ISSN: 2581-5989
PubMed - National Library of Medicine - ID: 101738774

International Journal of Dental Science and Innovative Research (IJDSIR)
IJDSIR : Dental Publication Service
Available Online at: www.ijdsir.com
Volume – 3, Issue – 6, November - 2020, Page No. : 179 - 184

Prosthetic Management of Hard Palate Perforation caused due to Rhinosporidium infection in a child with Dimorphic Anemia: A Case Report

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Citation of this Article: Dr. Deshraj Jain, Dr. Alka Gupta, Dr. Unnati Gedam, Dr. Amit Varma, "Prosthetic Management of Hard Palate Perforation caused due to Rhinosporidium infection in a child with Dimorphic Anemia: A Case Report", IJDSIR- November - 2020, Vol. – 3, Issue - 6, P. No. 179 – 184.

Type of Publication: Case Report

Conflicts of Interest: Nil

Abstract

Palatal perforation is a rare condition and can be due to congenital or acquired causes. The etiologies of acquired palatal perforations include developmental disorders, infections, malignancy, and drug abuse. Palatal lesions generally present in the form of ulcers and in severe forms as perforation. This case report illustrates the prosthodontic management of palatal perforation caused due to Rhinosporodiosis in 18 months old child diagnosed with dimorphic anemia. With the help of this case report, we try to touch on the spectrum of etiologies of palatal lesions and their necessity for appropriate management.

Keywords: Rhinosporidium infection, Dimorphic Anemia, Obturator, Palatal perforation, Case report.

Introduction

Palatal perforations create communication between the oral cavity and the nasal cavity. Palatal defects may be congenital or acquired. Acquired defects can occur due to trauma, bacterial, viral, or fungal infections. Palatal perforation is more common in immunocompromised patients as they are at risk for opportunistic infections that may invade the palate and cause its perforation. Rhinosporidiosis is a rare, infectious disease caused by Rhinosporidium seeberi. The disease predominantly occurs in warm, tropical climates, with over 90% of cases

found within the Indian subcontinent. The nose and nasopharynx are most commonly involved (70% of reported cases), with ocular manifestations accounting for 15% of cases. [1] It is a granulomatous inflammatory disease-inducing polyp of the affected mucous membrane. Rhinosporidiosis affects all ages but is most common in the third and fourth decades with a male predilection. [2] The goals of prosthetic rehabilitation include separation of oral and nasal cavities which helps to improve the basic functions, such as speech, swallowing, and chewing. These defects are more difficult to restore because they are generally lined with respiratory mucosa and poorly keratinized squamous epithelium, which makes it difficult for the patient to tolerate the prosthesis.

This case report illustrates the prosthodontic management of palatal perforation in an 18-month-old child diagnosed with dimorphic anemia. Palatal perforation caused due to rhinosporidiosis at this age is extremely rare and has not been reported in literature earlier. This case is significant taking into consideration, the age of the patient and palatal perforation caused by this disease.

Clinical Report

Case Presentation: A 18-month-old boy, was referred to the Department of Prosthodontics, Government College of Dentistry, Indore (M.P.) for prosthetic rehabilitation of a palatal perforation (Figure 1). History revealed that the patient was diagnosed with Rhinosporodium infection which led to the perforation of the palate. On general physical examination, the child had poor built, pale appearance, hypopigmented hair but conscious and afebrile. On extra-oral examination festering wound, open nasal cartilage, nasal septum, pyriform apparatus unusual undercuts, and other tissue abnormalities were noted. On intra-oral examination, palatal perforation was found extending from lip to covering three-fourth of the anterior palate, obliterating the labial vestibule. Residual necrotic tissue was visible in the perforation with exposed bone. Teeth no. 53, 56, 62, 63, 66 were seen erupting with only their occlusal surface visible.

A detailed case history revealed that the patient was apparently alright before 6 months. The patient was admitted to the Pediatric medicine department, Maharaja Yeshwant Rao Hospital, Indore (M.P.), with chief complaints of not accepting feed, accompanied by cold and fever, and keeping dull. On 3-4 days after admission, the patient developed complaints of respiratory disorders and peri-oral and peri-orbital swelling which hardened further with pus discharge. Pus discharge was initially noted from the right ala of the nose and later inside the oral cavity. On routine blood investigations and bone marrow aspirate, the patient was diagnosed with dimorphic anemia. CT scan of the paranasal sinuses (CT-PNS) revealed extensive soft tissue thickening involving bilateral ethmoid complexes, maxillary sinuses protruding into the nasal cavity with significant obliteration. Debridement of necrotic tissue was done under general anesthesia, by the Department of Oral & Maxillofacial Surgery, and biopsy of the sample was sent which revealed Rhinosporodium seeberi. Simultaneously patient was managed for severe anemia with blood transfusion, antifungal drugs, antibacterial Drugs, and multivitamins.

Later when the patient was accepting oral fluids well and vitals were stable, the child was referred to the Department of Prosthodontics for fabrication for management of palatal perforation. Following complete evaluation and thorough explanation of treatment goal and procedure, fabrication of a feeding plate or palatal obturator was planned. An explanation of the procedure using visual aids allays early anxiety and parents usually get enthusiastic with the prospect of becoming active participants of the child's treatment.

Impression making: A prefabricated perforated impression tray of auto polymerizing acrylic resin (DPI Autopolymerizing acrylic resin, Indore, Madhya Pradesh) was selected, evaluated intraorally, and was adjusted in accordance with the defect of the patient (Figure.2). Polyvinyl siloxane putty impression material (Ad- Sil Putty, Prime Dental Pvt. Ltd., Indore, Madhya Pradesh, India) was mixed, and the loaded impression tray was seated in the patient's mouth until the impression material adequately covered the anatomy of the upper arch. Care was taken to ensure the registration of good detail and depth for prosthesis construction. During the impression making the infant was held with his face down to avoid aspiration. Once the impression material was set, the tray was removed, an impression (Figure. 3) was evaluated for its completeness and the oral cavity was examined for any residual impression material. The impression was poured with type III gypsum product (Kalstone; Kalabhai Karlson, Indore, Madhya Pradesh) to obtain a definitive cast (Figure. 4).

Prosthesis design and fabrication: The size of the palatal perforation was measured from the cast using a vernier caliper (Digital Vernier Caliper; Sealy Power Products, Suffolk, UK) which was found to be 3cm x 1.5 cm. The region showing perforation was filled in with base plate wax (Modeling wax; Deepti denal products, (Rolex Modelling wax no. 2 ASHOO SONS), to approximate the contour and topography of an intact alveolar arch form and the prosthesis was fabricated with auto polymerizing acrylic resin (Travelon Clear Denture Material, Dentsply India, Gurgaon, India) of 2–3 mm thickness to provide structural integrity and to permit the future adjustments (Figure 5). The prosthesis was finished and polished to ensure that all tissue borders are smooth and highly polished, especially in the area of the labial frenum and oral portion that will contact with the tongue. The prosthesis was carefully inserted in the child's oral cavity and observed for a few minutes. The child was able to suck and swallow without gagging or struggling.

Extra-oral retentive wire: Two holes were drilled in the anterior part of the obturator and orthodontic wires were tied to these holes. The wires serve the purpose of extra-oral retention. The wires were contoured according to the lip curvature.

Retentive taping with surgical adhesive tape: The prosthesis was secured extra-orally to the cheeks and bilaterally by retentive taping with surgical adhesive tapes. A broader base tape (0.5x2 inch) (Generic laboratories Wound Care, St Paul, MN) was applied to the child's cheeks bilaterally (Figure 6).

Prosthesis delivery and instructions: A feeding obturator was inserted into the patient's mouth and it was checked for fit, comfort, and retention. Before delivering the prosthesis, final finishing and polishing were done. The appliance was placed in the child's oral cavity and the patient's mother was asked to feed the baby and it was noted that there was no nasal regurgitation and the child was successfully ready to feed with the feeding obturator in-situ with none discomfort. Parents were given post-insertion instructions and taught how to insert the plate in the oral cavity and how to feed the infant. Parents were asked to take care of the oral hygiene of the infant and regarding the cleaning of the feeding plate. They were also instructed to thoroughly clean the baby's oral cavity with a soft cloth soaked in warm water after every feed. The patient was seen after 24 hours for adjustment, and then the patient was followed up regularly after every 1 month.

Discussion

Rhinosporidiosis is a rare, infectious, chronic granulomatous disease-inducing polyps of the affected mucous membrane. It is caused by Rhinosporidium seeberi, an endosporulating microorganism. Its first

description was given by *Sir Guillermo Seeber* in Argentina. *Ashworth* in 1923 gave the nomenclature Rhinosporidium Seeberi. [3] The disease is most prevalent in warm, tropical climates, with over 90% of cases found within the Indian subcontinent. The nose and nasopharynx are most ordinarily involved at over 70% of reported cases, with ocular manifestations accounting for 15% of cases. [1-2] The disease commonly affects the nose (80%), nasopharynx (15%), oropharynx (3%), lacrimal sac, and the nasolacrimal duct (2%). The male to female ratio is found to be 10:4.[4] According to *Sinha et.al*, there have been more reported male cases compared to females, but the difference is minimal (57% to 43%), with the majority of cases involving young adults. [2] Rhinosporidiosis is most common in the third and fourth decades. The younger age groups may be at risk due to the likelihood of young adults participating in outdoor activities. No person-to-person transmission is noted. [4]

The role of imaging in the diagnosis and management of Rhinosporidiosis is very crucial. Imaging modality like CT scan of nose and paranasal sinuses reveal the precise site of attachment and extent of involvement. A definitive diagnosis depends on histological examination with Immunohistochemistry primarily conducted through excisional biopsy, scraping of superficial lesions, or fine-needle aspiration. [4] The histopathological sections will show sporangia in multiple stages of maturation enclosed within a thin wall. The sporangia can vary from 50-1000 μm in diameter.[5] Rhinosporidiosis infection is primarily treated with surgical intervention however medical management has been reported as an adjunctive treatment. Complications of rhinosporidiosis are relatively rare. Disseminated infection may occur, leading to osteolytic lesions of bone. The disease is fatal if it affects the tracheobronchial tree where it can cause obstruction of the airway. Despite the rare case of recurrence or dissemination, the prognosis is generally very favorable. [6] Differential diagnosis includes pyogenic granuloma, Squamous or conjunctival papilloma, hemangioma, arteriovenous malformation, chronic infection, oncocytoma, sebaceous adenoma, squamous carcinoma foreign body, Conjunctival cyst, and Inflamed pinguecula. [2][7][8]

In the present case, the histopathological sections showed cystically dilated spaces with sporangia embedded in fibro collagenous tissue along with mixed inflammatory cell infiltrate and areas of necrosis suggestive of Rhinosporodoisis. Peripheral smear depicted moderate anisopoikilocytosis and leucocytosis with neutrophilia and bone marrow aspirate depicted diluted marrow. These results were suggestive of hypocellular diluted marrow showing features of dimorphic anemia. Palatal perforations are commonly reported in immunocompromised patients as they are more prone to opportunistic infections.

The goals of prosthetic rehabilitation include separation of oral and nasal cavities which helps to reinforce the essential functions, like speech, swallowing, and chewing. These defects are harder to revive because they're generally lined with respiratory mucosa and poorly keratinized squamous epithelium, making it harder for the patient to tolerate the prosthesis. Feeding such children poses challenges to the parents. Feeding obturator helps in minimizing the feeding problems in palatal perforation. *Neha. et al.* fabricated the feeding appliance to Resolve Recurrent Ear Infections in an Infant with Isolated Cleft Palate in a similar manner [9] It normalizes tongue position resulting in better speech, reduces feeding time, provides a positive psychological impact on the parents, and promotes the quality of life which is important in preparing the child for future corrective surgery.

Conclusion

This paper addresses the fact that Rhinosporodiosis, a rare fungal infection can lead to palatal perforations in children if accompanied by some debilitating diseases. Palatal perforations may predispose the patient to hyper-nasal speech, fluid leakage into the nasal cavity, and impaired masticatory function. This severely affects the patient's quality of life. A properly fabricated palatal obturator solves the purpose. It is extremely essential that such patients should be provided with adequate nutrition and meticulously monitored for their oral hygiene.

Declaration of Patient Consent

The authors certify that they have obtained all appropriate patient consent forms. In the form, the patient(s) has/have given his/her/their consent for his/her/their images and other clinical information to be reported in the journal. The patients understand that their names and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

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Legends Figures

Fig 1: Image showing palatal perforation.

Fig 2: Pre-fabricated perforated tray made with autopolymerizing resin with the help of stock tray.

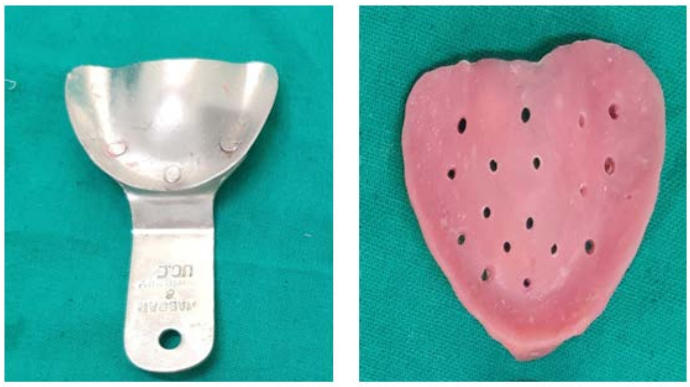

Fig 3: Impression was made with Polyvinyl siloxane putty impression material

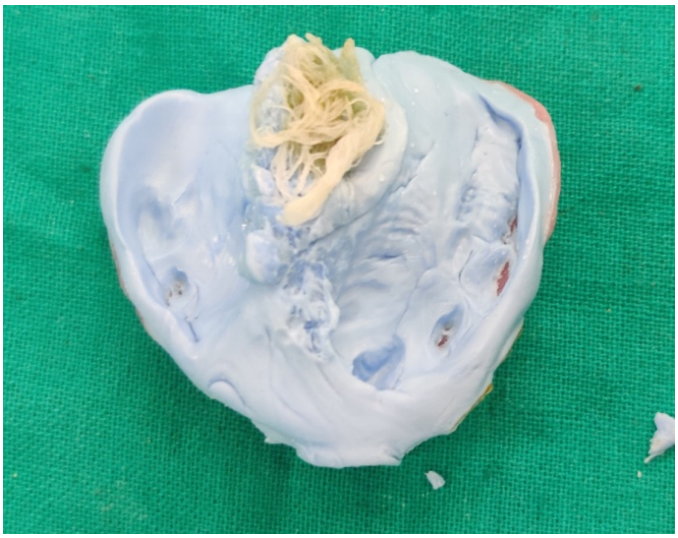

Fig 4: Beading and boxing was done and cast poured with die stone.

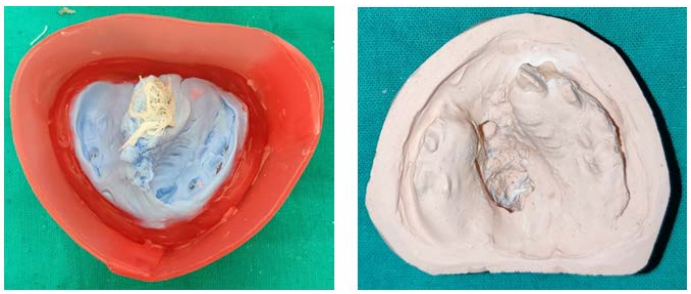

Fig 5: Prosthesis fabrication with autopolymerizing resin.

Fig 6: Prosthesis was secured extra-orally by surgical adhesive tape.