Dental Management for Patient with Kartagener’s Syndrome

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Abstract

Kartagener’s syndrome (KS) is a disease characterized by a triad of manifestations, which include, situs inversus (mirror image orientation of internal organs), bronchiectasis, and sinusitis. There is only one reported case for KS. Therefore, this case report aims to illustrate the oral findings of a pediatric patient with KS and to provide detailed management. Coordination and collaboration with the medical team by the systematic, proactive way rather than reactive way, will result in successful dental management and fewer complications. Management of the pediatric patient with Kartagener syndrome should be directed toward: 1- Maximize the efforts in behavior management techniques to perform the treatment on the dental chair to avoid possible respiratory complications related to general anesthesia. 2- Collaboration with medical healthcare professionals to identify and solve the predisposing factors for mouth breathing in patient with KS. 3- Expanding the efforts to identify and standardize the methods of early recognition of oral breathing in children and early management.

Keywords: Kartagener’s Syndrome (KS); Dental Management; Bronchiectasis; Sinusitis

Introduction

Kartagener’s syndrome (KS) is a disease characterized by a triad of manifestations. These manifestations include situs inversus (mirror image orientation of internal organs), sinusitis, and bronchiectasis [1-3]. The KS is included in a group of diseases characterized by primary ciliary dyskinesia (immotile cilia) [4-8]. The incidence of KS is reported at 1: 20,000 Caucasians, with no known racial predilection [1-3].

The KS is caused by an autosomal recessive gene defect; however, the specific gene conferring KS have not yet been identified [2,4]. The primary ciliary dyskinesia and associated disorders are characterized by abnormal cilia function, which will cause in sufficient mucociliary clearance, as well as defect in spermatozoa [5,6].

Since the early symptoms of KS are not totally distinguished, the diagnosis of KS is difficult. The first signs of KS are constant and difficult to treat nasal congestion or rhinitis that begins on the first days of life. Another sign is a persistent cough within the neonatal period. However, it is not conclusive signs of KS [7-9]. The diagnosis is usually done by studying mucociliary clearance and/or histologically through bronchial biopsy [4,5].

Because of compromised respiratory system, KS patients are at high risk of chronic respiratory infection, which can lead to severe respiratory problems in later life. Therefore, the treatment modalities for KS are directed at preventing the chronic airway infection by airway clearance with daily courses of physiotherapy. Up to our knowledge, there is only one reported dental case for KS. Therefore, the aim of this case report is to illustrate the oral findings of pediatric patient with KS and to provide detailed dental management [9].
**Case Description**

An 8-year-old female Asian was referred by the hospital of King Abdulaziz University for dental management. On presentation, the parent’s complaint was, ‘Our child has cavities, and we want to get them filled’. The patient never had any dental treatment previously. The patient was a known case of KS and diagnosed during the first year of life. The patient was suffering from frequent infections on the chest and otitis media since birth. She had the classical KS manifestations, including situs inversus, bronchiectasis, and sinusitis (Table 1). The family history revealed no history of KS and/or any other hereditary disease. Regarding the respiratory tract infection, it was treated with numerous courses of antibiotics. The antibiotics choices were varied, according to sensitivity tests carried out at that time, and were administered either intravenously or orally as an oral suspension.

<table>
<thead>
<tr>
<th>Summary of the medical history of the patient</th>
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<tr>
<td>Case of Kartagener’s syndrome (classic triad)</td>
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<tr>
<td>History of heart defect in birth repaired by its own, without surgical intervention.</td>
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<tr>
<td>Hospitalized three times for surgery (two ear tube and tonsillectomy)</td>
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<tr>
<td>Recurrent chest infections and otitis media.</td>
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<td>Frequent use of antibiotic</td>
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*Table 1*

There was negative history of abnormalities in the primary dentition. Regarding oral habits, the patient has a habit of thumb sucking since infancy and mouth breathing. The history of brushing was insufficient, and the diet was containing multiple snacks between meals. The patient was drinking bottle water on daily basis and no history of professional application of fluoride.

On clinical examination (Figure 1), no extra oral abnormalities were found. The patient was found to have angle class 2 molar relationships in both sides, and an anterior open bite with a posterior tendency for a cross-bite. Regarding periodontal health, the patient was found to have mild plaque induced gingivitis and stains. Regarding caries, the patient was found to have caries in the primary posterior teeth with badly decayed lower first primary molars. An orthopantomogram and Bitewing radiographs (Figure 2) showed the patient to be in the mixed dentition with developing first premolar, second premolars, canines and second molars. In the initial visit, the behavior of the patient was rated to be 4 (definitely positive) according to Frankle’s classification (1962).

*Figure 1: Pre-operative intra-oral photography and extra-oral picture.*

The final diagnosis and problems list were identified and formulated as, consideration of the medical condition consequences on the oral health status and dental management, inadequate oral hygiene, diet containing cariogenic food, multiple carious teeth, remaining roots, thump sucking habit, mouth breathing and open bite. The primary treatment plan objectives were to establish optimal oral health for the patient and to provide safe dental treatment. Consent was obtained and medical consultations were sent to the patient physicians and the recommendation was to administer daily low dose of prophylactic antibiotic during treatment course (20 mg/kg amoxicillin).

Therefore, antibiotic was prescribed and the patient was treated in four visit on the dental chair, using local anesthsia lidocaine 2% with Epinephrine 1:100:000 vasoconstrictor. The rubber dam was used in all restorative treatments procedures. The posterior teeth were restored with either Resin dental restorations or stainless steel crowns according to the extension of caries. The non-restorable posterior primary teeth were extracted and space maintainer was applied. Regarding thumb sucking habit, straight forward discussion was made with the patient and she convinced to quite the habit. Therefore, we decided to apply an appliance as reminder after full explanation to both the child and parents (Table 2 and figure 3).

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<thead>
<tr>
<th>Conservative/Medical Objectives</th>
<th>Medical consultation</th>
<th>Prophylactic antibiotic</th>
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<tr>
<td>Preventive Objectives</td>
<td>Oral Prophylaxis</td>
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<td>Topical Fluoride Application</td>
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<td>Restorative objectives</td>
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<td>Orthodontic Objectives</td>
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<td>Habit breaking appliance</td>
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<td>Orthodontic Consultation</td>
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*Table 2: The treatment approaches and objectives.*
Discussion

Coordination and collaboration with the medical team by the systematic, proactive way rather than reactive way will result in successful dental management and fewer complications. In our patient, different consultations were sent to the patient’s physicians with a detailed explanation of treatment, and they recommended to give a prophylactic antibiotic. The prescription of low dose prophylactic antibiotics during treatment was in agreement with a previous medical recommendation for the management of a patient with KS, as they are at higher risk of airway infection [9].

In regards to nonnutritive sucking, it considered normal in infants and young children, and other than anticipatory guidance interventions are usually not recommended prior to eruption of permanent teeth [11]. However, our patient was having the habit until the age of eight. In addition, the habit effect was superimposed by the mouth breathing, and both resulted in unfavorable changes on the dentition. Therefore, a straightforward discussion was made with the patient and a reminder was applied to remind the patient until she quit of the habit.

Although, our patient was referred for orthodontic management, but there is evidence that, there is relationship between the open bite that caused by mouth breathing, and chronic nasal and sinus obstruction disease. This has explained the high incidence of anterior open bite in patients suffering from cystic fibrosis [10]. The apparent cause of this habit in our patient is the airway disorder. Although, our patient was on a twice daily hypertonic saline 7% to clear the nasal airway from secretions, but it was not enough to prevent the oral breathing, which may be explained by the respiratory insufficiency.

The respiratory insufficiency was confirmed by using the pulse oximetry, and it shows 97% oxygen saturation in the dental visit. The early diagnosis and detection of mouth breathing in children is more challenging compared to adult. This is because that in adult the skeletal features of the face and jaw are already apparent. Pediatric dentists may be the first healthcare professionals to have contact with a
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mouth breathing child. Although, many articles described the consequences of mouth breathing, few studies investigated the key parameters for clinical recognition of mouth breathing, especially in children. Therefore, we recommend expanding the efforts to standardize the methods of early recognition of oral breathing in children and early management.

Conclusion

In conclusion, the management of pediatric patient with Kartgner syndrome should be directed toward:

1. Maximize the efforts in behavior management to carry the treatment on the dental chair to avoid respiratory complications related to general anesthesia.

2. Collaboration with medical healthcare professionals to identify and solve the predisposing factors for mouth breathing in patient with KS.

Bibliography


