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CLINICAL PAPER

Zygomaticomaxillary Osteomyelitis due to COVID-19 Associated Mucormycosis (CAM): A Case Series of 10 Patients

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Abstract

Aim To highlight the incidence of osteomyelitis due to CAM and to elucidate the mode of spread of infection from maxilla to zygomatic bone, to highlight how that is distinct from other cases of zygomatic osteomyelitis due to other etiologies.

Methods A standard protocol of treatment of the cases of CAM with zygomatic involvement based on our own outcomes was furnished. All 10 patients were treated with dual antifungal therapy and aggressive surgical resection via extraoral approach, in conjunction with functional endoscopic sinus surgery (FESS).

Results Ten out of 116 patients of CAM reporting to our institute presented with zygomatic bone involvement with an incidence rate averaging at 8.6%, whereas in previous literature osteomyelitis of zygomatic bone was extremely rare with an incidence pattern of just 1.42%.

Conclusions The treatment protocol followed by the authors gave good outcomes to all patients treated, with no mortalities.

Keywords COVID-19-Associated Mucormycosis (CAM) · Zygomatic osteomyelitis · Rhinomaxillary mucormycosis · Amphotericin B · Posaconazole · Total maxillectomy · Hemi-maxillectomy · Weber Ferguson incision

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Introduction

The second wave of the coronavirus disease 2019 caused by the novel Coronavirus or the SARS COV 2 has led to an increase in superinfections which prior to the beginning of the current pandemic were rarely ever reported. There is particularly a surge in cases of COVID-Associated Mucormycosis (CAM) which in common parlance has been termed 'The black fungus.' Mucormycosis which in prior times used to be a rare fungal infection is becoming rampant in many regions of India with the state of Maharashtra being one of the worst affected. Yet another curious finding is the involvement of the zygomatic bone in mucormycosis cases which have been reported to our center in these past few months. The maxilla is the most common jaw bone being affected by fungal osteomyelitis and is more commonly associated with diabetes mellitus. Clinically, mucormycosis occurs in one of the four forms: rhinocerebral, pulmonary, gastrointestinal and disseminated. Rhinocerebral form is the most

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common, representing one-third to one-half of all cases of zygomycosis. This is further subdivided into two subtypes: a highly fatal rhino-orbito-cerebral form which is invasive and may involve the ophthalmic and internal carotid arteries and a less fatal rhino-maxillary form which involves the sphenopalatine and greater palatine arteries, resulting in thrombosis of the turbinate and necrosis of the palate [1]. The clinical hallmark of mucormycosis is vascular invasion resulting in thrombosis and tissue infarction/necrosis [2]. This tissue infarction has been speculated as the cause of necrosis of the bone leading to fungal osteomyelitis. A review of the existing literature in the past or in pre-COVID times shows a low incidence of zygomatic bone osteomyelitis. In a survey of 141 cases of osteomyelitis by Adekeye et al. only 2 patients (1.42% of the study population) had involvement of the malar bone. [3] There now is however a noticeable increase in the number of patients presenting with zygomatic osteomyelitis secondary to CAM, a phenomenon which is highlighted in the following case series. It should be noted that there are no structured studies in the literature highlighting the incidence of CAM infection involving the zygomatic bone.

Patients and Methods

A descriptive study was planned at a tertiary care center, involving all patients with CAM of the paranasal sinuses and having a history of COVID-19 infection. Patients included in this study presented to the department of Oral and maxillofacial surgery over a period of 7 months (January 2021 to July 2021). Ethical approval was obtained from the Institutional Ethics Committee.

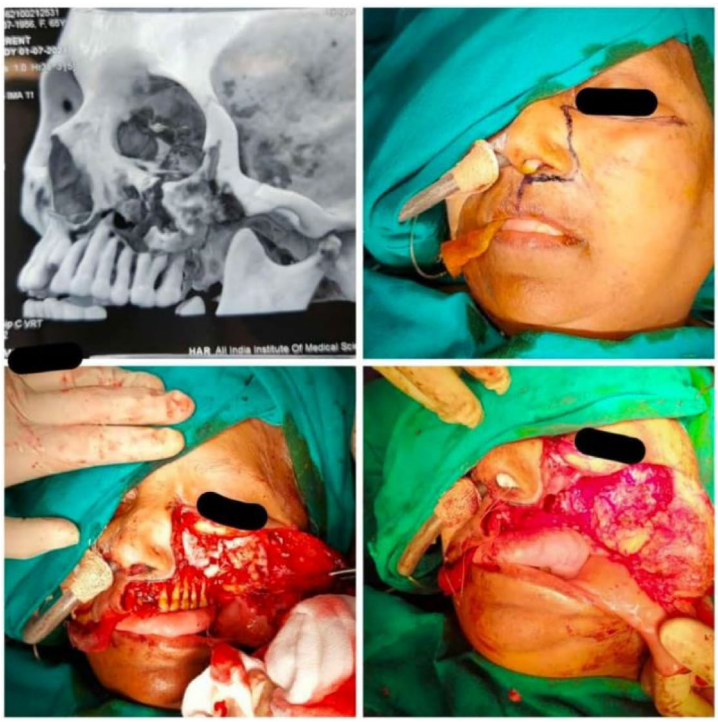
A total of 116 patients reported to our center of which 10 patients had involvement of zygomatic bone with an incidence rate of 8.6%. Four of the patients were female and 6 were male. Mean age of the patients was 56.4 years.

All the patients had a history of previous COVID-19 infection. Eight of the ten patients were admitted in COVID-19 wards and 2 of the patients were under home isolation being treated on an out-patient basis. Supplemental oxygen therapy was required for 6 patients during their COVID-19 infection treatment. Nine of the patients required Inj. Methylprednisolone during their treatments. Four patients were given Inj. Remdesivir treatment. Four of the patients developed type 2 Diabetes Mellitus during their COVID-19 treatment phase and the other 6 patients were known cases of type 2 Diabetes pre-infection. Two of the patients underwent dental extraction of teeth after discharge from COVID-19 hospitals and went on to develop CAM thereafter, incidentally affected on the same side as the dental extraction site. Seven of the ten cases had a longstanding history of hypertension and 1 patient had benign prostatic hyperplasia (BPH). Four of the patients developed type 2 Diabetes Mellitus during their COVID-19 treatment phase and the other 6 patients were known cases of type 2 Diabetes Mellitus. It is of note that all the 10 patients exhibited poor glycemic control which was a significant predisposing factor for the opportunistic infection. The patients did not report any other relevant medical history. Intra oral findings observed in the patients were, mobility of teeth in all of the cases, pain in teeth in 8 cases, multiple draining sinuses in 3 cases, palatal fistula was observed in only 2 cases and exposed bone intra-orally was present in 1 patient. Seven of the patients had nasal findings such as nasal stuffiness, perinasal edema and pain. Ocular findings which included pain, periorbital edema and proptosis were encountered in 4 patients. Neurologic findings were present in only one of the cases which was headache. Rhino-orbito-maxillary subtype was present in 4 patients. The rest of the cases were rhino-maxillary subtype. Rhino-orbito-cerebral subtype was not encountered. Either Contrast Enhanced Computed Tomography (CECT) of PNS + orbit + brain or plain CT HEAD or MRI was advised to the patients in preoperative period.

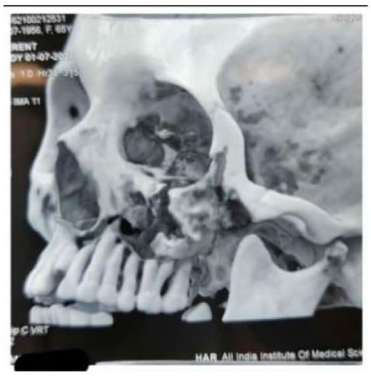
KOH mount of Pus culture/swab culture of all patients as well as tissue biopsy in all the patients presented with signs and symptoms of CAM showed broad-based, ribbon-like, non-septate hyphae with wide-angle branching (approximately 90°) in the laboratory diagnosis, thus confirming the diagnosis of CAM.

Surgical procedure was tailored according to each patient's findings and by extension of the infection. Endoscopic, open, and combined approaches were utilized with aggressive resection of the involved bone and sinus debridement (ethmoid, sphenoid, frontal and maxillary) followed by antifungal therapy to eradicate infection. Dual antifungal drug therapy with Amphotericin B and Posaconazole were given in all diagnosed cases of CAM. Tab Posaconazole 300 BD was given as a loading dose followed by 300 mg OD for 3 months and Injection Amphotericin B (Conventional/Lipid complex/Liposomal) was given 3 days pre-operatively and 7–15 days post-operatively. Overall, a protocol of aggressive surgical debridement was followed. All of the 10 patients first underwent Functional Endoscopic Sinus Surgery (FESS). Six out of ten patients had right sided zygomatic bone involvement and the remaining 4 had left sided zygomatic involvement. Treatment given to 4 of the patients was total maxillectomy + zygomatic resection + FESS and the remaining 6 patients underwent hemi-maxillectomy + zygomatic resection + FESS all via extraoral approach taking Weber Ferguson incision with Dieffenbach extension. Zygomatic resection extended till frontozygomatic suture and zygomatic arch in 2 of the cases. All the cases showed bilateral maxillary involvement of varying degrees of severity but it can be observed that the side which showed zygomatic involvement had more advanced progression of disease in

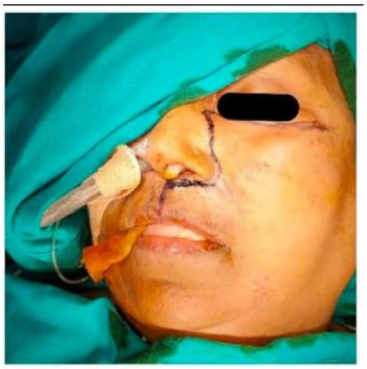
ipsilateral maxilla. Taking note that all 10 cases had unilateral zygomatic involvement, on the side without zygomatic bone involvement, treatment given in 3 of the cases was maxillary sinus debridement via Caldwell-Luc approach and the other 7 underwent hemi-maxillectomies. Seven of the cases underwent inferior nasal turbinectomies. All the cases had concomitant involvement of either unilateral or bilateral ethmoid and/or sphenoid and/or frontal sinuses. Pterygoid plate involvement was present in 1 case and pterygopalatine fossa was involved in one of the cases. Out of the 4 cases with orbital involvement one underwent lateral canthotomy and debridement of pus pouches and infected periorbital tissue. There was complete preservation of ocular movements post-operatively in this case due to meticulous surgical technique. 3DCT head image and intraoperative images of a patient are depicted in Fig. 1a–d. Image of the excised specimen of diseased tissue is depicted in Fig. 2. 3DCT face images of a patient with involvement of left zygomatic bone are depicted in Figs. 3 and 4. The intraoperative images of above-mentioned patient are depicted in Figs. 5 and 6. The 15-days follow-up images of the same patient are depicted in Figs. 7 and 8. Figure 9 depicts the patient at the six-month follow-up. Figure 10 depicts the intraoral view of the patient on the six-month follow-up visit.



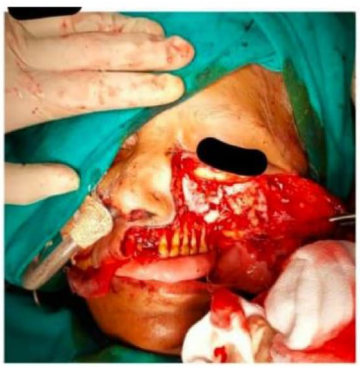
(a) 3DCT face image and intraoperative images.



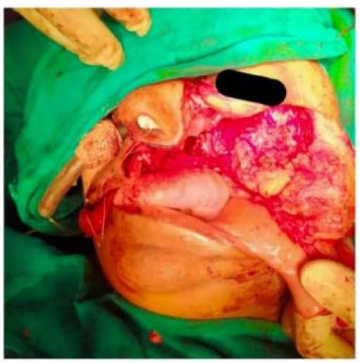
(b) 3DCT image showing zygomatic bone involvement.



(c) Marking of incision.

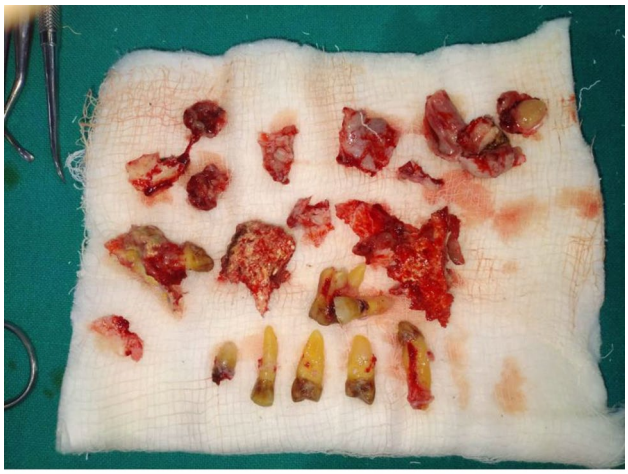


(d) Exposure of diseased bone.



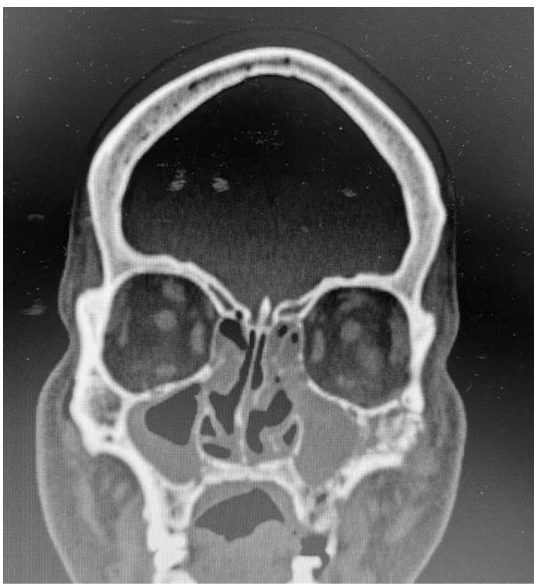
(e) Healthy bed of tissues after resection.

Fig. 1 3DCT face image and intraoperative images



Excised specimen

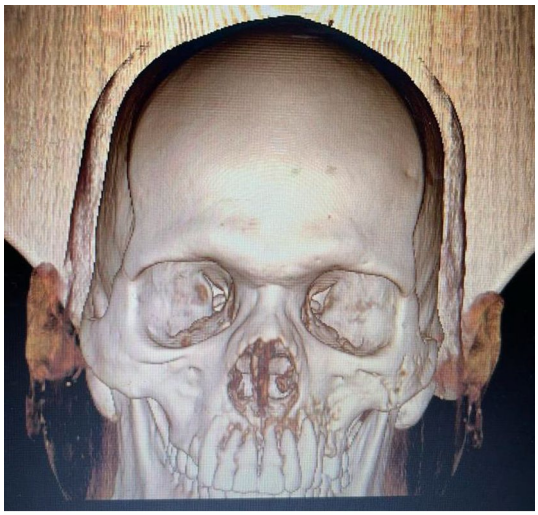
Fig. 2 Excised specimen



Coronal section showing involvement of left zygoma

Fig. 3 Coronal section showing involvement of left zygoma

Most common complication encountered in the post-operative phase was pain and post-operative edema. None of the cases developed wound site infection or wound dehiscence. Meticulous wound care, multivitamin supplementation and topical intraoral 0.1% Amphotericin B gel application rendered an uneventful post-operative period and patients were discharged on an average after 8 days. The patients reported for follow-up appointments after 3 days, 1 week and 2 weeks post-operatively with healthy healing wounds and were in good general condition.



3DCT representation showing left zygomatic bone involvement

Fig. 4 3DCT representation showing left zygomatic bone involvement

Intraoperative image showing diseased zygomatic bone

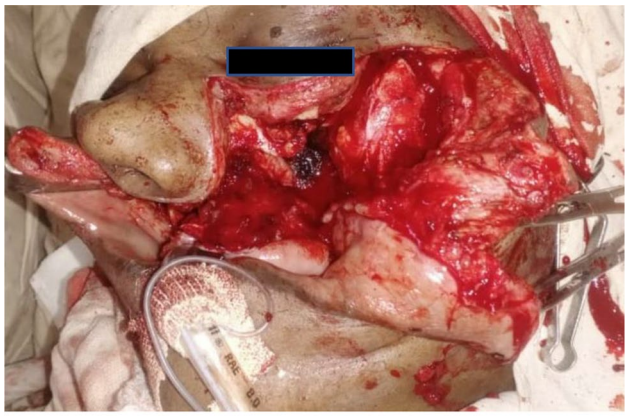
Fig. 5 Intraoperative image showing diseased zygomatic bone

Individual patient clinical features, treatment and outcomes are described in Table 1.

Discussion and Literature Review

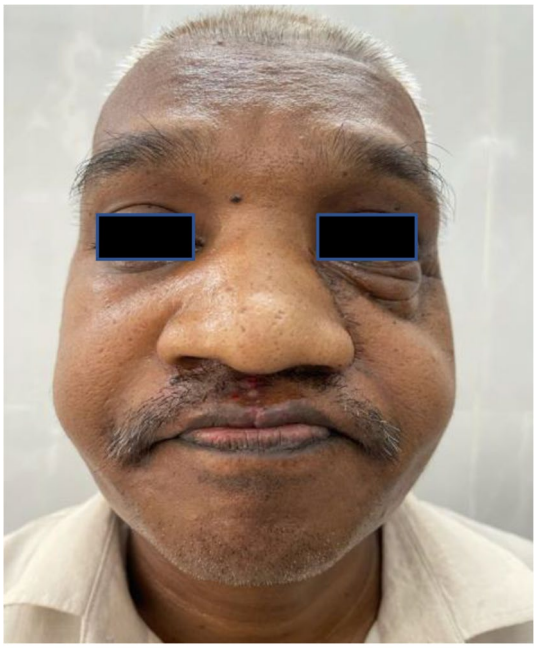
Osteomyelitis of bones in the middle third of the face is rare. A review of the literature shows that the mandible is the most commonly involved facial bone. Osteomyelitis of the maxilla is extremely rare. [3]

Osteomyelitis occurring due to fungal infection was rare and occurs in an indolent manner. Osteomyelitis is more



Healthy bed of tissues after resection

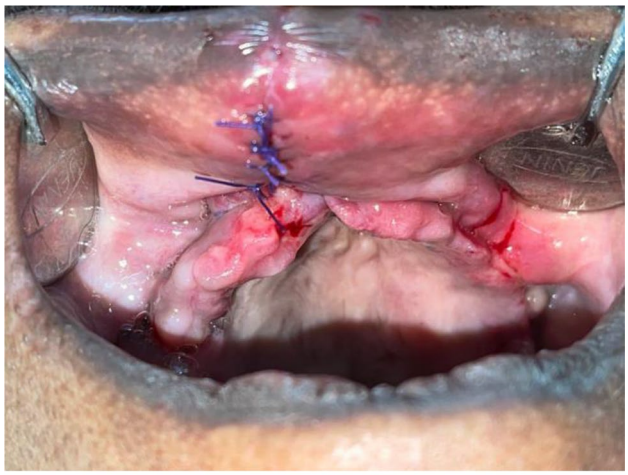
Fig. 6 Healthy bed of tissues after resection



15 day post operative image

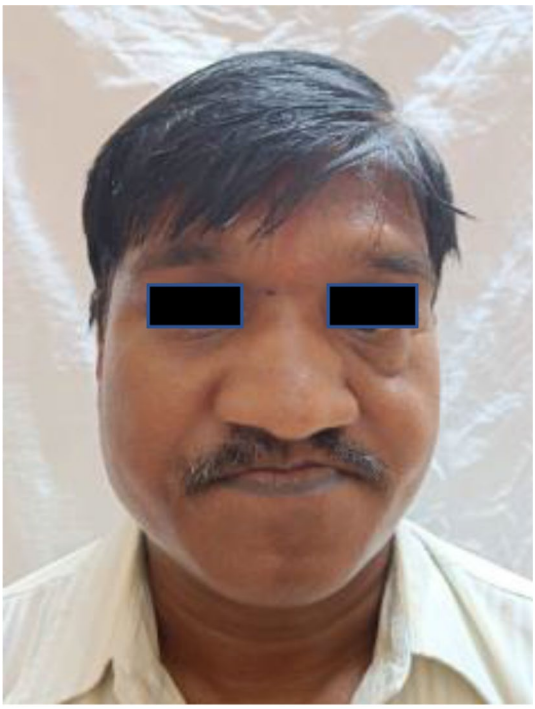
Fig. 7 15 day post-operative image

commonly seen in males (80.36%) than in females (19.64%), with a peak incidence in 30–39 years of age [4]. The maxilla is the most common jaw bone being affected by fungal osteomyelitis and is more commonly associated with diabetes mellitus. Among fungal osteomyelitis, Candida is the most commonly encountered followed by Aspergillosis and Mucormycosis. These organisms are from an original infection that has not been treated properly, commonly from dental extraction [5]. Niranjan et al., in their ten-year study reported that 52% of all the osteomyelitis cases were that of fungal osteomyelitis, whereas 48% belonged to the nonfungal category [6].



Healthy intraoral healing wound at 15 days post operatively

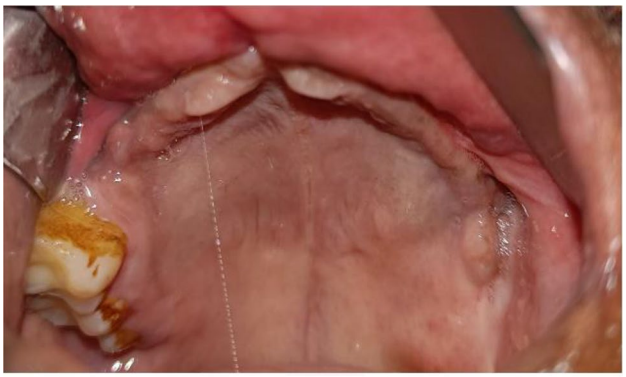
Fig. 8 Healthy intraoral healing wound at 15 days post-operatively



6 month follow up image

Fig. 9 6-month follow-up image

Adekeye et al. published a review of 141 cases of osteomyelitis of the jaws and reported the incidence of malar bone osteomyelitis to be only 1.42% [3]. The incidence of zygomatic bone osteomyelitis reported in our study of cases with CAM is 8.6%.



Intraoral view at 6 month follow up

Fig. 10 Intraoral view at 6-month follow-up

There are no other existing literature reviews or systematic reviews found in the existing pool of knowledge highlighting the incidence, causes and mode of spread of zygomatic bone osteomyelitis. There have been however isolated case reports and case series of zygomatic bone osteomyelitis. Most of these cases are due to tuberculosis and fungal causes (candidiasis, cryptococcus and aspergillosis). The majority of tuberculous cases of zygomatic osteomyelitis are due to hematologic spread of pulmonary tuberculosis by seeding of bacilli or by direct spread from neighboring structures [7]. There have been rare cases reported caused due to trauma and also idiopathic osteomyelitis of zygoma. An analysis of mode of spread of infection to zygomatic bone in a few rare cases reported in the literature is presented in Table 2.

Mucormycosis is an invasive fungal disease caused primarily by fungi belonging to the order Mucorales. This fungus usually acts as an opportunistic pathogen, seen in soil, decaying organic debris and frequently occurs in the patients with a compromised immune system. The leading predisposing factors for mucormycosis are uncontrolled diabetes mellitus, lymphomas, leukemias, renal failures, organ transplant, long-term intake of corticosteroids, immunosuppressive therapy and AIDS. Iron plays an important role in the growth of mucormycosis. Fungal hyphae produce 'rhizoferrin,' which binds iron fervently. This iron–Rhizoferrin complex is then taken up by the fungus and becomes available for its vital functions. In the cases of diabetic ketoacidosis, the patients are at high risk of developing mucormycosis, due to an elevation in the available serum iron [15]. Pertaining to the cases reporting to our institution is of special significance to note that all the 10 patients described exhibited poor glycemic control which was a predisposing factor for the opportunistic infection.

The infection develops after inhalation of fungal sporangiospores into the paranasal sinuses. The infection may then rapidly extend into adjacent tissues. Upon germination, the invading fungus may spread inferiorly to invade the palate, posteriorly to invade the sphenoid sinus, laterally into the cavernous sinus to involve the orbits, or cranially to invade the brain. The fungus invades the cranium through either the orbital apex or cribriform plate of the ethmoid bone and ultimately kills the host. Occasionally, cerebral vascular invasion can lead to hematogenous dissemination of the infection with or without development of mycotic aneurysms [16]. Upon visual inspection, infected tissue may appear normal during the earliest stages of spread of the fungus. Infected tissue then progresses through an erythematous phase, with or without edema, before onset of a violaceous appearance, and finally the development of a black, necrotic eschar as the blood vessels become thrombosed and tissue infarction occurs. Infection can sometimes extend from the sinuses into the mouth and produce painful, necrotic ulcerations of the hard palate [2]

In the present case series, the authors are of the opinion that the fungal spores enter via the nasal cavities and then spread to the paranasal sinuses. The maxillary sinus is thus invariably involved. The fungus erodes through the posterior aspect of the maxillary sinus gaining access to the infratemporal fossa. From the infratemporal fossa the infection directly invades into the body of the zygoma from the posterior aspect and in some cases also the arch of the zygoma. This has been deduced by the characteristic pattern observed in the CT imaging of each case which shows erosion of the posterolateral wall of the maxillary sinus with obvious haziness in the area of the infratemporal fossa. Retrospectively to co-relate with this clinically, at time of maxillectomy the posterior aspect of the maxilla and the pterygoid bone i.e., the infratemporal fossa and the structures therein were invariably diseased and required thorough debridement in all of the cases. This does indicate that spread to the zygoma from the maxilla could well be by way of the ipsilateral infratemporal fossa. According to the existing literature the zygomatic bone can be approached for resection intra-orally or extra-orally. For the cases in this series an open approach was preferred via Weber Ferguson incision to provide unhindered access to this posterior aspect and to be able to thoroughly debride the involved tissues of the infratemporal fossa. Although theoretically access to the zygoma can be achieved intraorally as well, it was deemed prudent by the surgical team to approach extra-orally for a more thorough, aggressive and definitive resection. The pathway of spread is elucidated by the flowchart in Fig. 11.

The incidence of zygomatic osteomyelitis in the literature is rare. Although a definitive cause for this has not been empirically stated in any of the past studies it can be speculated that it is due to the fact that the bone has a rich vascular supply and that there are no direct potential sources of infection such as teeth present in the bone. Additionally, the bone is not in any direct contact with the oral microflora nor is it in communication with the environment, it being a

Table 1 Table describing clinical features, treatment given and outcomes of patients involved in the study

Patient no	Age/ sex	Co-morbidities	Findings		Zygoma involved	Treatment		Outcomes
			Clinical	CT (sinuses affected)		Surgical	Antifungal	
1	43/M	DM, HTN	Maxillary left tooth pain and mobility, Draining sinuses, Left sided facial Pain and swelling, Nasal stuffiness	Left maxilla++ Right maxilla+ Nasal septum, Right ethmoid, Left zygoma	LEFT	Left zygomatic resection, left sided hemi-maxillectomy, right maxillary sinus debridement via Caldwell-Luc approach	*L- AmB Tab. posaconazole	Recovered
2	61/M	DM, HTN	Right sided facial pain and swelling, headache	Right maxilla++ Left maxilla++ Cavernous sinus, right ethmoid, bilateral sphenoids, right frontal, right zygoma	RIGHT	Right zygomatic resection, Bilateral hemi-maxillectomy	$^{+}$AmB Tab. posaconazole	Recovered
3	54/F	DM	Right sided facial pain and swelling	Right maxilla++ Left maxilla+ Bilateral ethmoids, Right zygoma	RIGHT	Right zygomatic resection, bilateral hemi-maxillectomy	AmB Tab. posaconazole	Recovered
4	73/F	DM, HTN	Right sided facial pain and swelling, Right periorbital edema, Ptosis of right eye, Vision loss with right eye	Bilateral maxillae++ Bilateral sphenoid, Bilateral ethmoid, Bilateral frontal, Right zygoma, Inflammatory edema of retrobulbar soft tissue	RIGHT	Right zygomatic resection, Bilateral hemi-maxillectomies	AmB Tab. posaconazole	Recovered
5	62/F	DM	Maxillary right tooth pain and mobility, Pain and swelling with right eye and cheek, Nasal stuffiness	Right maxilla++ Left maxilla+ Bilateral ethmoids, Right zygoma	RIGHT	Right zygomatic resection, right total maxillectomy, Left maxillary sinus debridement, Left inferior turbinectomy	L- AmB Tab. Posaconazole	Recovered
6	65/F	DM, HTN	Left sided facial swelling, Nasal stuffiness	Bilateral maxillae++ Left zygoma, Left ethmoid, Left sphenoid, Left pterygoid bone Left inferior, lateral and medial orbital walls	RIGHT	Right zygomatic resection up to frontozygomatic process with right lateral canthotomy, Right total maxillectomy, Left hemi-maxillectomy and pterygoid resection, Left inferior turbinectomy	L- AmB Tab. Posaconazole	Recovered

Table 1 (continued)

Patient no	Age/ sex	Co-morbidities	Findings		Zygoma involved	Treatment		Outcomes
7	59/M	DM, HTN, BPH	Left sided facial swelling and pain, Restricted eye movements with left eye, Nasal stuffiness	Bilateral maxillae++ Left zygoma body and arch, Bilateral ethmoids, Pterygopalatine fossa, Left Inferior rectus thickening, Inferior and medial and lateral walls of orbit	LEFT	Left side zygoma resection body and arch, Left sided total maxillectomy, Right hemi-maxillectomy, Bilateral inferior and middle turbinectomy	L- AmB Tab. Posaconazole	Recovered
8	52/M	DM, HTN	Left sided facial pain and swelling,	Left maxilla++ Right maxilla+ Left zygoma body and arch and frontozygomatic process, Bilateral ethmoid, Bilateral sphenoid	LEFT	Left zygomatic resection till frontozygomatic process, Left total maxillectomy, Right side maxillary sinus debridement via Caldwell- Luc approach, Bilateral inferior nasal turbinectomy	L- AmB Tab. Posaconazole	Recovered
9	46/M	DM	Right sided facial pain and swelling, Nasal stuffiness	Right maxilla++ Left maxilla+ Right zygoma, Bilateral ethmoid, Bilateral sphenoid	RIGHT	Right zygomatic resection, Bilateral hemi-maxillectomy, Right inferior turbinectomy	L- AmB Tab. Posaconazole	Recovered
10	50/M	DM, HTN	Left sided pain and swelling, Nasal stuffiness, Bloody red nasal discharge	Left maxilla++ Right maxilla+ Left zygoma Left ethmoid Bilateral sphenoids	LEFT	Left zygomatic resection, Left hemi-maxillectomy, Right maxillary sinus debridement via Caldwell- Luc approach, Left inferior turbinectomy	L- AmB Tab. posaconazole	Recovered

*L-AmB, Liposomal Amphotericin B; $^{+}$AmB, Amphotericin B Deoxycholate

++Extensive sinus involvement, +intermediate sinus involvement

Table 2 Review of literature describing zygomatic bone osteomyelitis with etiology, clinical features, mode of spread and treatment

Sr. no	References	Etiology	Clinical presentation	Likely mode of spread	Treatment given
1	Sethi et al. [7]	Secondary Tuberculosis	Erosion and sclerosis of left zygomatic bone with a soft-tissue abscess suggestive of osteomyelitis	Direct spread of infection from neighboring structures such as the orbit, paranasal sinuses, face and nasal mucosa	Anti-tubercular chemotherapy [7]
2	Rama et al. [8]	Primary tuberculosis	A sinus measuring 3×4 cm having undermined edges and yellowish pus discharge over the right zygomatic region	Primary extra-pulmonary zygomatic tuberculosis	Anti-tubercular chemotherapy [8]
3	Virendra Singh et al. [9, 9]	Secondary to pulmonary TB	Fluctuant swelling on the right zygomatic prominence	Hematologic seeding of bacilli	Surgical curettage and 4-drug antitubercular therapy [9, 10]
4	Takashi Matsuki et al. [11]	Cryptococcus	A tender swelling in the right zygomatic region and trismus	Isolated cryptococcal osteomyelitis of zygomatic bone	Fluconazole therapy for 6 months [11]
5	Arranz-Caso et al. [12]	Candidiasis	Tenderness of right zygomatic region	Self-inoculation of spores from muguet plaques on the oral mucosa to the exposed bone tissue (due to topical 5-fluorouracil treatment of the right malar region for actinic keratosis) by hand contact	Oral fluconazole, 200 mg/12 h, intravenous amphotericin B (0.5 mg/Kg/day, 40 days), a skin flap mobilized from the temporal region was implanted over the exposed bone [12]
6	Noroy et al. [13]	Aspergillus necrotizing otitis externa (NOE)	Blocked right ear for several months, associated with pain on mastication, without earache	Severe otitis externa with a foreign body in the external auditory canal, corresponding to cotton bud debris causing rare complication of Aspergillus NOE with temporomandibular arthritis and temporozygomatic osteomyelitis	Surgical curettage and 3 months of antifungal therapy [13]
7	Borle et al. [14]	Secondary infection due to trauma	Pus discharge from the cheek, infraorbital area, and lateral canthus region of the eye on the left side for 2 months	Incomplete immobilization despite direct fixation of zygomatic fracture	Sequestrectomy and debridement with sinus curettage [14]

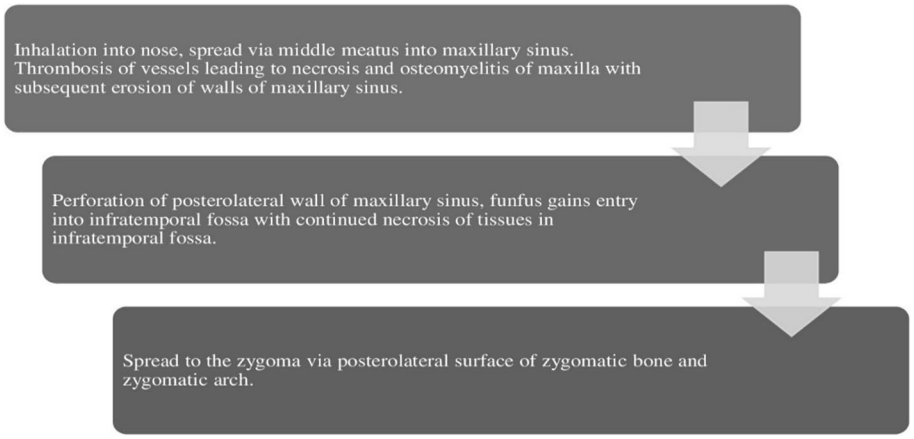


Fig. 11 The pathway of spread

solid, non-pneumatized bone, not bearing a sinus. With this in consideration an open approach was preferred via Weber Ferguson incision to provide unhindered access to this posterior aspect and to be able to thoroughly debride the involved tissues of the infratemporal fossa.

This case series attempts to highlight the unique nature of the mode of zygomatic involvement of the patients affected with CAM as compared to case reports and series presented in the past such as those due to tuberculosis, other fungi, trauma, etc. In our experience with cases of zygomatic bone osteomyelitis due to CAM, an aggressive surgical resection of bone and thorough debridement of diseased tissues combined with a dual antifungal drug therapy was the key to successful recovery. The authors recommend this treatment regimen for similar cases encountered.

Conclusion

The incidence of zygomatic bone osteomyelitis reported in our study of cases with CAM was found to be 8.6%. The mode of spread of infection to the zygomatic bone in cases of CAM is distinct from that of the other etiologies of zygomatic osteomyelitis reported in the past. The pattern of spread follows a distinct route and this should be kept in mind during the treatment planning and decision making for these cases. As the pattern of spread follows this predictable path the landmarks along the path of spread can serve as prognostic indicators for such cases and also serve as red flags for immediate intervention to prevent further spread and to reduce morbidity and to render a less challenging prosthetic rehabilitation. Considering the favorable outcome of the treatment protocol followed in this series, similar standard operating procedure can be employed in these cases encountered in the future.

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Declarations

Conflict of interest The authors declare that they have no conflict of interest.

Informed consent Informed consent was obtained from all patients.

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