Mucormycosis. Case report and literature review

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SUMMARY: Mucormycosis the life-threatening opportunistic fungal infection affects primarily immuno-compromised patients in an aggressive manner. Mucormycosis in the oral cavity is not common. The case we report presented with infra orbital swelling and pain with radiographic appearance of small area of bone destruction in maxillary anterior region. The patient also had uncontrolled type 2 diabetes. The patient underwent extraction of his anterior teeth 10 days back before he developed the swelling and pain. The primary diagnosis was osteomyelitis. After the histopathology diagnosis of mucormycosis the patient was immediately started with Amphotericin-b and surgical debridement of the involved site was done. The combination of medical and surgical treatment of mucormycosis will have good prognosis.

KEY WORDS: mucormycosis, amphotericin-b, type 2 diabetes, surgical debridement.

ΠΕΡΙΛΗΨΗ: Η μουκορμύκωση, μία απειλητική για τη ζωή ευκαιριακή μυκητιασική λοίμωξη, προσβάλλει κυρίως ανοσοκαταστολέντες ασθενείς. Εκδηλώνεται ως ρινοεγκεφαλική, πνευμονική ή διάχυτη μορφή σε ασθενείς σε κατάσταση ανοσοκαταστολής. Η μουκορμύκωση στη στοματική κοιλότητα είναι ασυνήθη. Στο παρόν περιστατικό ο ασθενής προσήλθε με οφθαλμικό οίδημα και πόνο με ακτινογραφική εικόνα μικρής περιοχής οστικής καταστροφής στην πρόσθια περιοχή της άνω γνάθου. Ο ασθενής εμφάνισε επίσης μη ελεγχόμενο σακχαρώδες διαβήτη τύπου II. Υποβλήθηκε σε εξαγωγή των προσθίων δοντιών του 10 ημερών πριν απαντήσει το οίδημα και τον πόνο. Η αρχική διάγνωση ήταν ακτινογραφο-κτιστική ακατάσταση χειρουργική διεξαγωγή της μουκορμύκωσης στον ασθενή. Ο ασθενής έμεινε επίσης με ελεγχόμενο σακχαρώδες διαβήτη τύπου II. Υποβλήθηκε σε εξαγωγή των προσθίων δοντιών του 10 ημερών πριν απαντήσει το οίδημα και τον πόνο. Η αρχική διάγνωση ήταν ακτινογραφο-κτιστική. Μετά την ιστοπαθολογική διάγνωση μουκορμύκωσης η ακτινογραφο-κτιστική διάγνωση ήταν ακατάστατη στον ασθενή. Ο ασθενής ακολούθησε ιατρική κεραμική διαμόρφωση και οικοδομή κεραμικής θεραπείας της προσθετικότητας. Περιλήψη: Η μουκορμύκωση είναι μία απειλητική για τη ζωή ευκαιριακή μυκητιασική λοίμωξη, προσβάλλει κυρίως ανοσοκατεσταλέντες ασθενείς. Εκδηλώνεται ως μυκητιασική λοίμωξη στην στοματική κοιλότητα. Ο ασθενής προσήλθε με οφθαλμικό οίδημα και πόνο με ακτινογραφο-κτιστική ακατάσταση. Η αρχική διάγνωση ήταν ιστοπαθολογική διάγνωση μουκορμύκωσης. Η συνδυασμός των ιατρικών και οικοδομικών θεραπείας της μουκορμύκωσης έχει καλή πρόγνωση. ΛΕΞΕΙΣ ΚΛΕΙΔΙΑ: μουκορμύκωση, αμφοτερικίνη-β, διαβήτη τύπου II, κεραμικής θεραπείας. Παρελήφθη: 6/3/2021 - Έγινε δεκτή 8/3/2021
INTRODUCTION

Mucormycosis is an invasive disease caused by a group of filamentous molds. The most common causative organism associated with Mucormycosis are Rhizopus species (Reid et al.). It has poor prognosis. It is a rare terminal complication of uncontrolled diabetes and other chronic debilitating disease. The incidence of the disease increased due to widespread use antibiotics and steroids. The fungi are normally avirulent and are able to invade the tissue only when the general resistance is low. The pathogens of mucormycosis involves inhalation or ingestion of sporangiospores of conidia. The nosocomial outbreaks of mucormycosis have been associated with medical waste, contaminated bandages, medical equipment, and ventilation systems (Kancharu et al.). By oxidative and non-oxidative killing mechanisms the mononuclear cells eliminate the fungal spores in healthy individuals. The various risk factors associated with mucormycosis are diabetes mellitus (poorly controlled and ketoacidosis), organ transplantation, autoimmune disorders, immunosuppressive therapy, HIV, burns, iron overload and malnutrition (Kancharu et al.). Mucormycosis can be classified based on anatomical location as: rhino-orbital - celebral - mucormycosis, pulmonary, cutaneous, gastrointestinal tract, disseminated and uncommon sites (bones, peritoneum, heart) (Prabhu et al., Palacios et al.).

CASE DESCRIPTION

A 55 year old male patient came with the chief complaint of pain and swelling in upper front teeth region for the past 4 days and the also complained of foul smell and nasal discharge. The patient had type 2 diabetes and hypertension for the past 15 years and was taking medication. There was no history of trauma on the affected side. On intraoral examination, there was swelling and ulcer in relation to 12, 13 and 14 region. Probing on bleeding was seen, and upper right buccal mucosa with black necrotic gingival tissue extending to maxillary central incisors. There was slight infraorbital swelling was seen, and upper right buccal mucosa with black necrotic gingival tissue extending to maxillary central incisors. There was no nasal discharge from the nose. On examination the patient was well nourished and no lymph node enlargement was noted. There was no nasal blockage on the right side. There was no nasal discharge. The patient had type 2 diabetes and hypertension for the past 15 years and the also complained of foul smell and nasal discharge. On intraoral examination, there was swelling and ulcer in relation to 12, 13 and 14 region. Probing on bleeding was seen, and upper right buccal mucosa with black necrotic gingival tissue extending to maxillary central incisors was seen (Figure 1&2). Dental history of the patient revealed that the patient had undergone uneventful extraction of right maxillary canine and lateral incisors 10 days back. Panoramic radiograph showed small area of bone destruction in right maxillary tooth region and generalized horizontal bone loss. Laboratory examination of blood sugar level, HbA1c, full blood count, erythrocyte sedimentation rate, retroviral screening was done. Gingival biopsy was taken for the histopathology diagnosis. Before laboratory and histopathology results, the patient with the provisional diagnosis of osteomyelitis was started with cephalixin, metronidazole and analgesics.

ΕΙΣΑΓΩΓΗ

Η μουκορμύκωση είναι μία επιθετική νόσος που προκαλείται από μία ομάδα νηματοειδών μυκών που ανήκουν στην τάξη Mucorales και Entomophthorales. Ο συνηθείστερος αιτιολογικός οργανισμός που σχετίζεται με τη μουκορμύκωση είναι το είδος Rhizopus (Reid και συν.). Έχει κακή πρόγνωση. Είναι μία σπάνια καταληκτική επιτοκία του μικρόκοκκου, με κλινική διάγνωση και διαγνωστικά μέτρα της νόσου. Αποτελεί μία σπάνια καταληκτική επιτοκία του μικρόκοκκου, με κλινική διάγνωση και διαγνωστικά μέτρα της νόσου. Είναι σημαντικό ότι η μουκορμύκωση μπορεί να ταξινομηθεί με βάση την εισπνοή ή εισήγαγη των σπόρων του μυκητιακού. Η μουκορμύκωση μπορεί να ταξινομηθεί με βάση την εισπνοή ή εισήγαγη των σπόρων του μυκητιακού. Η μουκορμύκωση μπορεί να ταξινομηθεί με βάση την εισπνοή ή εισήγαγη των σπόρων του μυκητιακού.
The blood sugar levels were elevated and HbA1c level was 9, retroviral screening turned out negative, WBC and ESR levels were elevated. The histopathology result came as mucormycosis. The patient was immediately started with amphotericin B and the surgical debridement involved removal of the maxillary right central incisor and curettage of all necrotic tissues. Bismuth Iodine Paraffin antiseptic pack was placed. The sinus was irrigated with 0.2% Chlorhexidine and 3% hydrogen peroxide solutions and the pack was regularly changed every 7 days for a period of 3 weeks till the wound healed completely. In between the patient was also referred to endocrinologist on the basis of laboratory results before the surgical debridement. The patient was asked to take insulin to control the diabetes by the endocrinologist. The patient was followed up for another 2 weeks and the wound was completely healed.

**DISCUSSION**

Oral mucormycosis is a rare disease mostly seen in immuno compromised patients as studied in the literature. Hard palate has been reported to be the most most
common site of involvement leading to tissue necrosis (Epstein et al., Bakathir et al.). Mucormycosis has the following symptoms: headache, facial pain, swelling and fever. The disease most commonly begins in the palate and extends to the paranasal sinuses spreading through the surrounding vessels (Rani et al.). Gingival involvement is not common, which is usually accompanied by ulceration and bone destruction which in turn leads to mobility of the teeth. In normal hosts, Rhizopus are eliminated by mononuclear and polymorphonuclear phagocytes by the production of oxidative metabolites and cationic peptides. The oxidative capacity is lost in patients with diabetic ketoacidosis. The reduced chemotaxis and phagocytic efficiency in these patients permit the growth of innocuous organisms. The hyperglycemic state patients are highly susceptible to mucormycosis since they promote fungal growth (Shastri et al.).

Once it begins to grow, invasion of human host takes place through attaching of the fungus to the endothelial cell lining of blood vessels leading to haematogenous spread, thrombosis, bleeding or infarction (Nicolatou-Galitis et al.). This leads to the formation of black necrotic eschars that are formed on the nasal or palatine mucosa, characteristic feature of mucormycosis. There is also evidence that an area of ulceration or an extraction socket in the mouth in an immuno compromised patient can also be the port of entry for mucormycosis (Cohen et al., Shastri et al.).

Histologically, mucormycosis is characterized by tissue necrosis, the presence of numerous large thin-walled fungal hyphae size ranges between 5-30μm, which are non-septate, branched at right angles and have a ribbon-like appearance (Rani et al., Cheong et al.).

As the disease progresses aggressively, immediate therapy and medication for the underlying disease is essential (Kudo et al.). For the treatment of mucormycosis to be successful it should include aggressive and repeated surgical debridement of necrotic tissue, antifungal therapy, and immediate control of the underlying systemic diseases. Amphotericin B is most commonly used along with the surgical debridement according the literature review (Rai et al., Swain et al.).

CONCLUSION

The early diagnosis of mucormycosis and immediate start of antifungal therapy and surgical debridement along with underlying systemic disease treatment helps to reduce mortality and morbidity of this disease.

ΣΥΜΠΕΡΑΣΜΑΤΑ

Η πρώτη διάγνωση της μουκορμύκωσης και η άμεση έναρξη της αντιμυκητιασικής θεραπείας και του χειρουργικού καθαρισμού μαζί με την θεραπεία του υποκείμενου νοσήματος βοηθούν στη μείωση της νοσηρότητάς και της νοσηρότητας αυτής της νόσου.
Mucormycosis/Μουκορμύκωση

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