

Dental Consideration of Exposed Maxillary Bone Secondary to Mucormycosis in Acute Lymphoblastic Leukemia: Case Report

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การดูแลทางทันตกรรมของภาวะรอยโรคกระดูกเปิดในช่องปากของผู้ป่วยเด็กที่เป็นมะเร็งในเม็ดเลือดขาวมีการติดเชื้อราไมโคร: รายงานผู้ป่วย 1 ราย

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Introduction

Mucormycosis is an invasive fungal infection which arises due to immune compromised state of individuals.¹ It is the second most common mucormycosis after aspergillosis caused by *actinomucor*, *rhizopus*, *rhizomucor*, and *mucor* species. The most important risk factor for mucormycosis development is malignant hematological disorders, bone marrow transplantation and prolonged corticosteroid administration in immunocompromised patient.² Predisposing factors also includes malnutrition, prematurity, HIV infection and diabetes mellitus.³ The characteristics of *mucor* species infection is rapid vascular invasion followed by vascular thrombosis and tissue necrosis.⁴ The patients of leukemia having impaired immunity are more prone for mucormycosis infection.⁵ Usually, mucormycosis presents as an acute infection and manifests as rhinocerebral, pulmonary, gastrointestinal, cutaneous or disseminated forms.⁶ Immunosuppressive condition of the body rise the chance of opportunistic fungal infection.³ Uncontrolled invasive fungal infection can cause destruction of surrounding tissues and bone and, can cause exposure to external environment.⁵ Exposed maxillary bone can occur due to trauma, bacterial infection leading to osteomyelitis, viral infection like herpes zoster, fungal infection such as mucormycosis, and malignancies. Exposed maxillary bone may cause oro-antral communication and fistula which is an unusual communication between the oral cavity and the maxillary sinus. The aim is report a case of exposed maxillary bone due to mucormycosis in a nine year old child of acute lymphoblastic leukemia (ALL) for the dental consideration.

Case report

A 9-year-10-month old Thai boy with relapsed acute lymphoblastic leukemia visited dental department of Queen Sirikit National Institute of Child Health (QSNICH), Thailand for dental examination and treatment. The child was diagnosed as acute lymphoid leukemia (ALL) with febrile neutropenia at the age of 2 years and 7 months. After confirmatory diagnosis of ALL, the child was following the treatment protocol of ALL. At the age of 2.9 years the child was admitted in QSNICH and received induction chemotherapy using low risk ALL protocol until recovery. Following recovered from ALL and the child was on every 3 months follow up treatment status. During follow up treatment hematological status, cardiovascular system, respiratory system, gastrointestinal system and renal system was investigated and was found stable. At the age of 9.2 years recurrence of ALL occurs with the symptoms of low fever followed by high fever, vomiting, headache and blurred vision. The manifestations diagnosed as relapse ALL with CNS involvement (high risk). The child was recommended to follow

treatment protocol for high risk ALL. The child was received chemotherapy along with radiotherapy. After four months of chemotherapy and two months of radiotherapy invasive fungal sinusitis were developed. The chemotherapy was stopped and anti-fungal therapy was started. The patient was planned to continuing the chemotherapy. Invasive mucormycosis involves nasal septal cartilage, nasal bridge and mucosa of floor and lateral nasal cavity. Bone exposed at upper buccal plate of the left lateral incisor connected to the floor of nose (fig 2) occur which is secondary to *mucor* species infection.

The examination of the oral cavity initially showed generalized gingivitis (fig 1) and bone exposure occurred in at upper buccal plate of the left lateral incisor connected to the floor of nose (fig 2). Prolonged retention of lower right primary molar, deep dental caries in upper right and lower left permanent molar was noted. Examination of occlusal topography of maxilla (fig 3) and maxillofacial computed tomography (CT) images revealed extension of lesion and mucosal thickening of the left maxillary sinus. (fig 4).



Fig 1 Generalized gingivitis



Fig 2 Exposed maxillary bone in vestibule of mouth

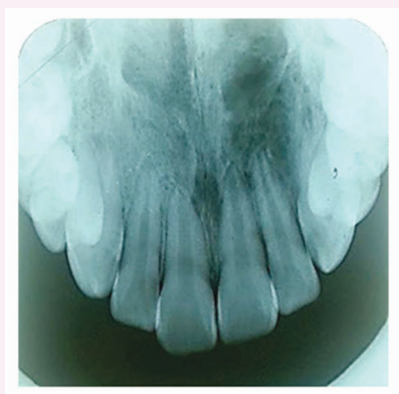


Fig 3 Occlusal topography of maxilla



Fig 4 Thickening of left maxillary sinus in CT scan

Generalized gingivitis developed due to lack of proper oral hygiene care. The oral hygiene instructions were given to the child and his mother. The patient was advised to use the soft tooth brush and wet cotton for cleaning the teeth, especially upper anterior teeth. Lower left second primary molar was extracted due to prolonged retention in oral cavity. Upper right and lower left permanent molar also noted deep carious lesion. Filling of the both permanent molar with restorative material was done after removal of carious lesion. All primary and permanent dentition remaining in the oral cavity were sealed with resin sealant in order to prevent initiation of new carious lesion. Remaining tooth structures were reinforced by applying fluoride varnish. Complete dental

rehabilitation was achieved (fig 5). Bone exposure that occurred in the vestibule of the mouth just over the left upper lateral incisor (fig 2) was surgically closed by the help of Otorhinolaryngology surgeon. During the follow up visits, it was found that gingivitis was subsided except maxillary anterior region due to lack of proper brushing in that particular area. History revealed that, child felt pain due to maxillary sinus involvement during brushing the upper anterior region. The child and mother, both were instructed to carefully clean the bone exposed area and upper anterior teeth using soft wet cotton to prevent accumulation of plaque. Now, all dental problems are subsided and child is in regular follow up at the dental clinic



a. Upper teeth



b. Upper and lower anterior teeth



c. Lower teeth

Fig 5 Intra-oral photography showing good oral hygiene after complete oral rehabilitation

Discussion

Mucormycosis associated with hematological malignancy is a severe infection with high mortality rate.⁷ Recent studies has revealed that, hematological malignancies were underlying diseases in 58–60% cases of mucormycosis.⁷ Multidisciplinary approach is required for optimum management of mucormycosis. More than 50% patient with mucormycosis demonstrates rhinocerebral involvements which entangle nose, paranasal sinuses and palate.⁹ The common manifestations of invasive mucormycosis includes black necrotic lesion of palate and could be involving gingiva, cheek mucosa and pharynx.¹⁰ Other atypical symptoms may include facial pain, sinus pain or odontalgia.⁸ Dental pain and tooth mobility due to intraoral involvement leads the patient to visit dentist. More precise dental examination might have state the need for further investigation of the condition. It is imperative that all dental practitioners record the patient's medical history and modify their diagnostic or therapeutic actions accordingly health status of patient.⁸ Management of invasive mucormycosis includes periodic debridement of necrotic tissue, anti-fungal therapy as well as maintenance of underlying disease.¹ Dental health rehabilitation and maintenance of oral health is necessary for prevention of further infectious involvement.³ After subside of all infectious condition, regular monitoring and periodic follow-up is required in order to prevent recurrence.

Conclusion

Although invasive mucormycosis is a rare infective disease, but attentive acquaintances of dentist can plays a vital role for elimination of dental pain, restore periodontal health and complete rehabilitation of the mouth. Following establishment of perfect oral hygiene and elimination of vulnerable tooth infections, underlying disease condition can be maintained and successful outcome is expected.

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