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CASE REPORT Open Access

Case report of spontaneous sino-cutaneous fistula in chronic rhinosinusitis

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Abstract

Background Chronic rhinosinusitis (CRS) is a common disease characterized by inflammation of the paranasal sinuses, with symptoms such as nasal blockage, facial pain, hyposmia, and headache. While these symptoms are often treated as a common cold, the disease can lead to serious complications if left untreated. We present an unusual case of CRS in which the patient presented to the ophthalmology department with persistent pus discharge from the left upper eyelid for 6 months, despite being treated with multiple courses of antibiotics and undergoing incision and drainage procedures.

Case presentation A 69-year-old male patient presented with a 6-month history of persistent pus discharge from the left upper eyelid, despite being treated with multiple courses of oral antibiotics and three incision and drainage procedures. He also complained of yellowish nasal discharge and bilateral nasal blockage for the past year but did not seek medical attention for these symptoms. Physical examination revealed pus discharge from the left upper eyelid and widening of the nasomaxillary groove. Nasoendoscopic examination showed bilateral grade 3 nasal polyps with mucopus secretion. CT scan of the paranasal sinuses revealed that the odd presentation was due to the chronic inflammatory process in the paranasal sinuses, which had led to osteitis and hyperostosis. This patient underwent endoscopic sinus surgery (ESS) with left frontal trephination and fat obliteration of the left frontal sinus. The postoperative histopathological report confirmed the diagnosis of benign inflammatory nasal polyp with acute on chronic inflammation. The patient was asymptomatic at 3-month follow-up post-operation.

Conclusion Our case report highlights the importance of considering CRS as a possible underlying cause of such unusual presentations, even when the symptoms may appear unrelated. Early diagnosis and treatment of CRS can prevent serious complications and improve patient outcomes. The inflammatory process of CRS leading to osteitis and hyperostosis of the paranasal sinuses complicates the clinical presentation, distorts the usual anatomical structures, and may complicate the operation. CT scan of the paranasal sinus is highly recommended before any sinus surgery. At times, ESS may not be sufficient to eradicate frontal sinus disease. In these cases, an external approach is necessary with or without obliteration of the sinus cavity.

Keywords Chronic rhinosinusitis, Hyperostosis, Osteitis, Nasal polyps

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Background

Chronic rhinosinusitis (CRS) is a common inflammatory condition of the paranasal sinuses that lasted for more than 3 months and affects approximately 11% of the adult population worldwide. It is more prevalent in Europe, USA, and Brazil with a rate of 5–15%. On the other hand, in the Asia region such as Korea, China, and Singapore, the prevalence rate is reported as 7%, 8%, and 2.7%, respectively [1, 2].

Despite its high prevalence, CRS is often misdiagnosed and undertreated as a common cold, leading to significant morbidity and reduced quality of life for patients. In some cases, CRS can also lead to serious complications such as meningitis, brain abscess, and periorbital abscess, due to the close anatomical proximity of the sinuses to these structures.

These complications are related to the inflammatory process of CRS, leading to osteitis of paranasal sinuses, and eventually causing bony erosion, and sometimes later in the disease phase, the bone undergoes thickening or hyperostosis [3–5].

In this case report, we present an unusual presentation of CRS where a patient presented with persistent pus discharge from the left upper eyelid for 6 months. The patient had previously been treated with multiple courses of oral antibiotics and incision and drainage procedures without significant improvement. The ultimate diagnosis was made through imaging studies, which revealed hyperostosis and osteitis of the paranasal sinuses.

This case underscores the importance of considering CRS as a potential diagnosis in patients with unusual presentations of persistent ocular symptoms. A high index of suspicion, along with proper history taking, endoscopic examination, and appropriate diagnostic evaluation, can lead to timely diagnosis and treatment, potentially preventing serious complications.

Case presentation

A 69-year-old male patient who has no known medical illnesses presented to the ophthalmology clinic with a 6-month history of persistent pus discharge from the left upper eyelid, despite being treated with multiple courses of oral antibiotics and three incision and drainage procedures. He also complained of yellowish nasal discharge and bilateral nasal blockage for the past year but did not seek medical attention for these symptoms.

Physical examination revealed pus discharge from the left upper eyelid and widening of the nasomaxillary groove. Nasoendoscopic examination showed bilateral grade 3 nasal polyps with mucopus secretion. A biopsy of the bilateral nasal polyp was taken under local anesthesia and reported as inflammatory polyp.

CT scan of the paranasal sinuses showed soft tissue thickening of the left upper eyelid, communicating with the left frontal sinus (Fig. 1), soft tissue densities in all paranasal sinuses, with sclerotic and thickened bony walls of maxillary, ethmoid, and sphenoid sinuses bilaterally (Figs. 2 and 3). The scan also revealed erosion of the left frontal, right ethmoid, and right sphenoid sinus walls with communication with left frontal meninges, prepontine cistern, and both orbits.

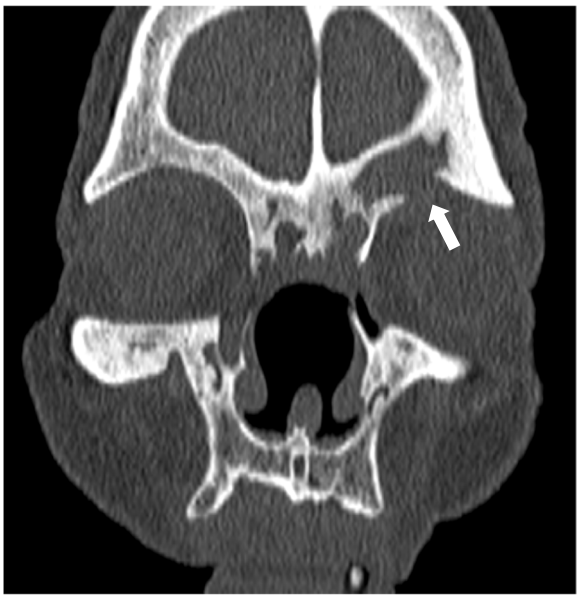

Fig. 1 Coronal view CT scan showing soft tissue thickening of the left upper eyelid, communicating with the left frontal sinus (white arrow)

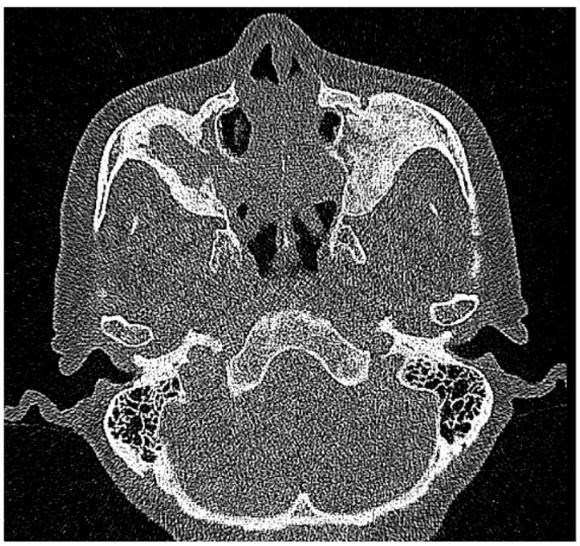

Fig. 2 Axial view CT scan showing sclerotic and thickened bony walls of bilateral maxillary sinuses

Given the suspicion of malignancy, an image-guided system (IGS) biopsy was performed via endoscopic sinus surgery (ESS) to confirm the diagnosis. However, bilateral maxillary, anterior, and posterior ethmoid air cells were sclerotic and had to be drilled for access to the respective sinuses. Left frontal trephination had to be performed due to hyperostosis of the paranasal sinuses resulting in total occlusion of the left frontal

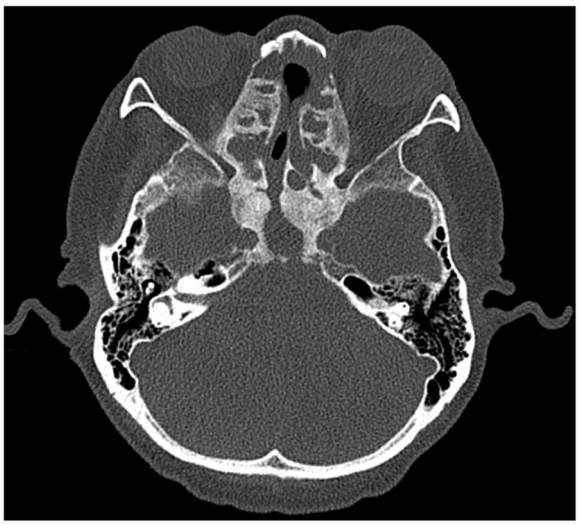

Fig. 3 Axial view CT scan showing sclerotic and thickened bony walls of ethmoid and sphenoid sinuses

sinus recess. The left frontal sinus was then obliterated with a fat graft.

The postoperative histopathological report confirmed the diagnosis of benign inflammatory nasal polyp with acute chronic inflammation. The patient was asymptomatic at 3-month follow-up post-operation.

Discussion

CRS is a common benign disease; however, the chronic inflammatory process of CRS causes bony erosion, and hyperostosis of paranasal sinuses is still not well understood. Studies have shown that not only CRS is associated with mucosal inflammatory changes but also the inflammatory process of CRS can reach the bone and lead to osteitis, which causes rarefaction and/or demineralization of bone, loss of trabeculae, cortical destruction, focal sclerosis, and loss of anatomical landmarks [3, 5].

The bone undergoes reabsorption or perforation during the early stage of CRS, but during the later phase of the disease, the bone will undergo thickening or hyperostosis [4]. A study by Kim H. Y. [6] shows a significant increase in the bony thickness of paranasal sinuses and a reduction of maxillary sinus volume in CRS pediatric patients.

Most patients with underlying CRS with nasal polyps will present with bony erosion, rarely present as hyperostosis of the bone [7]. In our case, the patient with long-standing neglected CRS presented with persistent pus discharge from the left upper eyelid due to ongoing osteitis and hyperostosis of all paranasal sinuses. The inflammatory process in the early stages of CRS leads to bone resorption and erosion of the orbital wall and anterior table of the left frontal sinus. In the later stage of the disease, hyperostosis develops, obliterates the left frontal ostium, and alters the normal paranasal sinus drainage system. As the inflammation continues and the mucopus secretion accumulates in a confined paranasal sinus, pressure increases in the confined left frontal sinus, causing exertion towards the eroded bony defect and eventually breaching the weakest point. In this case, it is the anterior table of the left frontal sinus. Patient with such sinocutaneous fistula usually has a history of frontal sinus fracture or frontal sinus surgery. Spontaneous sinocutaneous fistula due to CRS is very rare [8].

The CT scan of the paranasal sinusis a mandatory diagnostic tool before endoscopic surgery especially when there is evidence of complications such as in this case, fearing that the area may have bony changes and loss of anatomical landmarks, whereas magnetic resonance imaging (MRI) serves as the gold standard in diagnosing intracranial complications for its superior ability to demonstrate soft tissue abnormalities [9]. Besides CT scan and MRI, IGS will assist the surgeons to identify the distorted anatomical structures and assure that all the critical structures are in real time intraoperatively.

Usually, frontal sinus diseases can be successfully managed endoscopically. However, endonasal frontal sinus surgery may not be successful if the disease cannot be reached or long-term stable drainage is not established. In these cases, an external approach is necessary. Osteoplastic flap procedure with fat obliteration has been hailed as the definitive frontal sinus procedure for these cases [10].

Common indications for the frontal osteoplastic flap with sinus obliteration include chronic frontal sinusitis after endonasal surgery, frontal sinocutaneous fistula, frontal sinus fractures involving the frontal sinus drainage pathways, and osteomas [8]. The technique of frontal sinus obliteration with fat harvested from the abdomen dates back to the 1950s as proposed by Bergara [11] and Tato et al. [12].

Rainer Weber [10] also described the technique of osteoplastic frontal sinus surgery published by Weber et al. [13] It is essential to remove the frontal sinus mucosa completely by curette or drill to reduce the risk of mucocele [8, 9], followed by obliteration of the frontonasal duct with cartilage and a layer of temporalis fascia, galea periosteum, and fixation with fibrin glue. Lastly, obliterate the sinus cavity with harvested abdominal fat and covered with a bony flap. Among the 75 primary cases, the success rate of the surgery was more than 90% where seven cases needed revision and 9.8% developed mucocele [9]. Besides that, Abla Eledeissi proposed a similar surgical technique for treating traumatic frontal sinus fracture [14].

Several materials are recommended for frontal sinus obliteration, such as autogenous grafts like bone, fat,

temporalis fascia, or muscle and alloplastic materials like hydroxyapatite bone cement, methyl methacrylate, calcium phosphate bone cement, and glass ionomer. Overall, autogenous abdominal fat is the most well-studied and recommended by most surgeons [14].

Several studies have shown a high success rate and low risk of complications for frontal sinus surgery with fat obliteration. Zonis et al. reported a failure rate of 3% among 100 cases operated on with the Montgomery technique [15]. Calcaterra and Strahan reported that 23 of 24 patients with fat obliteration had complete eradication of the disease [16].

The most comprehensive series was that of Hardy and Montgomery, in which 250 patients were investigated, with a median follow-up of 8 years. Of 208 patients with obliteration of the frontal sinus, only 6% of cases had to be revised due to acute wound infection or recurrent chronic infection. There was no report on the occurrence of mucoceles [17].

Despite the high success rate and low risk of complications, long-term follow-up is still advocated for these patients to monitor the occurrence of persistent headaches, recurrent disease, and mucocele.

Conclusion

CRS symptoms may present similar to the common cold. However, it should not be taken lightly as CRS may lead to serious and even fatal complications.

The inflammatory process of CRS leading to osteitis and hyperostosis of the paranasal sinuses complicates the clinical presentation, distorts the usual anatomical structures, and may complicate the operation. CT scan of the paranasal sinus is a helpful diagnostic tool, and it is highly recommended before any sinus surgery.

At times, ESS may not be sufficient to eradicate frontal sinus disease. In these cases, an external approach is necessary with or without obliteration of the sinus cavity.

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Authors' contributions
IP and LY performed the surgery. SY collected medical history and drafted the paper. RR performed study design and article redaction. All authors have read and approved the final manuscript.

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Declarations

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Not applicable

Consent for publication
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Competing interests
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