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Orofacial Fusariosis Infection presenting with a complaint of Dental and Orofacial Pain in a Patient with Diabetes Mellitus

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Abstract

Fusarium species can cause opportunistic fungal infections, which usually present in the hospital setting, mainly in immunocompromised patients with hematologic malignancies. Early diagnosis and treatment is associated with improved survival in Fusariosis and other deep fungal infections.

We describe a patient seen in an outpatient oral medicine clinic with a two-month history of dental and facial pain, difficulty chewing and a history of Type 2 diabetes. He had seen a dentist 8 days previously, who extracted the upper right posterior teeth. No cultures or biopsies were done at the time of extractions. Examination revealed multiple gingival fistulas, suppuration from the recent extraction sockets, and radiolucent changes in the maxilla. A referral to the ear-nose-throat department was made the next day. Further investigations included CT scans, blood tests, nasal endoscopy, and culture from a nasal pus swab which grew Fusarium Solani, leading to the diagnosis. Treatment included antifungals, antibiotics, surgical excision, and control of hyperglycemia, however, local spread continued. Death occurred 7 months after occurrence of initial symptoms.

Our patient had an unusual orofacial presentation of Fusariosis, with dental pain and multiple gingival fistulas mimicking bacterial dental abscesses. In addition, his only risk factor was Type 2 diabetes. With the increasing incidence of diabetes worldwide and the ubiquitous presence of Fusarium species, dentists should consider these infections in the differential diagnosis of unusually virulent infections, even in immunocompetent patients.

Keywords

Fusariosis; deep fungal infection; dental pain; orofacial pain; non-healing extraction sockets; osteomyelitis

Introduction

Fusarium species are filamentous nondermatophyte fungi belonging to the class of Deuteromycetes of the order Moniliales [1, 2]. These organisms are present in plants and soil [3]. The Fusarium species known to be opportunistic pathogens in humans are F. Solani, F, oxysporum, F. moniliforme, F.proliferatum, F. chlamydosporum, F. Verticillioides, F.dimerum [1, 4]. Of these, F. Solani is the most virulent [3].

Fusarium species can cause infections when they are introduced into the body usually via breaks in the integrity of the skin or through the airborne route [5, 6]. The severity of infection may range from localized cutaneous infections, locally invasive infections to disseminated infections. Localized infections may occur in patients with preserved immune function while disseminated infections usually occur in immunocompromised patients [6].

Localized infections caused by Fusarium species include keratitis, onychomycosis, cellulitis, peritonitis and mycetomas [4, 5]. Disseminated infections caused by these organisms are generally seen in patients with severe immunosuppression due to hematological malignancies, stem cell or allogenic bone marrow transplants and organ transplant [4]. Patients with acute leukemia and prolonged neutropenia are at a greater risk of disseminated Fusariosis [6]. Diabetes mellitus, trauma, insect bites, and corticosteroid use are other less common predisposing factors [6-8].

Clinical manifestations of disseminated fusariosis may include high fever refractory to treatment, cutaneous lesions, sinusitis and pulmonary symptoms [6, 8]. Fusarium infection rarely presents with oral manifestations. These may occur secondary to seeding from circulating organisms in disseminated disease [1]. Oral involvement presenting as necrotic ulcers of gingiva, palate, and tongue has been reported [2].

The suspicion of Fusariosis or other deep fungal infections may be raised by consideration of the history, radiological presentation, clinical examination and biopsy findings [9]. The diagnosis of Fusariosis is made when blood cultures or cultures from tissue swabs are positive for Fusarium species [5, 8]. Histopathology may show presence of vascular invasion and acute branching septate hyphae, however on histopathologic examination Fusarium species are not easily distinguishable from other molds especially Aspergillus species. Polymerase chain reaction (PCR) based diagnostic methods have been described for detection of Fusarium species, and when available can provide more timely results without the delay necessary for growth in culture. In addition, when immunocompromised patients are on prophylactic antifungal medications, there may be an inhibition of the growth of the organisms in culture, making the PCR based methods more suitable [10]. However these diagnostic tests are not easily available.

The prognosis of disseminated Fusariosis is poor, with a high observed fatality rate [11]. In immunocompromised patients, the prognosis is influenced by the degree of immunosuppression [6]. Early diagnosis and treatment of Fusariosis, and when possible the underlying predisposing condition, result in an improved prognosis [8].

Treatment comprises of antifungal agents such as voriconazole, along with surgical debridement of localized infections. In addition, Granulocyte - colony stimulating factors (G-CSF) in neutropenic patients, and reduction or discontinuation of immunosuppressive medications may be necessary [6].

Case Report

A 62 year old male patient, presented to the Oral Medicine Department at Bhartiya Vidyapeeth Dental College (Pune, India) on July 4th, 2014, with a complaint of inability to eat due to pain on chewing, as well as a dull, continuous pain in the right and left jaw. Onset of these symptoms was 2 months previously. In addition, he also reported nasal congestion, severe headaches, malaise and intermittent fever for the past 1 month. He did not report presence of symptoms or lesions on the skin.

He had seen a dentist and had extractions of his upper right posterior teeth 8 days previously, and had been on the antibiotic cefixime for 4 days. No cultures or biopsies were done at the time of extractions.

His medical history was significant for diabetes which was diagnosed 2 years previously. He did not report any other past or current medical conditions, hospitalizations or surgeries. He was on medication for diabetes but couldn't recall the specific name, in addition, he reported that he did not take it regularly. On intraoral examination, the marginal gingiva in the maxillary arch appeared to be firm and resilient with normal contour and position at the cementoenamel junction. However, the attached gingiva was soft and edematous with multiple sinuses, in the region from the upper right first premolar to upper left 2nd premolar, with no mobility of the teeth evident. The alveolar mucosa showed non-healing extraction sockets in the upper right second premolar, and upper right 1st and 2nd molar region with exposure of bone (Figures 1 A-C). On palpation, the extraction wounds were not painful or tender, but suppuration was present. The palatal mucosa, on inspection appeared edematous with hyperpigmentation. On palpation, it felt soft and boggy in consistency but was non-tender.

Radiographic evaluation included a Panorex and occlusal radiographs which revealed presence of bony abnormalities (Figures 2A, B).

The patient was referred to the ENT (ear, nose, and throat) department on the next day and subsequently admitted to the hospital. Further investigations done in July 2014 included a CT scan, blood tests and a nasal endoscopy.

The CT scan revealed involvement of maxillary sinus with obliteration of the maxillary ostium and infundibulum bilaterally. The uncinate process and left middle turbinate were deossified. The nasal septum was deviated on left side with bony spur and focal breach. Blood tests included a CBC with differential which was within normal range, a cANCA test to rule out Wegener's granulomatosis which was negative. Blood glucose levels exceeded 300 mg/dl, which prompted a referral for management of diabetes and he was started on insulin and glimepiride.

The diagnostic nasal endoscopy findings done on July 12th 2014 included presence of a black necrotic crust in the right nasal cavity over the septum with exposed bone, granulations over the posterior floor with exposed bone. The middle meatus showed presence of thick pus. The left nasal cavity showed a marked deviated nasal septum with mucopus. The pus was collected and sent for culture and FESS (Functional Endoscopic Sinus Surgery) was scheduled. The patient was started on ceftriaxone, metronidazole and amphotericin.

The Right FESS procedure was done under general anesthesia on July 15th 2014 which showed the presence of a black necrotic ulcer on the inferior part of the septum, granulation over the posterior floor and posterior septum and thick pus in the middle meatus on the right side. The ulcerated and necrotic tissue was removed and sent for histopathological examination. The findings were suggestive for, but couldn't confirm mucormycosis. The culture report from the previously obtained nasal pus swab (obtained during the diagnostic endoscopy procedure) was received a week after FESS procedure on July 22nd 2014 which showed the growth of Fusarium species (speciation was confirmed as F. solani on July 29th 2014).

Medications used since onset of symptoms had included the antibiotics Ceftriaxone, metronidazole, ciprofloxacin, ampicillin-cloxacillin, and the antifungals amphotericin B, voriconazole. Intravenous Amphotericin was continued until July 28th 2014 (for a total of 17 days), however no improvement had occurred and he was switched to oral Voriconazole 200mg twice daily which was started on July 30th 2014, and was continued for about 1 month.

Subsequent surgical procedures included a debridement with partial maxillectomy under general anesthesia on August 12th 2014, followed by another debridement procedure on August 20th 2014.

The patient was discharged on September 1st 2014 on oral Voriconazole, and follow-ups scheduled.

Follow up phone calls revealed that the infection had spread to the orbit, however the patient had refused any further treatment and passed away in December 2014, about 7 months after initial presentation of symptoms of facial pain.

Discussion

Fusariosis is a relatively rare condition. In recent years, several cases of Fusarium infection involving the oral cavity and/or paranasal sinuses have been reported, mainly in immunocompromised individuals [12]. Often the diagnosis of Fusariosis is made during hospitalization for treatment of underlying hematologic malignancies, or shortly afterwards when the index of suspicion is high [13].

However a few cases of Fusariosis have been reported in immunocompetent persons [4, 5, 12].

Our patient, presented to an outpatient dental clinic with complaints of dental pain. He reported no current or past health problems other than a history of diabetes. He did report that he did not take his anti-diabetic medications regularly and his baseline blood tests revealed uncontrolled diabetes. In addition, the presentation was unusual as the mucosal lesions appeared as multiple fistulas on the gingiva, which somewhat resembled dental abscesses. No oral mucosal ulcerations were present.

The usual presentation of deep fungal infections in the oral cavity includes presence of ulcerations which may be chronic, deep and non-healing. The occurrence of osteomyelitis or fistulas is unusual. Although a few case reports of aspergillosis and mucormycosis have been reported with similar clinical presentation as our patient [9, 12].

Dentists may be consulted by patients with deep fungal infections when the early presentation includes oral or facial symptoms. Clinical examination findings of non-healing, deep ulcerations or extraction sockets, edematous tissue, exposed necrotic bone, multiple fistulas, and radiographic findings of bony destruction such as radiolucent changes, particularly in patients with a history of underlying immunosuppressive disease, diabetes, hematological malignancies or neutropenia should raise suspicion for deep fungal infections.

Biopsy is an important early step in the diagnostic process which can guide further investigations including cultures (tissue swabs/blood cultures). Because it is difficult to differentiate between fungal organisms on histopathologic examination alone, culture may be necessary to make the specific identification. This is crucial for selection of the most effective treatment.

With the increasing incidence of diabetes worldwide and the ubiquitous presence of Fusarium species, dentists should consider these infections in the differential diagnosis of unusually virulent oral infections, even in immunocompetent patients. In the rare cases with early oral presentation, dentists may be the clinicians most suited to initiate the diagnostic steps leading to a timely diagnosis in patients with deep fungal infections including Fusariosis.

Figures

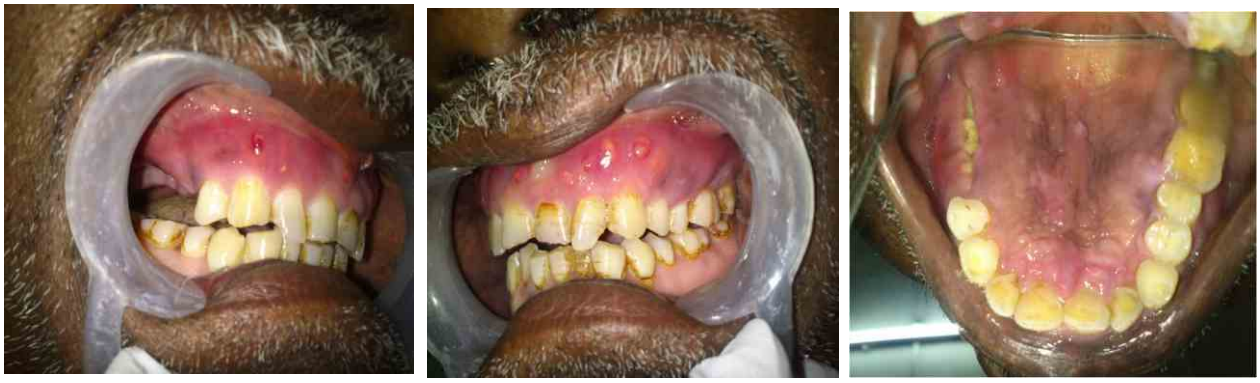

Figure 1 (a-c): Presence of multiple fistulas on the maxillary gingiva, and non-healing recent extraction sites with suppuration.

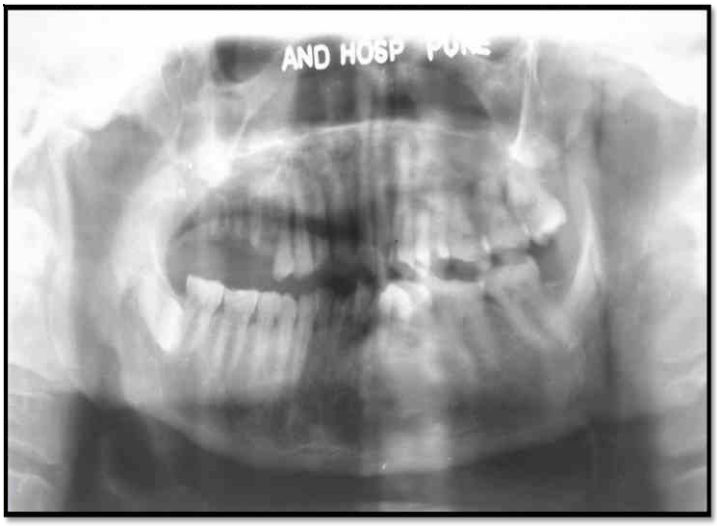

Figure 2a: Panorex showing non healing extraction sockets

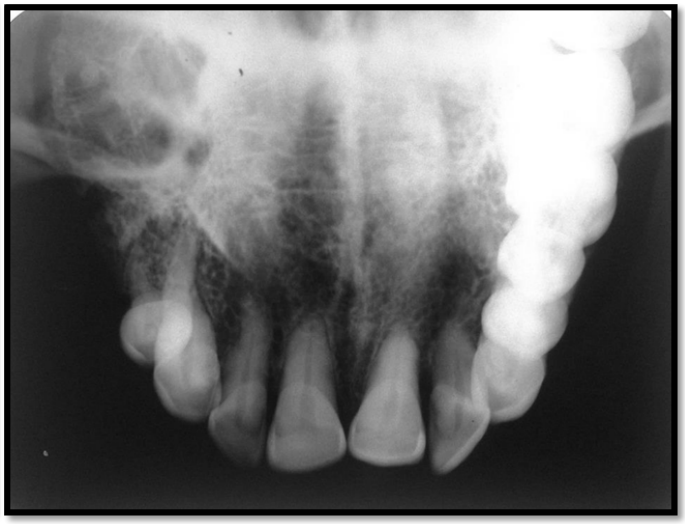

Figure 2b: True maxillary occlusal radiograph showing generalized rarefaction of the bone

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