

Batwing incision for a case of bilateral fungal zygomatic osteomyelitis

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SUMMARY

Zygomatic osteomyelitis is a rare occurrence due to rich collateral blood supply of bone. A man in his 30s presented with complaints of pain over bilateral cheek and pus discharge below the eye on lateral aspect. He was a known case of COVID-19 associated mucormycosis postendoscopic debridement of sinuses 3 months back. Radiology revealed bilateral destruction of zygoma with discharging sinus. Microbiological analysis confirmed aseptate hyphae in pus, and a diagnosis of bilateral fungal zygomatic osteomyelitis made. Under general anaesthesia, sequestrectomy done using bilateral lateral rhinotomy with extended Dieffenbach's approach (batwing incision). Postsurgery 3000 mg of liposomal amphotericin was administered. There was no enophthalmos or restricted eye movements postoperatively. Follow-up MRI suggested minimal inflammatory enhancement in maxillary sinus. Patient was discharged on oral antifungals.

BACKGROUND

COVID-19 associated mucormycosis (CAM) is on a rise in tropical countries such as India. Even after adequate surgical debridement and amphotericin administration, there have been a number of cases presenting with osteomyelitis of facial bones. We describe a case of chronic suppurative fungal osteomyelitis due to mucor involving bilateral zygomatic bones. On review of literature, there have been some case reports on unilateral maxillary and zygomatic bone osteomyelitis due to trauma, bacterial and fungal causes. This is the first case of bilateral zygomatic osteomyelitis reported in English literature with its treatment options. A bilateral limited lateral rhinotomy with extended Dieffenbach's incision was given for exposure of bilateral zygoma and its frontal process for desired osteotomies and debridement. This incision has been termed as batwing incision as such modification of facial incision has not been used before.

CASE PRESENTATION

A man in his 30s presented to otolaryngology outpatient department of AIIMS with history of bilateral facial pain and black lesion with pus discharge below the lateral part of both eyes for 1 month. The patient is a known case of CAM postendoscopic sinus debridement followed by oral posaconazole administration in another hospital 3 months back. There was no history of any ophthalmological or neurological complaints. On examination, the swelling over the malar prominence was mildly indurated and green-coloured purulent pus could



Figure 1 Markings of incision, a bilateral-limited lateral rhinotomy with extended Dieffenbach's approach (batwing incision). Arrow showing inclusion of fistulous site in incision.

be expressed from right side ([figure 1](#)). Diagnostic nasal endoscopy revealed bilateral healthy nasal cavity with patent sinuses postendoscopic debridement status. On microbiological analysis, fungal stain showed presence of aseptate hyphae. On contrast enhanced CT, bony erosions were present in bilateral right posterolateral wall of maxilla, zygomatic arch, roots and its frontal process with discharging sinus on right side ([figure 2](#)). A diagnosis of bilateral fungal zygomatic osteomyelitis was made. Patient underwent bilateral zygomatic sequestrectomy via external approach. After tarsorrhaphy, a bilateral lateral rhinotomy with Dieffenbach's lateral extension to lower eyelid incision (batwing incision) made with inclusion of diseased skin ([figure 1](#)). Intraoperatively, zygomatic bone was necrosed with discharging pus and surrounding

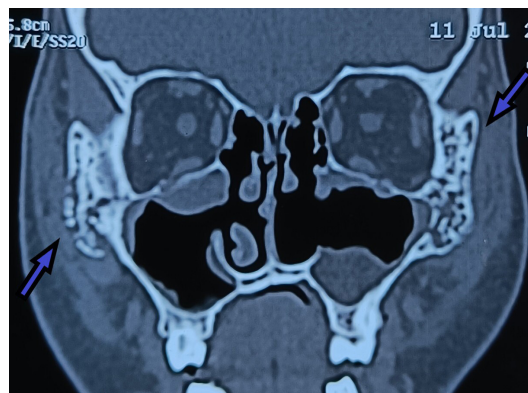


Figure 2 Contrast enhanced CT of face in coronal section showing bony erosions in bilateral zygoma extending superiorly till frontal process (marked arrow) and medially involving posterolateral wall of maxilla.



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Figure 3 Depicting bilateral cheek flap elevation and exposure of desired segments of zygoma and maxilla.

unhealthy soft tissue. Skin flaps were elevated using diathermy (figure 3). Right zygoma was completely exposed and osteotomies were given at its temporal process, superior end of frontal process and lateral to inferior orbital foramen at zygomatic arch using reciprocating saw. Lateral entry made into maxillary sinus and inspected for any residual necrotic tissue (figure 4). After local debridement, temporalis muscle was approximated to infraorbital rim to prevent enophthalmos. Similar steps repeated on left side. After achieving complete haemostasis, wound closed in two layers. Postoperatively, patient was given intravenous antibiotics and pain killers for 3 days. Intravenous liposomal amphotericin was also given up to 3 g in next 2 weeks of ward stay. There were no visual complaints and sutures were removed on postoperative day 7.

INVESTIGATIONS

Preoperative blood investigations show HbA1C: 5.2, total leucocyte count: 0.00598/L, serum ferritin: 332 ng/mL, IL-6: 15 pg/mL, D-dimer: 255 ng/mL, CRP: 28.6 mg/L, fasting blood sugar levels: 90 mg/dL. Above investigations signifies mucormycosis developing in a non-diabetic man. Raised serum markers shows COVID-19 recovery.

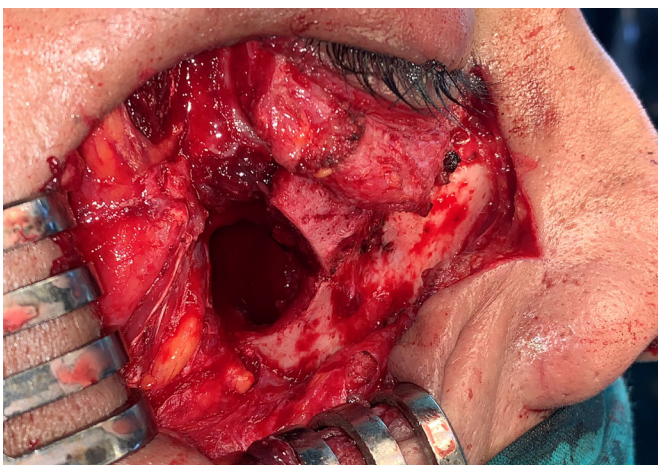


Figure 4 Intraoperative exposure after removal of zygoma and posterolateral wall of right maxilla. Temporalis muscle fibres were rotated infraorbitally and sutured to form a sling and prevent enophthalmos.

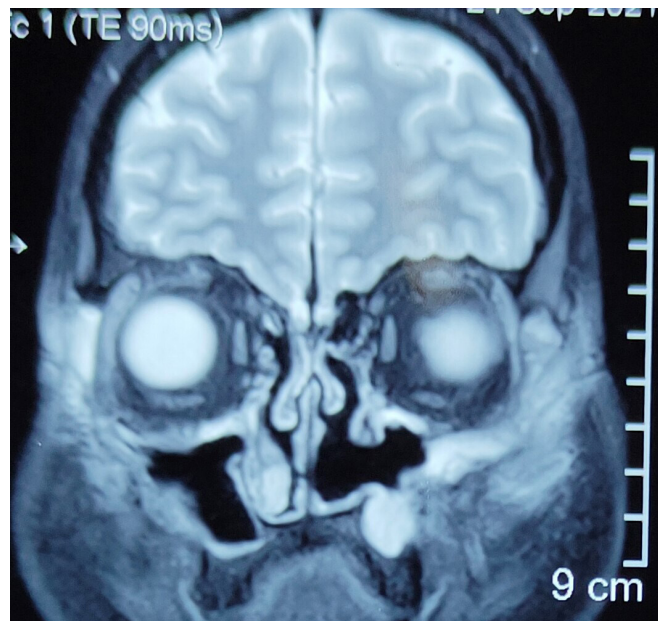


Figure 5 Contrast enhanced MRI of face in axial section showing minimal mucosal enhancement in maxillary sinuses with no evident residual disease.

OUTCOME AND FOLLOW-UP

Contrast enhanced MRI was done after 2 weeks which showed mild heterogenous enhancement in posterior wall of bilateral maxillary sinus, suggestive of mucoid secretions and no residual disease (figure 5). There were no restriction in eye movements or enophthalmos. Patient was discharged in healthy condition. Patient was followed up at 3 months and had no fresh complaints or signs suggestive of residual disease (figure 6).

DISCUSSION

Osteomyelitis in zygoma and maxilla is a rare occurrence as compared with mandible due to rich vascularity and thinner cortices that promote rapid healing.¹ Characteristic features include bone destruction with sequestrum formation, formation of extraoral sinuses and facial disfigurement. We presented a case of chronic suppurative osteomyelitis of bilateral zygoma which was fungal in origin. Susceptible aetiologies that result in this process are comorbidities such as diabetes, immunosuppressive therapy, malignancies, chemo and radiotherapy.² The only immunosuppressive aetiology in our case was COVID-19 infection and its treatment with systemic steroids which resulted in CAM.³ The fungus penetrates the arterial walls producing thrombosis and infarction of tissues.⁴ Early debridement and antifungals are the mainstay of treatment in such cases. Our case was one of the rarest in presentation as it affected bilateral



Figure 6 Post-operative picture of healed scar after 2 weeks.

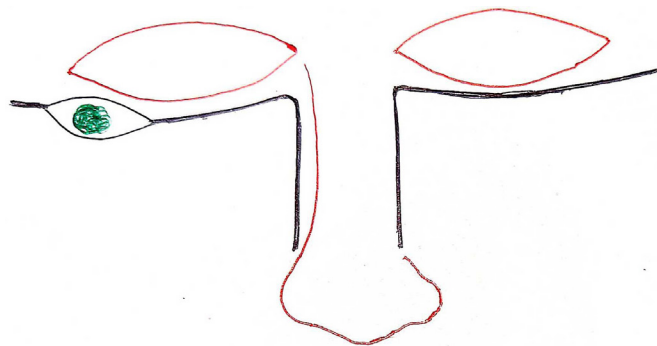


Figure 7 A line diagram depicting markings of batwing incision (drawn by SY).

Patient's perspective

After recovering from COVID-19, I developed a fungal disease of the sinuses for which I underwent surgery in a nearby hospital. But some days after surgery I started having pus discharge below my eye on both sides, for which I was referred to AIIMS-Delhi. Here, after some investigations, doctors diagnosed it to be the same fungal infection and it had already damaged both my cheek bones. I consented to surgery for the same, and afterwards I received amphotericin. Now since last 1 month I am fine with no difficulties in vision or any other nasal symptoms. Doctors have advised me for a regular follow-up.

Learning points

- ▶ Osteomyelitis of bilateral zygoma is very rare and differential must include a sinonasal disease or diseases that spread with haematogenous route.
- ▶ For diagnosis, a concurrent clinical, radiological and microbiological analysis is required.
- ▶ Mucormycosis generally results in bone infarction due to direct penetration of arterial walls and subsequently producing thrombosis.
- ▶ Treatment in such cases is emergency surgical debridement and amphotericin administration.
- ▶ Batwing incision is another name for bilateral lateral rhinotomy with extended lateral infraorbital Dieffenbach's incision.
- ▶ Long-term follow-up with radiology and clinical examination is required at frequent intervals.

zygoma. Review of literature had such case reports of only unilateral involvement of maxilla and zygoma. Routine surgical management in such cases is the removal of necrotic bone with sequestrum followed by debridement of surrounding tissues.⁵ A similar treatment protocol was followed using a batwing incision (bilateral-limited lateral rhinotomy with lateral extension of Dieffenbach's incision) (figure 7). Other approaches described in literature were Weber-Fergusson and sublabial incisions.⁶ In our case, as the disease involved the frontal process of maxilla, an external approach rather than sublabial approach was preferred.⁷ These cases should have a long follow-up with multidisciplinary management postoperatively, specially with ophthalmology, endocrinology, nephrology and haematology.⁷ Though loss of bilateral zygomatic bone causes aesthetic handicap of facial features for the patient, it can be minimised by creating a sound incision over face and preventing enophthalmos using temporalis muscle flap.⁶

Contributors SY is the assisting surgeon, has drafted the case report and followed up the case in ward. Line diagram is made by SY. HV is the corresponding author and primary surgeon in the treatment of the patient. AshS and AbhS helped in the review of literature, taking pictures of patient, taking consent and postoperative follow-up.

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Case reports provide a valuable learning resource for the scientific community and can indicate areas of interest for future research. They should not be used in isolation to guide treatment choices or public health policy.

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