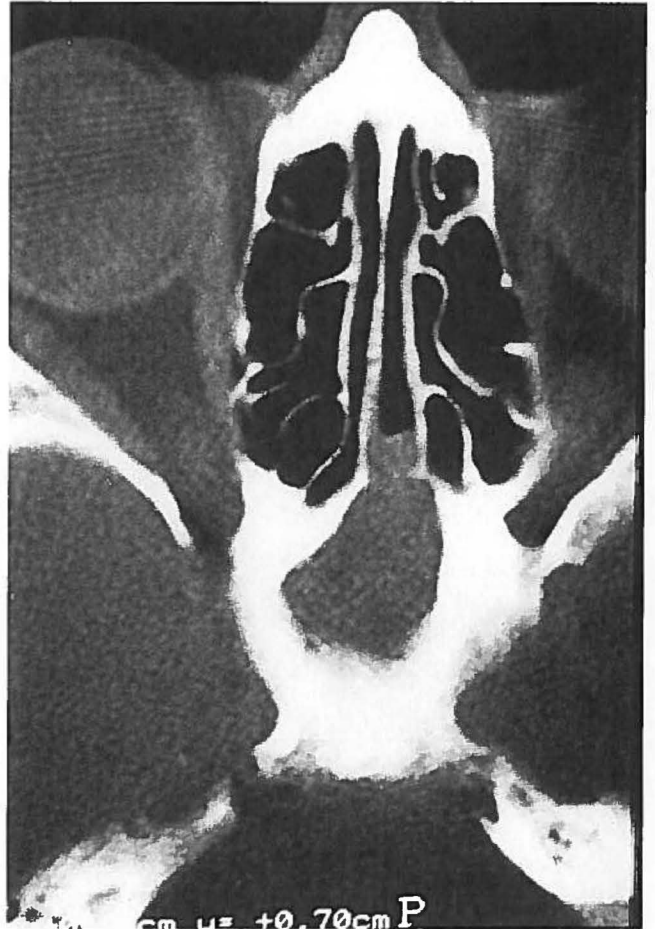


Figure 6. Pre-operative CT in axial view (F/48). An homogeneous opacification of the left sphenoid sinus is surrounded by an important bone condensation.

isolated homogeneous opacification within the sphenoid sinus. A remarkable bone condensation around the sphenoid sinus was even found in one case (Figure 6). A clean regular sphenoidal cavity was still encountered many months after sphenoidotomy.

#### RESULTS

Ten cases of sphenoidal aspergillomas are presented. Table 1 shows the different data on each studied patient.

Two-thirds of cases were females. The population was mostly between 40 and 60 years of age with a mean age of 46 year old. The most frequent presentation was solitary sphenoidal involvement (7/10). We also encountered a bilateral sphenoidal localisation, a sphenoid-ethmoidal presentation, and a unilateral

pansinus disease. Unilaterality was observed in nine patients. All cases had rhinological symptoms with or without headaches. Chronic posterior nasal drainage, unresponsive to medical therapy, and nocturnal chronic cough were the two major complaints. Unexplained chronic dysphonia was also found. A history of endodontic treatment with intracanal fillings were found in only two patients. Pre-operative endoscopy showed a regional inflammation in the sphenoid-ethmoidal recess of all cases, a discrete mucopurulent discharge in six patients, and some polyps in two others.

Pre-operative CT results correspond in Table 1 according to each patient. Aspergillomas appeared as heterogeneous opacities in five cases and as homogeneous opacities in five others. Three patients presented numerous microcalcifications within the opacities. A pseudotumoural appearance with some bone lysis was seen in four patients. Major bone condensation was observed in one case. Additional magnetic resonance imaging (MRI) was performed on two patients with an extensive skull-base bone lysis. On a T<sub>1</sub>-weighted MRI, it showed a nodular hypo-intense signal which could not be enhanced by gadolinium and was surrounded by a diffuse hypersignal. On a T<sub>2</sub>-

Table 2. Results of each of the four diagnostic tools.

sex/age	histology	direct smear	cultures	serology
F/58	-	+	-	-
M/69	+	+	+	-
F/44	+	+	+	-
M/47	-	+	-	-
F/39	+	+	-	-
F/53	+	+	-	-
F/37	+	+	-	-
F/25	+	+	+	-
M/41	+	-	-	-
F/48	+	-	-	-

weighted MRI, the aspergilloma was iso- or hypo-intense to the brain.

Four diagnostic tools were systematically used in each patient (Table 2). Histological examination revealed the presence of branching hyphae in eight out of 10 patients. Direct smear was also positive in eight cases. Fungal cultures were least sensitive with only three positive results. The only identified pathogen was *Aspergillus fumigatus*. Positive cultures were always associated with positive results on direct smear and histological examination. Precipitines for *Aspergillus fumigatus*, *A. flavus*, *A. niger* and *A. terreus* were measured by an immunodiffusion technique. Serology for *Aspergillus fumigatus* was also analysed by immuno-electrophoresis and with the "HAI test" with normal dilution values <1:320. No positive serological result was found. We have a mean follow-up of 27 months, ranging from 10 to 44 months. No recurrence of the disease was observed using ESS.

#### DISCUSSION

The slow development of aspergillomas associated with a non-specific symptomatology makes the diagnosis of sphenoidal aspergilloma very challenging. Aspergillomas of the paranasal

sinuses in immunocompetent hosts have been well documented in the Sudan or other Third-World countries (Saeed and Brookes, 1995). Rowe-Jones and Freedman (1994) reported only 15 cases of aspergillosis of the sphenoidal sinus in the English literature between 1979 and 1994. Few cases have been reported in the French literature during that period (Salvan et al., 1993). Advances in rhinology with the wider use of CT and nasal endoscopy have permitted earlier and more frequent diagnoses. In our personal experience, the sphenoid sinus is the second most frequently affected sinus after the maxillary sinus. Concerning the symptomatology, a discrete post-nasal discharge, a predominantly chronic nocturnal cough, non-specific headaches, and even unexplained dysphonia should alert the physician. These symptoms have also been reported by Miglets et al. (1978) and Simmons et al. (1982). A nasal endoscopy must be carried out in all patients presenting such symptoms to precisely assess the sphenoid-ethmoidal region for any mucopurulent discharge or polyps. These non-specific symptoms have led some authors to conclude that the diagnosis of paranasal aspergillosis can be confusing (Blitzer and Lawson, 1993).

Our patients with any history or endoscopy suggestive of fungal infection have a systematic CT to optimize the assessment. The importance of this diagnostic tool in fungal sinus disease has already been recognized in the literature (Zinreich et al., 1988; Stammberger et al., 1984). Affected sinuses are always subtotally or completely opacified. Occasional micro-calcifications were found in three cases.

Such micro-calcifications in an opacified maxillary sinus may correspond to some dental amalgam (Legent, 1989). This phenomenon may also be due to a fungal concretion with the deposition of calcium salts (Stammberger et al., 1984). The fact that only two out of our 10 patients have had a dental treatment history supports the second theory. Nevertheless, the pathophysiology of aspergillomas remains unknown. Most authors describe aspergillomas as an homogeneous opacity within the sinus cavity without bone lysis.

Jahsdoerfer et al. (1979) concluded that the distinction between the non-invasive and slowly invasive aspergillosis is not so clear. In our series, aspergilloma appeared as a pseudotumour on CT with apparent bony lysis in two out of seven patients with single sphenoidal involvement, and in two out of three cases with multi-sinus infection. Hartwick and Batsakis (1991) suggested the invasive potential of aspergillosis in chronic and indolent sinusitis as a possible mechanism of bone lysis. Expansion forces within the obstructed sinuses might be another possible explanation for the thinning and deformation of the sinus walls. MRI was systematically performed in cases with large skull base involvement (two cases). In fungal disease an iso- or hypo-intense signal is seen in T<sub>2</sub>-weighted MR images (Figure 2). Higher concentrations of iron and manganese in fungal concretions could also explain these findings (Zinreich et al., 1988). Pre-operative findings were always very characteristic. A green fungus ball was often seen. The typical glue filling the affected paranasal sinuses – and often discussed in the literature – was encountered in only two out of 10 patients. In these cases, serologies were negative.

Four diagnostic tools have been systematically used in our 10 patients: histology, direct smear, fungal cultures, and serologies for *Aspergillus* (Table 2). Direct smears were positive in eight patients, histology was positive in eight patients, and fungal cultures were positive in only three patients. All serologies were negative, excluding an allergic aspergillosis. These four diagnostic tests should be carried out in any suspected case of aspergilloma. A single positive result at either histology, direct smear or fungal culture in association with a negative serology and consistent macroscopical findings during ESS are required for the diagnosis of aspergilloma.

All cases were successfully managed by surgery. A wide sphenoidotomy was often performed under local anaesthesia (6/10). A posterior ethmoidectomy and a total ethmoidectomy were jointly performed in two patients. The initial configuration of the paranasal cavities was restored in the postoperative period of four of our cases. Exposed dura at the skull base was observed endoscopically to be slowly covered by a new lining of bone and epithelium (Figures 4-5). This observed regeneration of bone and normalisation of the anatomy after surgery confirm the reversibility of the bone erosion encountered with this pathology. This reversibility may be due to the effect of reducing inflammation on the balance between osteogenesis and osteolysis in the post-operative period. The epithelial lining normalized within six weeks after surgery, leading to a clean and regular paranasal cavity in eight weeks. The usefulness of ESS in the management of sphenoidal aspergillomas and the reversibility of the bone changes has been demonstrated.

#### CONCLUSION

We report 10 cases of sphenoidal aspergillomas. This disease has been considered very unusual before the wide use of endoscopy and CT in rhinology. A non-specific symptomatology leads to the diagnosis at history. Nasal endoscopy is characteristic with some oedema in the sphenoid-ethmoidal recess in all cases. On CT, various appearances can be found: heterogeneous or homogeneous sinus opacifications, subtotal or total sinus opacities with or without bone lysis. Aggressive characteristics suggestive of neoplasia can even be found with extended areas

of bone lysis at the skull base. Surgery using ESS has proven its reliability in all cases. A wide opening of the sphenoid sinus must be followed by a careful extraction of all fungal materials. Histology and direct smear are the two most useful diagnostic tools. Any patient with a large area of exposed dura must be followed closely after surgery. No recurrence of the disease under these conditions has been observed after a mean follow-up of 27 months.

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