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Case Report

Acute invasive fungal rhinosinusitis presenting with subperiosteal cheek abscess: A case report

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ABSTRACT

Mucormycosis is a severe, rapidly progressive fungal infection, seen in immunocompromised patients. We report an unusual case of mucormycosis of maxilla, presenting only with cheek abscess. The patient was a young male with uncontrolled diabetes. This presentation of mucormycosis is rare, complete examination and high degrees of suspicion are necessary. The rapid progression in lack of active treatment is often fatal.

1. Introduction

Acute rhinosinusitis (ARS) is expounded as the sudden onset of symptoms of nasal congestion, nasal discharge and obstruction along with facial pain or pressure and hyposmia or anosmia for a duration of <12 weeks.[1] There is inflammation of the paranasal sinuses mostly caused by viral and later bacterial infection, and thus typically presents with double-sickening pattern i.e. symptoms that worsen after an initial improvement.[2,3]

A saprophytic fungus, mainly mucor, rarely may cause a dangerous and rapidly progressive sinusitis, in an immunocompromised patient.

Here, we present a rare case of acute invasive fungal rhinosinusitis, in a young male with uncontrolled diabetes, presenting with only cheek swelling complaint. Performing incision and drainage for the cheek abscess, without looking for possible causes, would have been fatal to the patient.

The objective is to highlight that minor looking cheek abscess may be a tip of iceberg, i.e., more sinister pathology.

2. Case Summary

A 33-year-old male, presented to Otorhinolaryngology outpatient clinic with chief complaints of swelling of right side of cheek with loss of sensation, low grade fever since past 1 month. There was complaint of severe headache for 5 days. Swelling was insidious in onset, progressive, diffuse causing asymmetrical face. Patient gave history of repeated massage of the swelling with hot fomentation. Both the swelling and headache were not relieved on taking over the counter medications. There was no history of any nasal discharge or post nasal drip. There was no history of animal contact, dental pain, extraction or filling in recent time. He denied any recent maxillofacial trauma or cheek bite while chewing food. There was no history of any discharge from

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swelling. There was no history of difficulty mouth opening or chewing. He was a known case of type 2 Diabetes Mellitus, on Ayurvedic treatment for same. There was no history of hypertension, tuberculosis or bronchial asthma.

On local examination, there was facial asymmetry was evident due to the diffuse swelling 2cm X 2cm on the right cheek with loss of nasolabial fold. There was limited eye opening on right side compared to opposite side. Dark pigmentation was present on the right cheek and ala of nose. Swelling was diffuse, with no marked edges. Skin over swelling was shiny (Fig 1). Patient was febrile (100-degree F). On palpation, there was local rise of temperature and tenderness. Swelling was immobile with cystic to firm consistency at places. Fluctuation was present with transillumination absent. Examination of nose revealed left grade II deviated septum. Mucoid discharge was present in right nasal cavity along with slight blackening of the right vestibule. Left nasal cavity was normal. Bilateral maxillary and frontal sinuses were non tender as on the right-side patient had loss of sensation over the cheek. Intraoral examination of palate and teeth were normal, with no discolouration, swelling or discharge. Ear, pharynx, larynx and neck examinations were all within normal limits.

Diagnostic Nasal Endoscopy revealed pale with black at places right nasal cavity mucosa, oedematous middle turbinate, yellow uncovered bone over the lateral wall, mucopurulent discharge coming from the right middle meatus with no sensation on probing.

Computerised Tomography (CT) of paranasal sinuses showed right maxillary and ethmoidal sinusitis (Fig 2).

Laboratory data gave Haemoglobin as 12.2g% with raised total leucocyte count of 16,310/mm^3 with 65% polymorph cells. It also showed raised HbA1c level of 15.7% and random blood sugar level was 426mg/dL. The blood urea and serum creatinine were within normal limits. The urine examination revealed ketone bodies.

The patient was taken up for incision and drainage of cheek abscess via sublabial route under local anaesthesia. Approximately 5ml purulent collection was removed and sent for culture sensitivity. Intravenous antibiotics, Ceftriaxone 1gm 12 hourly and Metronidazole 500mg 8 hourly, were started empirically. Since patient had uncontrolled blood sugars insulin therapy, Human Mixtard 8unit morning, night and regular insulin according to sliding scale was started with medicine opinion. There was no associated ketoacidosis. Daily dressing was done with betadine-soaked wick in the abscess cavity. Endoscopic maxillary and ethmoid sinuses debridement were done under local anaesthesia, to remove the necrosed, inflamed mucosal lining on 3^{rd} day of admission (Fig 3). Intraoperatively, there was purulent discharge from maxillary sinus freely flowing into the nasopharynx from eroded medial wall of maxilla. The cavity was lavage and packed with antiseptic soaked ribbon gauze.

The excised mucosa was submitted for KOH mount in saline and histopathology in formalin. KOH mount revealed presence of aseptate, broad fungal hyphae. The histopathology confirmed presence of mucormycosis (Fig 4) and subsequently Amphotericin B deoxycholate therapy (1.0-1.5mg/kg/day) was started, after test dose. Cumulative dose of 2 gm of Amphotericin was given over a period of 6 weeks, with complete resolution of disease, with no complications due to therapy. At the weekly follow up for a month, endoscopic debridement was done in OPD, to remove excessive crusts. The patient was advised to continue nasal douching with normal saline. The monthly follow up till 9 months was done. The patient was symptom free and had no recurrence of disease, on endoscopic evaluation, and controlled blood sugar levels on oral hypoglycaemics.

Fig. 1: Clinical picture at presentation showing diffuse swelling of the right cheek with dark pigmentation of overlying skin

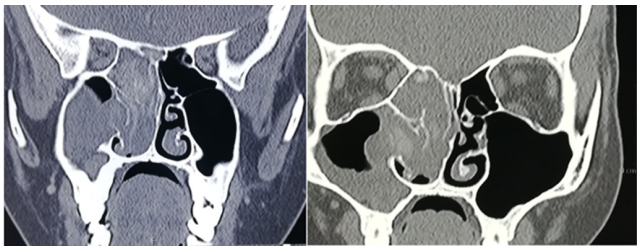

Fig. 2: Coronal section of Computed Tomogram showing soft tissue opacification of right maxillaand ethmoid sinuses

3. Discussion

Many consider pain of the facial region to be sinusitis, and most people are often right. Maxillary sinusitis can accompany nasopharyngitis often at times. There are however other causes including migraines or dental caries and rarely it can be idiopathic.

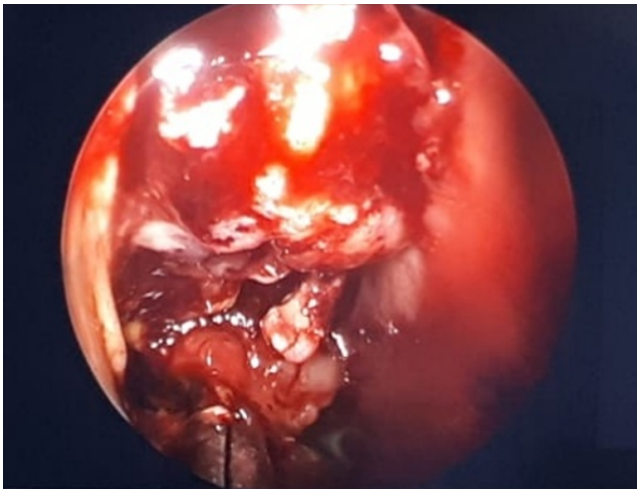

Fig. 3: Intraoperative endoscopic picture, showing necrosed mucosa and raw bonepresent on the superior and posterior region of medial wall of maxilla and inferior turbinate.

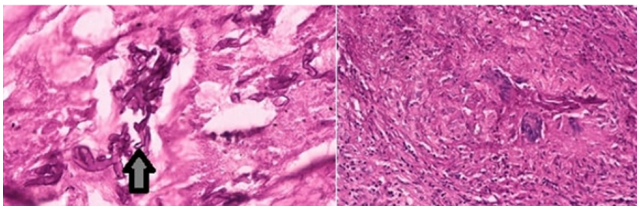

Fig. 4: Histopathology under PAS stain showed presence of broad aseptatefungal hyphae showing branching at right angles characteristic of mucormycosis (arrow marked)

Tests which can detect the symptoms of acute onset includes- swabs of the nasal cavity, ultrasound scans and lavages which can be diagnostic. In a tertiary care centre, more appropriate testing can be done for chronic conditions, including biopsy of the affected mucosa, frequency of ciliary beat and endoscopic inspection of sinuses etc. for which endocopic apparatus are required for accurate testing.[4,5]

Mucormycosis include a spectrum of infectious diseases due to Zygomycetes. Zygomycetes are a fungi group known for producing hyphae which have a ribbon-like appearance and sexually reproduce with the formation of special spores called zygospores. The pathogenic fungi are found abundantly in the earth, excrement etc. and can be obtained from the cultures of swabs of the nasal and oral cavities and throat swabs of people who are in good health and free of any disease. A variant of Zygomycetes, Mucorales can cause a varied and unique illness pattern. They are normally nonvirulent, and become infectious only when the resistance of the host organism is extremely low. Any break in the mucous membranes, tooth extrication in the oral cavity can provide a point of entry for organism in oromaxillofacial region, especially when the immunity of the person concerned is low.[6]

The infectious nature of the Mucorales is due to asexually formed spores. The spores which are miniscule can land on the mucosa of the oral and nasal cavities. In normal immunocompetent individuals, the immune system responds by phagocytosis and thus limits the further destructive nature of the spores being carried out. Failure to do so results in the formation of hyphae and further growth of the fungi. Neutrophils are not so effective in hyphae removal and any compromise to immunity of the individual will lead to the disease progression as the hyphal growth occupies the arterial vessels. This will lead to emboli formation and further causes necrosis of the regions affected. Vascular spread to the other parts of the body can result in septicemia and multiple organ failure.[7]

Increased blood sugar levels as a result of diabetes can make the host body more susceptible to infections due to reduced response of the body's immune system. It helps in the unrestricted growth of the fungi by decreasing the chemotactic efficacy and phagocytosis which prevents the usually harmless fungi to survive and proliferate in an acidic domain. In individuals who have ketone bodies due to diabetic ketoacidosis (DKA), Rhizopus oryzae species of fungi thrive by utilizing the ketone bodies,[8] by formation of ketoreductase enzyme. DKA throws the potential of the transferrin enzymes' iron binding into disarray. This results in a weakened immune response and further helps in the growth of Rhizopus. In the present case, the patient presented with diabetes mellitus.

Most clinically presenting features constitute rhinal, cerebral, respiratory and epidermal and rarely involves gastrointestinal symptoms or disparate forms.[9] In individuals with unrestricted blood sugar levels, rhinocerebral or rhinomaxiallary form of the disease presents most often.[10] Symptoms often include lethargy, pain and swelling of the face, headache, with mild rise in body temperature. The infection often starts in the oral or nasal mucosa and progresses to the sinuses by utilizing the ethmoidal, lacrimal blood vessels. The retro-orbital area is infected by direct invasion of the organism.[11] Vascular spread to further areas such as the brain, lungs can often be deadly to the infected individual.[12]

The other causes of oedema of the cheek includes squamous cell carcinoma, chronic infections such as syphilis, tuberculosis, asperigillosis and actinomycosis.[13]

On histopathological evaluation, broad fungal hyphae which are aseptate and had branching at right angles are observed.[14] The same was found in the present case. The fungal hyphae of Aspergillus sp. are septate, having a lesser width and often branches far more acutely.

On early detection, mucormycosis can be treated by removal of the diseased area surgically and by intravenously administering the antifungal amphotericin-B. The proper treatment of any immunocompromising conditions such as

diabetes mellitus is an important factor for full recovery.[15]

Our strength in management of this case was comprehensive ENT examination, supported by radiology and histopathology, which helped in early diagnosis and therapy, thus limiting the disease, with good outcome.

There were no limitations in diagnosis and management of our case.

4. Conclusion

Cheek abscess can be a rare presentation of mucormycosis. It is a dangerous, gradually intrusive fungal infection that can manifest in individuals with a compromised immune system, in our case uncontrolled diabetes. Early detection of the disease, as in our case and proper management can limit the disease and help in recovery without causing significant harm to the patient.

5. Declaration of Patients Consent

All required consent forms have been obtained from the patient. The patient has consented for the use of images and other clinical information for academic purposes. The patient understands that their identity will be concealed and there is no guarantee of anonymity.

6. Conflicts of Interest

The authors declare that there are no conflicts of interest regarding the publication of this paper.

7. Source of Funding

None.

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