The Need for Surgery of Isolated Sphenoid Sinus Disease Care Based on Imaging Findings

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Research Article

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Abstract

Background:

In general, ISSD may be detected due to clinical symptoms, but some ISSDs are found incidentally during radiological examinations for the assessment of unrelated diseases. This study aims to evaluate the clinical aspects of all ISSDs detected by image inspection.

Methods:

A total of 94 patients with ISSD were enrolled in this study. The study patients were classified into two groups: a group who improved with conservative treatment (n=25), and a group who did not improve with conservative treatment (n=69). Their clinical symptoms, medical histories, pathologies, and imaging findings were assessed.

Results:

The incidence of high signal intensity on T2-weighted images in the improvement group was significantly higher compared with the non-improvement group. Conversely, the incidences of bone erosion and expansion were significantly higher in the non-improvement group. Of the 42 cases receiving surgery, 10 cases (23.8%) had asymptomatic ISSD.

Conclusion:

This study suggests that ISSD with high signal intensity on T2-weighted images and/or without bone erosion and expansion in imaging findings could improve by conservative treatment or watchful waiting. Because some asymptomatic ISSD cases required surgery, proper diagnosis of ISSD regardless of clinical symptoms is important to prevent severe illness.

Introduction

Isolated sphenoid sinus disease (ISSD) is a relatively uncommon clinical condition among patients with paranasal sinus disease. As has been reported, ISSD is difficult to detect by conventional X-ray imaging; nonetheless, the number of patients with ISSD has been increasing because of the widespread availability and use of advanced imaging modalities including computed tomography (CT) [1–3]. Patients with ISSD may have various clinical symptoms including headache, nasal obstruction, and postnasal drip. Moreover, ISSD affecting adjacent anatomical regions may lead to severe complications such as visual loss and diplopia.

ISSD without clinical symptoms has been detected incidentally by imaging tests for other diseases or non-specific symptoms. Many studies have reported differential diagnoses and treatments of ISSD, but the majority of these studies focused on symptomatic ISSD requiring surgery. To the best of our knowledge, no study has evaluated all ISSDs, including cases without surgery. Among them, there are

cases improved by conservative treatment and asymptomatic cases detected incidentally. The aim of this study is to evaluate the clinical features, diagnostic results, treatments, and outcomes of all cases of ISSD.

Materials And Methods

Patients

A retrospective review of the Kochi Health Sciences Center's medical records from March 2003 to June 2018 was performed for the diagnosis of ISSD based on findings by CT scan and/or magnetic resonance imaging (MRI). Patients with concomitant involvement of other paranasal sinuses were excluded from this study. Also excluded were patients with nasopharyngeal neoplasm, head trauma, loss of consciousness, or tracheal intubation. Therefore, out of 151 patients diagnosed with ISSD, 55 patients who had neither surgery nor follow-up imaging tests and 2 patients with pituitary adenoma, who had bone erosion and underwent surgery by the neurosurgery department, were excluded. A total of 94 ISSD patients were enrolled in the study, which evaluated their clinical symptoms, medical histories, and imaging findings. We classified these ISSD patients into two groups: improvement group, a group who improved with conservative treatment; and non-improvement group, a group who did not improve with conservative treatment. According to the necessity of surgery, the non-improvement group was divided into a surgery group and a follow-up group without surgery.

The therapeutic principle for ISSD at our hospital is as follows. First of all, patients with cranial nerve neuropathy or suspicion of malignant tumor receive a recommendation for emergency surgery. Elective surgery is recommended for patients with suspicion of benign tumor, fungus ball, or mucocele with sinus wall erosion. Other patients receive conservative management, such as with antibiotics, or careful follow-up for 12 weeks or longer. After the conservative treatment, the patients with complete improvement are recommended to get regular follow-up, and the patients with residual ISSD without any clinical symptom and bone erosion are also recommended to get regular follow-up with imaging tests. Surgery is recommended to the symptomatic patients with residual ISSD after the conservative treatment. In the surgery group, the final diagnosis was based on histopathologic and microbiological examination of surgically resected specimens.

Clinical findings

We assessed clinical symptoms such as fever, headache, nasal obstruction, nasal discharge, visual loss, diplopia, and post-nasal drip. The asymptomatic ISSD cases were defined as patients without these symptoms. We examined the smoking status and the past history of immunosuppression, malignancy, and paranasal sinus surgery. The patients who had been smoking on their first visit to our hospital were defined as current smokers. The Brinkman index (number of cigarettes per day × smoking years) was calculated for former/current smokers. The patients who had diabetes mellitus and/or other immunodeficiency syndromes, and the patients with administration of immunosuppressant medication including oral steroids and antineoplastic drugs were considered to have immunosuppressive conditions.

Imaging findings

We evaluated findings such as bone erosion, sinus wall remodeling, bilaterality, expansion, complete opacification, and calcification on CT and/or high signal intensity on T2-weighted MRI images (Fig. 1). As for image inspections, 64 patients underwent both CT and MRI, while 17 patients underwent only CT, and 13 patients only MRI.

Statistical analysis

Values are presented as means ± standard deviation (SD). We used Fisher's exact test to compare the proportions of categorial variables among the groups. The comparisons of continuous variables among the groups used the Mann-Whitney U test. The threshold for significance was set at $p < 0.05$. All statistical analyses were performed with EZR (Saitama Medical Center, Jichi Medical University, Saitama, Japan), which is a graphical user interface for R (The R Foundation for Statistical Computing, Vienna, Austria). More precisely, it is a modified version of R Commander designed to add the statistical functions frequently used in biostatistics [4].

Results

Characteristics of subjects

Of the 94 patients with ISSD, 41 were female and 53 were male. The age at diagnosis ranged from 2 to 91 years (mean: 59.3 years old). As mentioned above, the patients were divided into two groups (improvement group, 25 patients; non-improvement group, 69 patients) (Figure 2). In the study period (March 2003 - June 2018), endoscopic sinus surgery was performed in 735 cases at our department. The incidence of endoscopic sinus surgery for ISSD was 5.7% (42/735). The characteristics of these patients are summarized in Table 1. Between the two groups, there are statistically significant differences in sex and age. A significant difference was also observed in incidence in the past history of immunosuppressive conditions and imaging findings in bone erosion, expansion, and high signal intensity on T2-weighted images. The improvement group had much a higher ratio of high signal intensity on T2-weighted images (17 out of 18 [94.4%]) than the 25.4% in the non-improvement group.

Clinical findings

Regarding clinical symptoms, there were no statistically significant differences between the two groups. The most common symptom was headache (35 patients [37.2%]). Twelve cases (12.8%) had cranial nerve neuropathy (visual loss and/or diplopia), three cases had both visual loss and diplopia, three cases had visual loss only, and six cases had diplopia only. In improvement group, one patient with diplopia refused to receive surgery and was treated with antibiotics (Table 2). The non-improvement group was separated into 42 patients in the surgery group and 27 patients in the follow-up group. The follow-up period in the follow-up group ranged from 3 to 149 months (mean ± SD: 50.5 ± 42.7 months). During the

period, none of the patients in the follow-up group exhibited other clinical symptoms or severe complications.

Relationship between pathological findings and clinical characteristics

The pathological diagnoses in the surgery group were obtained using surgical specimens. Table 2-4 shows the relationship between the pathological findings and clinical characteristics. In the surgery group, the diagnoses consist of 7 cases of sphenoiditis without nasal polyps (16.7%), 1 case of sphenoiditis with nasal polyps (2.4%), 11 cases of mucocele (26.1%), 12 cases of fungal rhinosinusitis (fungal ball, 8 cases, 19.0%; invasive fungal rhinosinusitis, 4 cases, 9.5%), 3 cases of allergic fungal rhinosinusitis (7.1%), and 3 cases of malignant tumor (7.1%). Headache, visual loss, and diplopia were frequently observed in the patients with invasive fungal rhinosinusitis or malignant tumor (Table 2). Sphenoiditis without nasal polyp or invasive fungal rhinosinusitis was frequently found in the patients with a past history of malignant tumors and/or immunosuppression (Table 3). As for imaging findings, the patients with mucocele, of which 6 out of 11 were asymptomatic, had high incidences of bone erosion (45.5%) and expansion (72.7%) (Table 4).

Asymptomatic ISSD

The 43 cases (45.7%) of asymptomatic ISSD are outlined in Figure 2. Asymptomatic ISSD was identified in 13 out of 25 cases (52.0%) in the improvement group, and 30 out of 69 cases (43.5%) in the non-improvement group. In the non-improvement group, the number of asymptomatic patients was small in the surgery group compared with the follow-up group. Of the 10 cases out of 42 cases (23.8%) in the surgery group, 6 cases were mucocele, and 1 case each of fungus ball, inverted papilloma, meningioma, and ectopic pituitary adenoma was also identified.

Discussion:

ISSD has various clinical symptoms and pathologies. Several review articles have reported on ISSD diagnosis and management [5–8]. Although ISSD usually can be detected based on clinical symptoms, patients with ISSD sometimes have no symptom. Fooanant et al. reported that 22.1% of ISSD (27 out of 122 patients) was detected incidentally [5]. Most studies discussed only surgical cases, and there were a few reports on asymptomatic cases or cases improved by conservative treatment [9, 10]. Even in the asymptomatic cases, because many kinds of diseases including malignant neoplasms are considered as a differential diagnosis for ISSD, a detailed interview and medical examination might be necessary for a definitive diagnosis. In this report, we evaluated all cases of ISSD in clinical symptoms, imaging findings, diagnostic results, and necessity of surgery.

The patients in the improvement group were considered as cases with acute sphenoiditis, although no pathological examination was done. In this study, we performed the conservative treatment for sphenoiditis if the patients did not have immunosuppression and cranial nerve neuropathy. Compared with the non-improvement group, the patients in the improvement group had high signal intensity on the

T2-weighted images, which means we could rule out a tumor and performed conservative treatment. Although previous reports recommended early surgical intervention for all patients with ISSD [11], some patients had the possibility to improve without surgery.

As for clinical symptoms, several studies reported that headache was the most common presenting symptom. The pooled rate of headache in ISSD was 71.4%, while the pooled rate of cranial nerve neuropathy was 16.3%. Among all the neuropathies, visual loss and diplopia were most commonly observed, followed by trigeminal neuropathy [7]. As for cases of malignant tumor, previous papers reported that 50-53.8% of them had cranial nerve neuropathy, 66–75% had headache, and 6.7% were asymptomatic [2, 12]. In this study, all three malignant tumor patients had either cranial nerve neuropathy (visual loss: 1 case; diplopia: 3 cases) or headache (2 cases). Although the malignant tumors in the study were detected in the advanced stage, the case of inverted papilloma was incidentally detected in an early stage as asymptomatic ISSD. The most common malignant tumor in the sphenoid sinus is squamous cell carcinoma (29.4%), and inverted papilloma have a 10% risk of malignant transformation to squamous cell carcinoma [13]. Because the prognosis for cranial nerve neuropathy associated with malignant tumor is very poor, when a tumor without clinical symptoms is suspected in the sphenoid sinus, surgery should be performed for early diagnosis and treatment before it causes severe complications such as cranial nerve neuropathy [14].

Moss et al. [7] reported 1,133 ISSD pathologies classified as sphenoiditis without nasal polyp (28.3%), mucocele (20.3%), fungal sinusitis (12.5%), malignant neoplasms (7.7%), intracranial lesions (7.0%), benign neoplasms (5.7%), sphenoiditis with nasal polyp (3.4%), and other lesions (4.7%). In our study, the pathologies in the surgery group were similar to those of previous reports. The most common pathology of asymptomatic ISSD in the surgery group was mucocele, which was 6 out of 11 (54.5%) cases, and the patients received surgery to prevent enlargement or infection of the mucocele. The etiology of the mucocele remains unclear. One theory regarding the cause of the mucocele is mechanical obstruction of the sinus, and 7 out of 11 (63.6%) patients with mucocele had a past history of paranasal sinus surgery, which could have caused a mechanical obstruction. Mucocele can progress slowly and asymptomatically until bone erosion or compression affects adjacent structures [15, 16].

Detection of ISSD is difficult by conventional X-ray examination, but recent advances in the availability and accuracy of radiological studies have made it much easier to detect ISSD. A tumor diagnosed by incidental findings in an asymptomatic or symptomatic patient during imaging for unrelated reasons is called incidentaloma. Though there were no studies about incidentaloma in the paranasal sinus, lesions detected by incidental imaging findings including incidentaloma are known as a modern medical crisis [17]. Sieskiewicz et al. reported that 46.8% of ISSD was detected by non-otorhinolaryngological departments, and delayed diagnosis was a problem because the clinical symptoms and signs of ISSD were usually rare and non-specific [18]. Ashida et al. reported ISSD was often asymptomatic, however, many cases were not referred otorhinolaryngological departments [19]. Radiological imaging is a great help to diagnosis, but the use of one modality alone—either CT or MRI—could lead to a missed diagnosis

[1, 12]. Therefore, we had to generally refer to imaging findings and additional examinations such as an endoscopic examination, blood test, and bacterial culture test comprehensively.

Study limitation

A limitation of our analyses is that we did not diagnose the follow-up group because they had neither surgery nor improvement from imaging findings. Almost all studies did not report a follow-up group without surgery because they did not diagnose any from pathological findings. None of our reported cases in the follow-up group showed worse clinical symptoms or complications of ISSD during the follow-up period. Because the follow-up group had much a higher rate of asymptomatic ISSD (20 out of 27 [74.1%]) than the other groups, they tended to reject the surgery in the diagnosis. Although we followed a watch-and-wait approach for these ISSD patients who rejected surgery, the validity of surgery for the follow-up group should be re-evaluated from a long-term perspective with consideration of systematic conditions and imaging findings.

Conclusion:

Our analysis suggests that ISSD frequently has temporal and asymptomatic cases without surgery. Any time that ISSD is detected, it has to be carefully diagnosed. This study suggests that the patients with only high signal intensity on T2-weighted images could improve by conservative treatment or watchful waiting. However, some asymptomatic ISSDs such as incidentaloma required surgery. Proper diagnosis of ISSD with or without clinical symptoms is important to prevent severe illness.

Abbreviations

ISSD: isolated sphenoid sinus disease, CT: computed tomography, MRI: magnetic resonance imaging, SD: standard deviation

Declarations

Ethics approval and consent to participate: This study was approved by the Institutional Review Board of the Kochi Health Sciences Center (IRB approval number: 181096) and conducted in compliance with the Declaration of Helsinki 2013.

Consent for publication: Not applicable.

Availability of data and materials: Not applicable.

Competing interests: The authors declare that they have no conflict of interest.

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Informed consent: Informed consent was obtained in the form of opt-out on the website.

Authors' contributions: KU, HT, SM, TO and SK: concept and design; KU, KK and AD: acquisition of data; KU, HT, SM, TO and SK: data interpretation; KU, HT, KK, AD, SM, YN, TO and SK: manuscript drafting and approval. All authors read and approved the final manuscript.

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Tables

Due to technical limitations, table 1-4 is only available as a download in the Supplemental Files section.

Figures

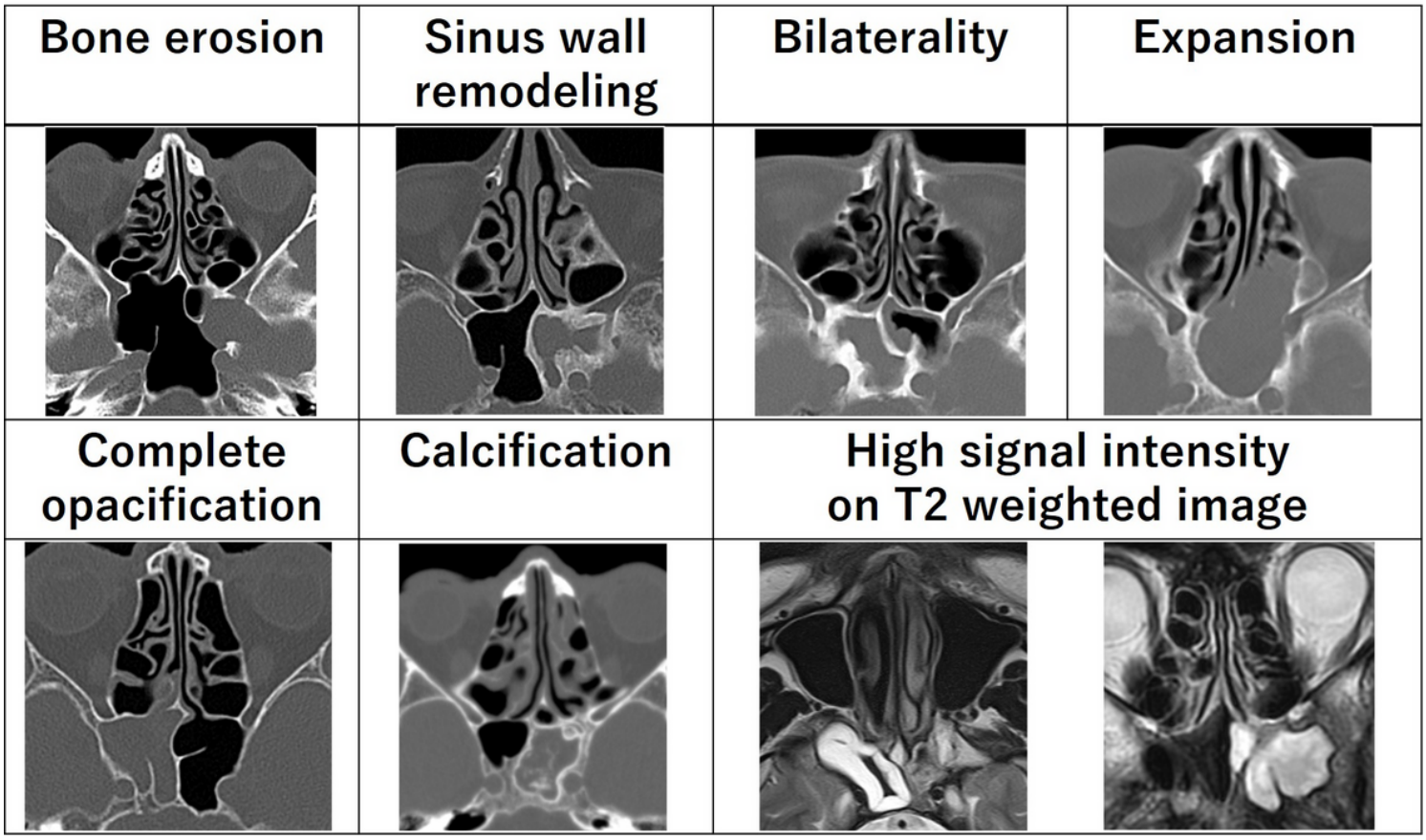

Figure 1

Imaging findings of isolated sphenoid sinus disease.

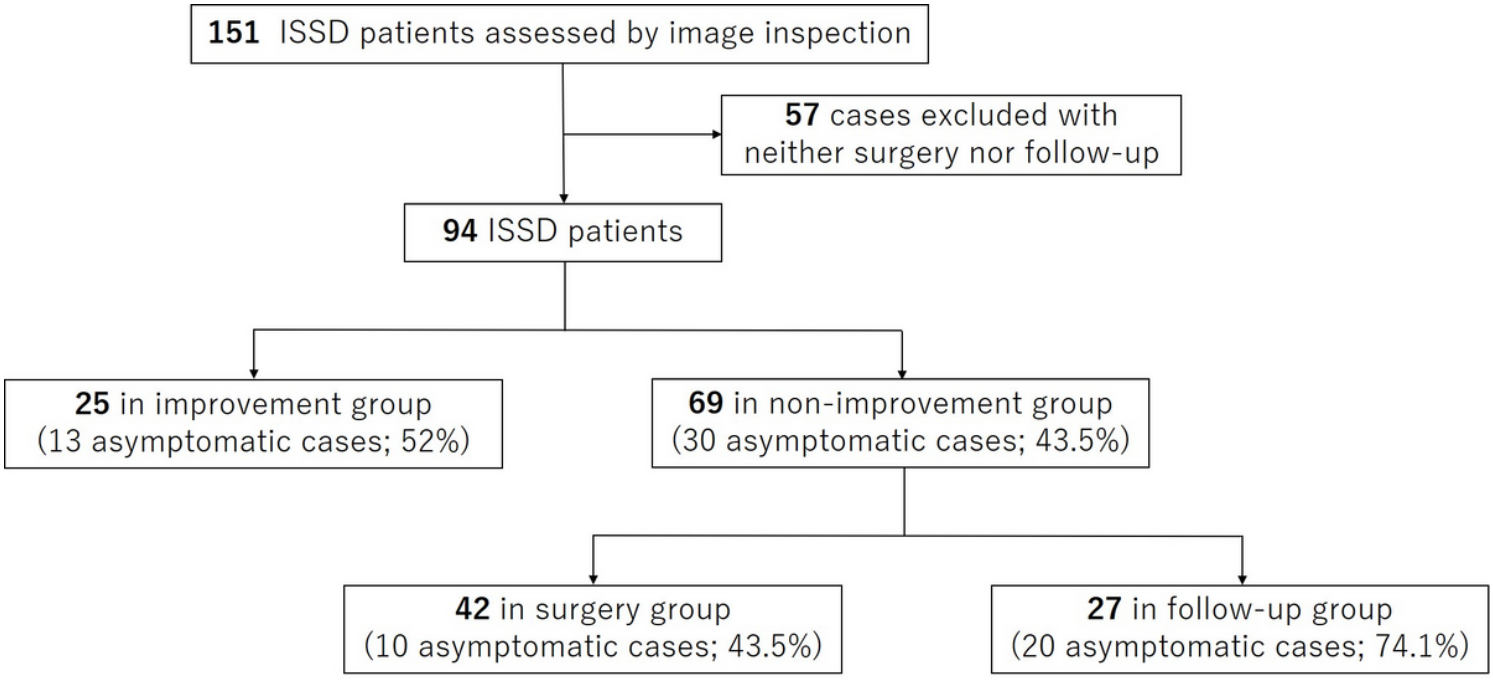

Figure 2

Flow diagram of included studies. (ISSD; isolated sphenoid sinus disease)

Supplementary Files

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- Table1.pdf
- Table2.pdf
- Table3.pdf
- Table4.pdf