Facial Translocation Approach for Management of Invasive Sinonasal Aspergillosis

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Abstract Aspergillosis is an uncommon fungal disease seen commonly in paranasal sinuses. It may be of noninvasive variety which is restricted to paranasal sinuses seen in healthy and immunocompetent hosts or may present as invasive form seen in immunocompromised hosts especially in diabetics and patients under long term immunosuppressant therapy. Along with paranasal sinuses invasive forms may encroach upon surrounding adjacent structures involving the orbit and extend intracranially. Orbital and intracranial involvement have a high mortality rate. Early diagnosis of the lesion and prompt aggressive management is important for better prognosis. Aggressive treatment includes surgical debulking and medical management mainly with systemic amphotericin B and other azoles. In orbito-cranial involvement, due to limited access to cranial base, it becomes difficult to surgically debulk the lesion with safety to surrounding important structures. Facial translocation approach gives adequate access to paranasal sinuses, orbit and anterior cranial base. Reported here are two cases of invasive cranio-orbito-nasal aspergillosis treated with facial translocation approach.

Keywords Fungal sinusitis · Invasive Aspergillosis · Facial Translocation · Orbito-zygomatomo-maxillary bone flap

Introduction

Aspergillus, is a fungal species of class Ascomycetes present ubiquitous. They are most common causative agents of fungal

sinusitis with *Aspergillus fumigatus* being most commonly implicated [1]. Invasive aspergillosis is an opportunistic infection occurring commonly in immunocompromised individuals. The prognosis worsens with the intracranial extension, may result in fatal outcome [2]. Therefore early diagnosis and aggressive therapy is important.

Facial translocation, a concept that originated from facial compartment disassembly, is a valuable approach to the skull base. They can be divided as “mini”, “standard”, “expanded” (vertically, medially, posteriorly), and “bilateral” facial translocation procedures [3].

We report two cases of patients with invasive aspergillosis of maxillary sinus and orbit treated with facial translocation approach. Importance of early diagnosis and aggressive treatment of invasive aspergillosis is also discussed.

Case 1

A 30-year-old male patient with 8 month history of pain and swelling on right side of the face and retro-orbital region reported to the Department of Oral and Maxillofacial Surgery in our institution. The patient's history revealed that he had been suffering from pain since last 8 months for which he reported to a regional hospital. He also noticed swelling which was gradually increasing. There, the lesion was biopsied and the report was confirmatory of a tubercular lesion of right maxillary sinus. The patient was started with the anti-tubercular regimen which continued for 6–7 months but he was not completely relieved of the symptoms. Then he reported to our institution.

On admission, he complained of severe throbbing pain in right maxillary sinus and retro-orbital region, radiating towards the temporal region on the same side. Diffused swelling on right side was also present extending supero-

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Fig. 1 Preoperative photograph showing swelling on right side and proptosis of right eye

inferiorly from the infraorbital region to the right angle of the mouth region which was tender on palpation. Clinical examination showed decreased visual acuity with proptosis of the right eye and numbness on right side of the face and palatal mucosa (Fig. 1).

On medical examination, he was diagnosed with hepatitis which may be drug induced with very high levels of SGOT and SGPT. He also had high blood sugar levels, but there was no previous history of diabetes. CT scan revealed a large heterogeneously enhancing mass lesion, invading the right maxillary sinus, eroding its medial and lateral walls, encroaching into subcutaneous soft tissue planes anterior to the right infratemporal fossa and extending up to right pterygoid plate and retro-orbital planes engulfing the optic nerve (Fig. 2a, b).

Medical treatment for hepatitis and diabetes was started and a biopsy of the lesion was performed through the right side maxillary sinus. Microscopic examination of the specimen showed a fibrous connective tissue stroma with heavy inflammatory cell infiltrate and focal abscess formation, wherein *Aspergillus* hyphae colonies were formed (Fig. 3).

After control of the blood sugar level, the patient was undertaken for surgery. Debridement and debulking of the lesion using facial translocation approach was planned, followed by long term course of anti-fungal drugs.

Standard facial translocation approach was used with modification of the standard incision. In standard facial translocation approach, superior incision continues from the nose to the inferior fornix of the lower eyelid and again through the lateral canthus horizontally to the preauricular area. We used, Weber–Ferguson incision starting with the midline split of the upper lip to the base of the columella and then deviated to follow the nasal contour of the medial canthus on the right side with an infraorbital extension of the incision from the medial canthus to expose the maxilla

Fig. 2 **a** CT scan demonstrating a large heterogeneously enhancing mass lesion, invading the right maxillary sinus, eroding its medial and lateral walls and extending up to right pterygoid plate. **b** Extension of the lesion in retro-orbital planes engulfing the optic nerve leading to proptosis

Fig. 3 Histologic section confirmatory of Aspergillosis

(Fig. 4). There was a fibrous, brownish-red growth eroding the anterior maxillary sinus wall which was debrided. Orbito-zygomatico-maxillary bone flap was temporarily

Fig. 4 Weber–Ferguson incision on right side

Fig. 5 Orbito-zygomatico-maxillary bone flap was removed temporarily for access to anterior cranial base

removed for access (Fig. 5). The surgical debridement of right maxillary sinus, medial wall of nose and retro-orbital region was done taking care not to damage the optic nerve (Fig. 6). Then, the orbito-zygomatico-maxillary segment was plated back (Fig. 7). Orbital floor and anterior wall of maxillary sinus defect were reconstructed using a 1.5 mm titanium mesh (Fig. 8).

Postoperatively, Amphotericin B with a dose of 1–1.5 mg/kg was administered intravenously on alternate days. The blood counts and serum creatinine levels were monitored every week and accordingly the dose was titrated by the physician. This was continued for 2 months. The patient was relieved of the symptoms with normal visual acuity and decreased pain (Fig. 9). But again after 15 days, the patient complained of pain in right temporal region. As the patient was of poor economic status, combination therapy (Amphotericin B+ Itraconazole) could not be administered and moreover looking at the severity of

Fig. 6 Debridement of the fungal mass with preservation of optic nerve

Fig. 7 Orbito-zygomatico-maxillary bone flap plated back

the disease we had to continue with the same regimen but we used the liposomal Amphotericin B variant this time as it has very less side effects compared to the standard amphotericin B deoxycholate. The patient again reported to our institution after 4 months in an unconscious state with GCS score of 3. CT scan of the head demonstrated extension of the lesion into the brain leading to aspergillosis induced encephalitis. Voriconazole (200 mg BD) was started and the patient showed some improvement with GCS score of 6. His condition remained steady for 15 days and then continued to deteriorate. The patient died 1 month later.

Fig. 8 Reconstruction of the floor of orbit and anterior wall of maxillary sinus

Fig. 9 Postoperative photograph after 2 months

Case 2

A 42 year old male patient reported to our department with the chief complaint of pain in right eye and headache since 2 months. In the beginning, the pain was dull and intermittent in nature which was relieved on taking medication. Gradually the pain became severe and continuous radiating towards the frontal and right temporal region. There was decreased visual acuity with restriction of the eye movements especially in the medial gaze of the right eye with exophthalmos and numbness on right side of the face.

Fig. 10 T1 weighted MRI showing hypointense soft tissue lesion involving the right maxillary sinus, nasal cavity and the orbit extending up to the frontal lobe

Medical history of the patient revealed that he was diabetic with high levels of blood sugar (fasting and post prandial). T1 weighted axial and coronal MRI examination showed an expansile soft tissue lesion involving the right maxillary, ethmoid and frontal sinuses invading the retro-orbital and medial orbital planes engulfing the optic nerve, inferior rectus, medial rectus and superior oblique muscles extending up to the inferior segment of frontal lobe. Erosion of the right inferior and middle turbinates with extension of the lesion into the right nasal cavity was also seen (Fig. 10). Biopsy was performed through the maxillary sinus which was confirmatory of aspergillosis.

The standard facial translocation approach was performed using the Weber–Ferguson incision, wherein the ipsilateral facial skin was displaced laterally and inferiorly along with the underlying maxilla, creating midline split of the hard palate (Fig. 11). Thorough debridement of the fungal growth was done and the orbito-zygomatico-maxillary bone flap along with the hard palate was plated back (Fig. 12).

The patient was administered liposomal Amphotericin B 1–1.5 mg/kg on alternate days and depending on the relief in symptoms reported by the patient and postoperative radiographic picture, the same treatment was continued for 4–6 months with monitoring of the blood counts and serum creatinine levels weekly after which the dose was titrated by the physician. The postoperative course was satisfactory with decrease in pain. The patient is fine and under follow-up.

Fig. 11 Standard facial translocation approach with midline split of the palate

Fig. 12 After debridement, retracted bone flap plated back

Discussion

With the improved aids, the diagnosis of paranasal sinus fungal diseases has increased in recent years. *Aspergillus*, a saprophytic mould, is a thermo tolerant fungi which grows in 15–53 °C range suitable for human environment. Out of 19 species of *Aspergillus* species, most common is *Aspergillus fumigatus* followed by *A. flavus*, *A. niger*, *A. terreus* [4]. Microscopically *Aspergillus* is a filamentous fungi 2–4 mm in diameter and septate hyphae. It can be distinguished from *Mucor*, which forms broader nonseptate hyphae and identified by dichotomous branching at 90° angle [5].

Infections of *aspergillus* include a wide range of clinical manifestations including allergic sinusitis seen in non

invasive variety to a life threatening acute invasive disease. Most common mode of inoculation of spores is via inhalation. Many reports suggest presence of *aspergillus* in apparently healthy individuals. This inoculation can present itself as respiratory tract disease involving tracheo-bronchial infections or as a sinonasal infection. There have been other sites which may be primarily involved including ear infections, disseminated aspergillosis involving CNS, ocular infections and bone infections [4].

Depending on host response and severity of infections, Aspergillosis may present itself as a noninvasive or an invasive form. Non invasive type of sinonasal aspergillosis is seen most commonly in immunocompetent individuals. It may be due to allergy causing localized fungal mycetoma or aspergilloma or “fungus ball” [6].

Invasive type of sinonasal aspergillosis is seen in immunocompromised patients usually associated with destruction of surrounding structures. Depending on amount of invasion it can be sinonasal, orbitonasal, and cranio-orbito nasal aspergillosis [6]. Invasive aspergillosis causes rapid spread and destruction via vascular invasion with coagulation necrosis of vessels causing mycotic thrombosis. This vascular invasion can lead to orbital or intracranial extension. This angioinvasive property results in more tissue destruction, beyond the appreciated area of clinical or radiographic extension [7].

When it invades orbit it causes proptosis, chemosis, ophthalmoplegia, retroorbital pain, decreased or no vision depending on optic nerve involvement as it causes interruption of blood supply to sensory nerve with extensive bony destruction [2]. Intracranial extension involvement may be with frontal bone via frontal sinuses, cavernous sinus, carotid artery or into anterior or middle cranial fossa via cerebral artery [7]. More the involvement of surrounding structure poorer is the prognosis. Intracranial involvement reduces survival with mortality of 40–80 % and is usually fatal [8].

Success of treatment of aspergillosis majorly includes two factors. Firstly assessment of patient’s immune status and secondly, the time of diagnosis. Status of host immunity is considered equally important to type of treatment therapy chosen. With immunocompromised status it shows more mortality rates. Also status of any underlying disease accounts for success of treatment. Remission of underlying disease shows a better prognosis while the relapse may increase the mortality. Hence, immunologic stimulation has been attempted by use of Granulocyte Macrophage stimulating factor to raise neutrophil counts and host resistance [7].

Delayed diagnosis usually results in delayed treatment in indolent stages or sometimes untreatable condition. This is usually due to lack of host inflammatory response towards the disease. The most important strategy is

improving the patient's immune status along with management of fungal disease [7].

Management of invasive aspergillosis is a combination of surgical debridement and chemotherapy. Extensive surgical debridement is essential as invasive form may involve paranasal sinuses, lateral nasal walls, orbits, or may extend intracranially. The debulking of the fungal lesion also improves response of medical treatment [2]. Various surgical approaches most commonly used in literature include Caldwell Luc procedure and Endoscopic Sinus Surgeries [9]. In this report, we suggest a facial translocation approach for improved access. The basic principle of facial translocation approach is to temporarily remove the facial bone unit and to reimplant and fix it after the tumor is resected. Specifically, in facial translocation approach, the facial osteotomies can be electively carried out according to the anatomic location of the tumor. In addition to tumors of the skull base, this technique has been found useful in the management of tumors of the paranasal sinuses and orbit [10]. As the floor of orbit was already eroded, this approach aided in approaching the posterior orbit inferiorly with preservation of surrounding structures. Along with debulking, medicinal treatment includes use of polyenes namely Amphotericin B as the first line drug of choice. Also combination of Amphotericin B with Itraconazole has shown a synergistic effect. A newer Azole agent, Variconazole has improved results.

To conclude, in cases of invasive aspergillosis early diagnosis and aggressive treatment i.e. extensive surgical debulking of lesion combined with an antifungal therapy may improve the prognosis. Also for such an extensive lesion, facial translocation approach proves to be beneficial, as it provides good surgical access because inappropriate surgical access may result in difficulty in

debridement and a risk of damage to surrounding vital structures. However, in cases of intracranial extension, the prognosis is poor and the result maybe fatal as seen in the first case.

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