Sino-orbital cutaneous fistula with endoscopy guided technique: A case series

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1. Introduction

Fistula formation in areas around the orbit is often associated with sinuses. Orbital fistula (a channel from sinus, orbit, to the outer skin) can occur following surgeries for malignant tumors of the orbit or paranasal sinuses, also due to trauma or radiotherapy. It may develop when the bony orbital and nasal walls are resected or penetrated to achieve local tumor clearance, because the malignant tumors can cause thinning and bony erosion. Fistula may also occur due to infection or the surgery itself. Incidence of fistula following orbital exenteration in cancer patients was ranged from 5.4 to 23% [1,2]. A study in Nigeria showed that 2 out of 228 chronic sinusitis patients (0.88%) developed frontal cutaneous fistula [3]. Aggressive local therapy of the nose and paranasal sinuses is essential as they lie adjacent to vital structures, orbit, and cranial cavity. A patient generally comes with a complaint of discharge from the fistula. Managing the orbital fistula with reconstruction is crucial; there are various methods, such as primary closure, split skin, full-thickness grafts, local, distant, and free flaps. In this study, the authors presented sino-orbital fistula cases with various etiologies, such as mucocele, chronic inflammation, and destruction caused by the tumor. The most commonly affected sinus is the frontal (60–89%). Fistula can occur with or without orbital/ nasal wall destruction and bony erosion. Before starting the treatment, it is essential to make a precise diagnosis of the etiology and rule out the possibility of recurrence. SOCF can be treated with conservative or invasive management, depending on the severity of the fistula.

Clinical discussion: Risk factors of developing SOCF are poor surgical technique, post-operative radiotherapy, concomitant immunocompromised state, diabetes mellitus, hypoproteinemia, or destruction caused by the tumor. It is essential to perform a thorough diagnostic examination with radiographic imaging to determine the specific cause before deciding on definitive treatment. By using the endoscopy-guided technique, long-term favorable results can be achieved. Multidisciplinary collaborative teamwork is needed to have a successful result.
2. Case presentation

2.1. Case 1

Female, 37 years old, came to outpatient clinic at our hospital with a complaint of continuous serous discharge from the skin defect under the eyebrow of the left eye. Two years prior, the patient had a history of frontal sinus mucocele and underwent transnasal endoscopic marsupialization to manage the mucocele. She was diagnosed with a sino-orbital cutaneous fistula (SOCF) of the left eye (Fig. 1A).

On ophthalmologic examination, there was a 15-mm diameter fistula at the central part of the left upper eyelid, beneath the superior orbital rim. Orbital CT Scan showed an expansive lytic lesion of the left frontal bone with a defect in the left maxillary sinus ostium (Fig. 1B, C).

She underwent joint surgery by oculoplastic and rhinology surgeons. During surgery, an incision was made around the fistula, followed by undermining the fistula canal until the end of the fistula (fistulectomy). Subsequently, the endoscopic endonasal surgery was initiated by removing the thickened bone narrowing the ostium. Then the procedure continued with removing pathologic mucosal tissues and preserving the healthy parts. Reconstruction using an abdominal composite full-thickness skin and fat graft was performed with naso-endoscopy guidance to see graft position in the frontal sinus. The wound was closed with polypropylene 6.0 sutures (Fig. 2).

After surgery, the patient was given oral and eye ointment antibiotics for 2 weeks, saline nasal irrigation, and intranasal steroid 200 μg twice a day for 12 weeks, along with regular postoperative care. The defect was successfully repaired, and no discharge was seen from the reconstructed orbital fistula. During follow-up at our outpatient clinic more than one year after surgery, the patient showed satisfaction, no complaint of fistula recurrence, and the wound healed well (Fig. 3).

2.2. Case 2

The male patient, 38 years old, came with a fistula at the nasal side of the right upper eyelid with recurrent mucoid-mucopurulent discharge (Fig. 4). The patient had a history of asthma and allergic rhinitis. Three times of surgical interventions still failed to manage the prolonged discharge from the fistula.

From the ophthalmologic examination, there was a 2-mm diameter fistula at the superomedial area of the right upper eyelid with mucoid discharge. Massages at the nasolacrimal sac were performed, resulting in no discharge. Anel's test (irrigation tests at the upper and lower puncta) resulted in no obstruction at the lacrimal ducts, and no leakage was seen from the fistula. Eye movement, visual acuity, anterior and posterior segment examinations were within normal limits. It was concluded that there was no connection between the fistula and the lacrimal excretory system; thus, the DCG examination was not performed. Before the patient underwent the surgery by oculoplastic and rhinology surgeons, a paranasal sinus CT scan was performed, showing opacification in the ethmoidal and frontal sinuses. The naso-endoscopic surgery we performed did not confirm a direct connection between the sinus and the orbit. Instead, we found severe inflamed residual cells of the frontal sinus that may extend to the orbital fistula. Another mandatory endoscopic sinus surgery was performed to prevent recurring inflammation.

An incision was made around the fistula as the first step of the surgery. The probe was used as a guide to follow from the proximal part of the cutaneous to the distal part of the tract. Next, the wound was reconstructed directly. After more than one year of follow-up, the patient still felt satisfied, no complaint of mass or discharge recurrence, and the wound healed well.

2.3. Case 3

Male, 39 years old, came with a complaint of discharge from left nasal superior orbit. The patient, who had a history of complete remission nasopharyngeal carcinoma, had been treated with chemotheraphy and radiotherapy one year prior. There was no family history of malignancy. Three months after remission, he presented with a lump at the superior left eyelid, proptosis, and double vision. From the ophthalmologic examination, there was a non-axial left eye proptosis due to superior nasal solid mass, pushing the eyeball downward to inferolateral and restricting eye movement to all directions (Fig. 5A). Anterior and posterior segment examinations were within normal limits. The right eye was within the normal limit. CT scan examination showed a 3-cm diameter solid isodense mass with contrast enhancement at the anteromedial orbit of the left eye expanding to ethmoidal sinus, pushing the eyeball and medial rectus muscle to lateral. Bone erosion was found at the medial wall, left preauricular lymphadenopathy, and pansinusitis.

The patient underwent anterior orbitotomy, ethmoidectomy, and a debulking tumor surgery. The surgery also included the removal of the adjacent periostium at the tumor bed. Histopathology resulted in squamous cell carcinoma (SCC) being poorly differentiated. One month post-operative, he was given 4 cycles of adjuvant chemotherapy. Ptosis and restriction of eye movement to all directions were noted. Three months after surgery, he experienced recurrent redness, swelling, and discharge at the scar tissue which resolved with oral antibiotics. CT scan examination showed no receding lesion at the tumor bed (medial extraconal left orbit) and pansinusitis (ethmoid and maxillary sinuses) with left medial rectus muscle and left inferior oblique muscle enlargement. The patient was diagnosed with a sino-orbital cutaneous fistula of the left upper eyelid post-SCC tumor excision (Fig. 5B-D).

An orbital fistula reconstruction was performed. Firstly, the superonasal sulcus fistula was identified beneath the superior orbital rim at
Fig. 2. Intraoperative pictures. (A) Frontal mucosal sinus from endonasal view showing the diameter of frontal ostium more than 8 mm to prevent re-stenosis. (B) Incision was made around the fistula followed by fistulectomy. (C) Dermofat graft taken from the abdomen was excised to fit the fistula wall. (D) The result was fixated to the tissue surrounding it using polypropylene 6.0 sutures.

Fig. 3. One year post-operative follow-up showed good wound healing.

Fig. 4. (A) Abscess of the medial canthus. (B) Purulent discharge from the abscess. (C) Abscess resolved, showing superomedial orbit fistula punctum.
the upper eyelid of the left eye. An incision was made around the fistula, then the fistula canal was undermined until reaching the end. Endonasal endoscopic sinus surgery was performed to remove residual cells, fibrotic tissue, and polypoid mucosa, then it was visualized that the medial wall of the orbit only covered by mucoperiosteum resulted from the previous SCC orbital tumor excision that had included the medial, oblique, and superior rectus muscle, and lamina papyracea. A probe inserted from the proximal end of the canal fistula reached the distal end in the frontal sinus. The reconstruction procedure was done using an abdominal composite full-thickness skin and fat graft which was positioned in the frontal sinus, under nasoendoscopy guidance (Fig. 6). The cutaneous wound was closed with polypropylene 6.0 sutures. This is our latest case, on 6 months of follow-up, patient showed no recurring fistula and the wound healed well. The patient hoped that the eyelid could be opened and the eye movement could improve.

3. Discussion

The various factors responsible for developing a naso-orbital cutaneous fistula (following surgery of this region) include poor surgical technique, post-operative radiotherapy, concomitant immunocompromised state, diabetes mellitus, hypoproteinemia, or a destruction caused by the tumor. The fistulas in this case series were formed by inadequate management from previous surgeries, severe inflammation of chronic rhinosinusitis associated with allergic rhinitis and asthma, and the removal of the previous orbital tumor.

In the first case, mucocele occurred because of stenosis due to fibrosis from previous frontal sinus surgeries and subsequently forming a fistula due to bony erosion and severe mucosal, orbital, and cutaneous inflammation. Mucoceles in paranasal sinuses had a space-occupying quality that might cause bone erosion and displacement of surrounding structures [5]. The frontal sinus is most commonly involved (60–89%), followed by the ethmoid (8–30%) and maxillary (<5%) sinus [6]. Bony erosion at the floor of the frontal sinus adjacent to the superior orbital wall may cause early displacement of the orbit in this case. While in the second case, recurrent sinusitis hematogenically extended to the skin, caused the fistula without either orbital/nasal wall destruction or orbital displacement.

The SOCF of the third case was caused either by bone erosion due to the tumor or by the tumor debulking surgery that created a large area of fibrotic tissues. The patient had a history of squamous cell carcinoma (SCC) at the left retrobulbar area, invading the ethmoidal sinus as the second primary tumor. The first primary tumor was nasopharyngeal carcinoma (NPC). In NPC patients, there was a 24% increased risk of developing second primary cancer compared with the general population. The most common second primary cancers in NPC are: oral/pharyngeal, salivary gland, sarcoma, skin, and leukemia/lymphoma [7,8]. Before starting the treatment, it is important to rule out the recurrence of the malignancy especially if the fistula has occurred late. The average interval between the first NPC and second primary cancer is 5.33 ± 4.68 years [7]. In this case, the patient had a second primary in 3 months after remission of the first primary.

Conservative management is preferred for a small asymptomatic fistula, such as maintaining local hygiene or direct closure [9]. A small fistula with bone loss may require grafts for appropriate repair. If the fistula diameter is >5 mm, it may trigger disturbing symptoms, like foul-smelling and crusting [10,11]. Surgery was indicated in the first and third cases because of its large size. In the second case, due to repeated failure episodes of conservative management with antibiotics, we performed reconstruction surgery with direct closure technique from the distal to the proximal of the fistula. In the first and second cases, although patients had undergone external approach reconstruction surgery, recurrent discharge still occurred; thus, nasoendoscopic approach was preferred.
Reconstruction surgery was chosen as the definitive treatment of superior palpebra fistula, using composite full-thickness skin and fat graft which includes the epidermis, full-thickness dermis, and subcutaneous fat to prevent traction on the orbital rim in the first case. This grafting method is chosen over local flap, split-thickness skin graft (STSG), or full-thickness skin graft (FTSG) because of the large size of the fistula (15-mm diameter) and fewer complications, such as contour deformities of surrounding tissues, scarring, or the possibility of volume depletion. The fat part of the graft provides better structural support and greater volume than FTSG. Other studies used autologous fat, hydroxyapatite cement, muscle, bone, or bioactive glass to obliterate the frontal sinus [12]. The frontal stenosis in the first case is resolved by maintaining the patency of ostium in 8-mm diameter with intensive post-operative care. Similar orbital and sinus surgery procedures were performed in the third case of the tumor patient. Most importantly, watchful observation has to be conducted for early diagnosis of recurring tumors.

SOCF diagnosis is concluded in the second case, even though there was a limitation in observing the distal end of the fistula in the ethmoidal and frontal sinus cavity due to its pinpoint size. Therefore, the fistulectomy and the wound closure were more straightforward to perform. Endonasal endoscopy-guided approach shows better results than external approach for patients with sinonasal disease-related orbital problems. It is functional and minimally invasive, with precise removal of diseased tissue, less scarring, and faster healing. The post-operative care is dedicated to having physiologic paranasal sinus mucosa in 6-month follow-up. SOCF recurrence can be prevented by the healing of paranasal and sinus mucosa integrity.

Because the third case is the latest case we observed, we could not have a longer period of follow-up. The patient was observed for 6 months, but we recommend minimum of 5 years follow-up to observe recurrence and prognosis.

4. Conclusion

In diagnosing SOCF, imaging modality should be performed to assess the anatomical state of the orbital and surrounding structures. Although, endonasal endoscopy-guided technique provides a better depiction of existing connection between nasal cavity and orbit. Composite full-thickness skin and fat graft is beneficial for large orbital fistula. Fistulectomy with direct closure is recommended for a small orbital fistula. Paranasal sinus post-operative care is mandatory to prevent the recurrence of SOCF. However, longer follow-up period and larger sample size are required to validate our suggestion. A five-year duration of follow-up should be considered for malignancy-associated SOCF. Collaboration between rhinology and oculoplastic surgeons is beneficial to adequately manage recurrent SOCF cases, to ensure the canal between sinonasal and orbital cavity can be closed completely.

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Ethical approval

N/A. This study is exempt from ethical approval from the institution.
Patient consent

Consent to publish personal information for publication or scientific purposes have been obtained from the patients orally and written. This case series does not contain any personal information that could lead to the identification of the patient. A copy of the written consent is available for review by the Editor-in-Chief of this journal on request.

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Declaration of competing interest

N/A.

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