

# **ABSTRACTS FOR ORAL CASE REPORT PRESENTATION**

## **CASE REPORT 1**

### **Management of Self-Mutilation Patients with Lesch-Nyhan Syndrome: Two Case Reports**

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**Introduction:** Lesch-Nyhan Syndrome (LNS) is a rare X-linked disorder caused by mutation in HPRT1, an important enzyme in the purine salvage pathway. Symptoms include overproduction of uric acid, dystonia, gout, intellectual disability, and self-mutilation. This report describes two patients diagnosed with LNS who came to us for management of self-inflicted oral injuries. Both patients' parents agreed that a conservative approach, avoiding teeth extraction, should be followed initially before any radical intervention.

**Case Reports:** We report two intriguing cases of two 3-year-old boys diagnosed with LNS who were referred to us for management of extensive traumatic oral ulcerations in 2018 & 2019. After a thorough discussion with both parents, removal dental appliances were fabricated for them. Parents were instructed and trained on insertion and anchorage, removal, cleaning of the appliances. One week later, frequencies of self-mutilation have improved and healing of lesion was observed. However, one patient subsequently damaged his appliance repeatedly and developed recurrent oral ulceration. After much consideration, his parents agreed for dental extractions under general anesthesia.

**Conclusion:** It is important to know about LNS and the effects of self-mutilation on the family, in order to know how and when to treat, thus giving patients and their family the best possible quality of life. Oral appliances represent a conservative approach to the issue and should be tried initially before resorting to dental extractions.

## CASE REPORT 2

### **Non-Syndromic Multiple Supernumeraries In Permanent Dentition: A Multidisciplinary Approach**

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**Introduction:** Non-syndromic multiple supernumeraries (NSMS) is a rare condition that usually affects male which can lead to various dental problems including tooth displacement, delayed eruption and malocclusion.

**Case Report:** This case describes the management of malocclusion caused by multiple supernumeraries (MS) in the premaxilla region of a healthy 9-year-old boy. Intraoral examination showed severely crowded upper incisors with the presence of five MS teeth causing impacted tooth 21, traumatic crossbite between a supernumerary and tooth 31 and palatal displacement of tooth 2. Two of the supernumeraries were of supplemental type resembling the maxillary lateral and central incisors. The teeth were differentiated with the use of cone beam computed tomography. After orthodontic consultation, extraction of multiple supernumeraries was performed under local anaesthesia. Tooth 21 was spontaneously erupted shortly after and simple orthodontic treatment was used to correct the crossbite and realign upper anterior teeth.

**Discussion:** NSMS usually occurs bilaterally and is often associated with hereditary factors. In this case, the supernumeraries erupted in the premaxilla region crossing the midline. Hereditary factors may have played a role as his elder brother also had a history of a single mesiodens.

**Conclusion:** A multidisciplinary approach is important in managing this case. The use of 3D analysis helped in deciding the teeth that need to be extracted and malocclusion was corrected using simple sectional orthodontics.

#### **Lesson Learnt:**

- 1) MS can lead to malocclusion and tooth impaction which need a multidisciplinary approach.
- 2) Prompt management of supernumerary teeth obstructing the eruption of permanent teeth is crucial to prevent further complications (impaction/cyst/resorption).

### **CASE REPORT 3**

#### **Too Little, Too Late...**

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**Introduction:** Rickets is thought to be a rare occurrence in Malaysia, where resources are adequate and easily accessible and there are abundant of sunlight all year long.

**Case Report:** A 3-year old Malay girl referred to us for dental management as she was diagnosed with nutritional deficiency rickets. She appears to be of a shorter stature and has bowing deformity on both legs. Laboratory investigations revealed low serum 25-hydroxyvitamin-D, serum calcium levels, high levels of serum parathyroid hormone and serum alkaline phosphatase, which are classical features of Vitamin-D deficiency rickets. Intraorally, there are yellowish-brown pitting over the labial surfaces on her upper teeth. Hypomineralisation is also present at the cervical region of molars. There is no evidence of tooth wear; all teeth are sound. Her general condition improved remarkably after treatment with oral vitamin D over a period of 1 year.

**Discussion:** Nutritional rickets caused by deficiency of Vitamin D, calcium or phosphate may cause hard tissue malformation. In this case, the long bone and dental developmental defects were present.

**Conclusion:** Long term follow-up, monitoring caries risk status in this child as progression of caries is expected to be faster in hypomineralised teeth. Aggressive preventive strategies should be applied to arrest existing carious process and to prevent development of new caries.

**Lesson Learnt:** Rickets, dental developmental defects

## CASE REPORT 4

### **Therapeutic Challenges in Management of Familial Oligodontia: Exploring Horizons in Dental Rehabilitation.**

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**Introduction:** Oligodontia can present with multiple anomalies of teeth, periodontium and jaws such as crown malformations, enamel hypoplasia, periodontal involvement, short roots etc., thereby complicating the oral rehabilitation of such patients.

**Case Report:** An adolescent Chinese boy presented with spacing in upper teeth. Examination showed mixed dentition with eight over-retained deciduous teeth and 14 unerupted permanent. Straight facial profile and normal skeletal dental base relations were noticed along with class III molar relation and edge-to-edge bite. Further evaluation showed that his sister and father had similar findings.

**Discussion:** Orthodontically, the case was quite challenging since multiple permanent teeth were missing and to reduce the inter-bracket span, primary teeth had to be engaged with utmost caution not cause more resorption of the roots. Hence, desired and controlled tooth movement was tough to achieve.

**Conclusion:** Dentists can play an important role in patients with oligodontia to “unearth” associated syndromes/systemic disorders which would have not caught attention elsewhere.

#### **Lesson Learnt:**

- Thorough clinical examination of oral cavity and various body systems along with panoramic radiographs, genetic tests and blood/biochemical evaluations is of utmost importance to rule out syndromes/systemic disorders and aid in accurate diagnosis.
- A good dental health team comprising of orthodontists, oral surgeons, implantologists, prosthodontists as well as pediatric dentists can help in complete oral rehabilitation.
- Various treatment options must be discussed with the patient/parent and decided upon before embarking on it

## **CASE REPORT 5**

### **Non-Invasive and Invasive Method in Diagnosing Tongue Lymphangioma in A Child: A Case Report**

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**Introduction:** Lymphangiomas are benign, hamartomatous tumors of lymphatic vessels that usually present at birth but they may go unnoticed until later in life. It is commonly located at head and neck, rarely in oral cavity. The common intraoral locations are dorsum of tongue, palate, buccal mucosa, gingiva and lips. Clinically lymphangioma presented as localized or diffused growth which may enlarge causing difficulties in speech, mastication or breathing.

**Case Report:** A 9-years-old boy presented with multiple reddish pink papillary and vesicles-like projection at mid dorsum of tongue for 4 years. It is associated with burning sensation especially during spicy food consumption. MRI was carried out as initial investigation, however no definite mass is detected. Incisional biopsy was performed and histopathological examination revealed lymphangioma.

**Discussion:** MRI is useful in evaluating extension of lesion, especially in cases of extensive lymphangioma with cystic spaces, or cystic hygroma. It is unlikely to be used in small oral lymphangioma especially in children. In most cases, oral lymphangioma is confirmed by incisional biopsy.

**Conclusion:** This case emphasizes the importance of investigation process to confirm the diagnosis which leads to proper management for oral lymphangioma.

#### **Lesson Learnt:**

There is limitation of MRI usage in detecting small lesion on tongue especially in children. In such case, incisional biopsy is recommended to confirm diagnosis of tongue lymphangioma.

## **CASE REPORT 6**

### **MIH: TO SMILE OR NOT TO SMILE? A CASE REPORT**

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**Introduction:** Extensive dental caries involving upper permanent incisors is rare in teenagers. More likely, predisposing enamel hypoplasia or hypomineralization increase the risk, ensuing in isolated carious lesions on these teeth. Diagnostically, it is challenging to distinguish the aetiology of these defects which can range from Turner's hypoplasia to molar-incisor hypomineralization (MIH). In Malaysia, MIH prevalence ranges from 14-16% , while globally it ranges between 2.4-44%. An individual affected with MIH is known to have increased risk of dental caries which can impair their quality of life.

**Case Report:** A 15-year-old girl presented with discoloured upper central incisors and pulp polyp on lower first permanent molars. Clinical examination revealed extensive enamel structural defects with dentin exposure complicated with underlying sensitivity and inflamed marginal gingiva of tooth 11 and 21. The upper central incisors were temporarized using glass ionomer cement to overcome sensitivity and allow healing of the surrounding gingival tissues. Home care measures were reinforced. Upon review, the healing of the gingiva showed remarkable changes. Tooth 11 and 21 was subsequently restored conservatively with glass ionomer and composite resin in layering technique.

**Discussion:** MIH presentation varies from simple opacities to severe forms. Post eruptive breakdown is evident in severe cases where dentin is exposed. The available treatment modality ranges from preventive, restorative to long-term maintenance.

**Conclusion:** Early detection and adoption of preventive measures is crucial for preservation of tooth structure.

**Lesson Learnt:** Early recognition of MIH could avoid compromising a patient's quality of life.

## **CASE REPORT 7**

### **Dentofacial Anomalies In Axenfeld-Rieger Syndrome: A Case Report**

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**Introduction:** Axenfeld-Rieger syndrome (ARS) is a rare autosomal dominant disorder characterized by ocular, craniofacial, dental and periumbilical abnormalities. Dental manifestations commonly reported include hypodontia, microdontia and taurodontia. Early diagnosis, a multidisciplinary approach and lifelong monitoring are vital in providing effective management of this challenging disorder.

**Case Report:** A 2 year-8-month-old girl was referred for unerupted front teeth. Her medical history then was non-contributory. However, she presented with facial dysmorphism, blue sclera and clinically missing 3 primary teeth. Radiological examination of the dentition revealed absence of most permanent tooth germs. Referral to the genetic department led to the diagnosis of ARS based on the clinical manifestations including, most importantly the congenital iris abnormalities. Her parents were counselled regarding the condition and further genetic testing was performed. Current dental management for this patient includes strict preventive measures and restorative management of her precious primary teeth. Following multiple medical interventions, she became apprehensive towards health treatment thereby complicating her dental care.

**Discussion:** Care of syndromic patients commonly necessitates multidisciplinary care and good communication. Fortunately, our patient was diagnosed early due to attentive dental personnels as well as proficient medical providers. Multiple ocular presentations have since emerged and more challenges are expected in the future, including dental issues that will require functional as well as aesthetic interventions.

**Conclusion:** Management of ARS requires complicated, long term medical and dental care. Early diagnosis has assisted in the prognosis of her ocular and dental issues.

**Lesson Learnt:** Hypodontia is not uncommon but when involving both dentitions, may indicate a serious condition such as ARS.

## **CASE REPORT 8**

### **Multidisciplinary approach of an adolescent with infraoccluded primary molars and oligodontia: A case report.**

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**Introduction:** Management of infra-occlusion with oligodontia may pose some challenges and requires a multidisciplinary approach. Oligodontia is a rare condition that usually occurs as part of a syndrome and seldom exists alone.

**Case Report:** A fit and well 16-year old Malay boy was referred for multiple missing teeth. He has missing maxillary first premolars and all mandibular premolars with retained mandibular second primary molars and left mandibular first primary molar. Right mandibular first primary molar was extracted few years ago in primary dental care and bone loss noted. The primary molars are severely infraoccluded. He also complains of “discomfort when eating on the right side of the mouth”. Radiographic findings showed enamel caries on all infraoccluded teeth and the retained primary molars were ankylosed. There is no family history of missing teeth. He was seen in Joint clinic and the agreed treatment plan involved restoration of the retained primary molars with indirect onlays, orthodontic treatment to distribute space and prostheses later. He also been referred for genetics counselling on parents’ consent.

**Discussion:** Hypodontia can be associated with a syndrome or as non-syndromic. The genes mutation that have been identified associated with oligodontia are MSX1, PAX 9 and AXIN 2.

**Conclusion:** Effective treatment plan of patients with oligodontia requires multidisciplinary approach to develop appropriate long-term treatment plan. Genetic counselling may beneficial for the patient to identify any genes mutation or syndromic involvement.

#### **Lesson Learnt:**

- Different treatment options on management of infraoccluded teeth,
- Complications and treatment challenges in patients with oligodontia and infraocclusion
- Holistic treatment approach includes genetic testing

## **CASE REPORT 9**

### **Osteomyelitis Of Mandible Secondary To An Unusual Cause: A Case Report**

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**Introduction:** Osteomyelitis is a bone disease with multiple etiologies. Here we describe an interesting case report of a mandibular osteomyelitis of an unusual origin.

**Case Report:** A healthy 15-year-old Indian girl presented in August 2018 with recurrent pain in the region of 35 and 36 for about 2 months. Clinical examination revealed mild left extraoral mandible swelling with the left buccal bone expansion without any evidence of odontogenic infection. Radiological finding was suggestive of osteomyelitis of left mandible. The diagnosis was further confirmed by histopathological examination following a biopsy, performed under general anaesthesia. Initial resolution of the lesion was remarkable with administration of antibiotics. However, she presented with recurrent pain on the left mandible shortly after the completion of antibiotics, with generalized areas of radiolucency involving her entire mandible. This necessitated haematological examination to rule out systemic bone disease which showed significant deficiency in Vitamin D. She was then commenced on a Vitamin D supplement under the care of an endocrinologist. The patient had since been asymptomatic with significant mandibular bone healing.

**Discussion:** Vitamin D is an essential component for intestinal absorption of calcium, magnesium and phosphate. A fall in vitamin D could lead to calcium insufficiency and an increase in the susceptibility to infection leading to impaired bone mineralization.

**Conclusion:** The occurrence of osteomyelitis though, is an uncommon feature in Vitamin D deficiency, but cannot be overlooked.

**Lesson Learnt:** A systemic cause should always be investigated if an osteomyelitis is not responding to the conventional therapy.

## **CASE REPORT 10**

### **Morquio Syndrome (MPS Type IV A): It's Challenges in Dental Management**

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#### **Introduction:**

Morquio syndrome (Mucopolysaccharidosis type IVA) is an autosomal recessive lysosomal storage disorder (LSD) caused by deficiency of Nacetylgalactosamine-6-sulfate sulfatase (GALNS) and cause accumulation of specific glycosaminoglycans (GAGs) mainly in cartilage and extracellular matrix (ECM) including teeth.

**Case Report:** A case of 6.2 years old healthy girl, referred to Paediatric Dental team for dental assessment and management. Patient was diagnosed by Geneticist with Morquio syndrome type IV A and under management and follow-up with Paediatric team. Clinical examination showed marked short stature, protrusion of the chest, kyphoscoliosis, platyspondyly, coxa valga, abnormal gait, laxity of joints, stubby hands and generalized hypoplastic primary dentition. Radiographically, she presented with normal presentation of permanent teeth according to her chronological age. Comprehensive dental treatment including stainless steel crowns, composite restorations and extractions performed.

**Discussion:** Further monitoring is still needed to assess the eruption status of permanent dentition and future treatment needs.

**Conclusion:** Early assessment and management of generalized hypoplastic primary dentition may contribute to beneficial effects to future eruption of permanent dentition.

#### **Lesson Learnt:**

1. Morquio syndrome might affect dentition hence, dental assessment is mandatory in every patient.
2. Early treatment in hypoplastic primary dentition prevent progress of the condition and further reduce risks of treatment under general anaesthesia.

## **CASE REPORT 11**

### **Minimally Invasive Approach in Aesthetic Management of Hypomineralised Incisors**

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**Introduction:** Molar Incisor Hypomineralization (MIH) is a developmental defect of enamel affecting first permanent molar and frequently incisors. The characteristic appearance of well-demarcated opacities and discoloration ranging from whitish cream to yellow brown results in aesthetically compromised maxillary incisors.

**Case Report:** A 14 years old girl presented with white patches on upper front teeth since eruption. She went to general practitioners who reassured her to accept the colour. She has no complaint of pain and sensitivity but alleged being teased at school. Clinical examination revealed well demarcated whitish cream opacities on maxillary right and mandibular left first permanent molars, maxillary right and left central incisors and mandibular right lateral incisor. The anterior teeth were clearly visible upon smiling. Diagnosis of Molar Incisor Hypomineralization (MIH) was made. Treatment plan discussed and accepted was resin infiltration.

**Discussion:** Discolouration in children and adolescence is often perceived least important and poses challenge due to large immature pulp. This contraindicate use of invasive technique such as composite restoration and veneer. Studies show that adhesion is inferior in hypomineralized enamel potentiate restoration failure. Resin infiltration is a minimally invasive, fast and well accepted. This report demonstrated the pleasing aesthetic outcome of resin infiltration (ICON®) for masking discoloration.

**Conclusion:** Predictable result and conservative approach of resin infiltration provide excellent option in aesthetically compromised hypomineralized incisor.

#### **Lesson Learnt:**

- Clinician should be attentive in teeth discolorations in children and adolescent especially when patient desire treatment.

## **CASE REPORT 12**

### **Case Report: Clinical Presentation And Orthodontic Management Of Siblings With Van Der Woude Syndrome**

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**Introduction:** Van Der Woude syndrome (VWS) is a rare autosomal dominant congenital disorder. Its distinctive clinical presentation is congenital pits on lower lip that appear on paramedian portion of the vermilion border of the lip. This occurs in concurrence with cleft lip and/or cleft palate.

#### **Case Report:**

Case 1 :

18 year old male young adult presented with VWS characterized by bilateral lip pits, left unilateral cleft lip and palate, and congenitally missing 22 and 35.

He was first treated at age 11, ABG which was done in 2014. Bonded retainer was placed and prosthesis was done to replace 22.

Case 2 :

The younger sibling, a 13 year old teenage girl presented to our clinic with VWS, also characterized by bilateral lip pits, and left unilateral cleft lip and palate and congenitally missing 22.

She first attended clinic at 11 years old and ABG was done in 2017 prior to orthodontic treatment.

**Discussion:** A rare congenital disorder that occurs only in 1:40000 and 1:100000 was seen in siblings of different sex.

#### **Conclusion:**

Orthodontic management of VWS siblings did not present any complication and their malocclusion were treated by alveolar bone grafting and fixed appliance.

**Lesson Learnt:** There are varied presentations of VWS and in this case we have observed lip pits as distinctive feature. In the literature there were problems of mucous secretion from the pits that may require surgical correction. Risk of inheritance from parent with lip pit and cleft is 39%, may be increased as the parents' sibling also presented with the same feature.

## **CASE REPORT 13**

### **Hypodontia : A Multidisciplinary Approach**

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**Introduction:** Children with hypodontia require treatment planning incorporating the aesthetic as well as functional aspects as research demonstrates missing teeth can negatively impact their self-esteem. This case report highlights the complexities of managing hypodontia in an 8 year old girl.

**Case Report:** PS an 8-year-old girl presented in with a complaint of “hating the look of her teeth”. She had multiple retained and carious primary teeth, infraoccluded 84, clinically missing 11 and 21. Radiographic examination revealed hypodontia of 15,35,31, and 41 as well as diminutive 12 and 22. A treatment plan was formulated involving targeted prevention and quadrant dentistry under local anaesthesia emphasizing on treatment of the carious primary teeth and later on composite build-ups on her 71,81, 12 and 22. Inhalation sedation was utilized to complete the course of dental treatment.

**Discussion:** The gene associated with incisor-premolar hypodontia had been hypothesized to be linked to diminutive maxillary lateral incisors. PS was teased at school due to appearance of her teeth. A decision was made to build-up 12,22,71 and 81 with composite after seeking input from the orthodontist as well as restorative specialist.

**Reflective learning:** Having to provide a treatment plan for a child with hypodontia often requires good communication between members of the multidisciplinary team as the treatment plan is often provisional and prone to change as the child grows.

**Conclusion:** As a paediatric dentist in a multidisciplinary discussion, we play a vital role in forming a holistic treatment plan that addresses the long-term outcomes and the child’s current complaint as well.

**Lesson Learnt:** If I could do things differently I would consider incorporating the composite build-ups from the initial treatment plan preventing her distress. However, at the end of treatment, PS was very happy with her smile and was no longer embarrassed by it.

## **CASE REPORT 14**

### **Ehlers-Danlos Syndrome: A case of catastrophic bleed from lip injury**

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**Introduction:** Ehlers- Danlos Syndrome (EDS) is a group of rare inherited disorders of the connective tissue presenting as generalised hypermobility of joints and loose, fragile, stretchy skin that bruises easily due to weakened connective tissue.

#### **Case Report:**

A 5-year-old boy presented to our Emergency Department (ED) with laceration wounds over the left angle of mouth left buccal mucosa after a fall. Suturing was done under local anaesthesia. The next day and day four of review the wound healing appeared normal. However, at day 10, the child was brought to ED with profuse bleeding from the previous sutured site. On presentation he was weak, pale, tachycardic with low pulse volume and BP of 70/40mmHg consistent with haemorrhagic shock. His estimated blood loss was 300cc. Resuscitation fluids were given and emergency haemostatic suturing was performed under local anaesthesia. He was given blood transfusion and subsequently resuturing with cauterisation of active bleeder vessel was done under general anaesthesia. He was managed as inpatient and discharged well post-operative on day 5. There was uneventful outcome on follow-up.

**Discussion:** Minor trauma in patients with EDS, can easily cause splitting of the dermis due to the fragility of skin and result in poor wound healing. It can also cause unpredictable tearing (rupture) of blood vessels that can lead to internal bleeding and other potentially life-threatening complications.

**Conclusion:** As collagen formation is affected, healing is compromised thus close follow up is necessary to prevent a catastrophic bleed.

**Lesson Learnt:** Due to the increase vascular fragility, it is important to avoid excessive manipulation of surrounding tissue to reduce risk of further breakdown and bleed.

## **CASE REPORT 15**

### **Management of A Severely Dilacerated Maxillary Central Incisor Complicated by Hypodontia**

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**Introduction:** Unerupted upper anterior teeth produces serious aesthetic and functional problems in a growing child. Dilacerated tooth is managed by either surgical exposure and traction, or surgical removal of the tooth based on the prognosis for proper alignment and long-term survival of the tooth.

**Case Report:** GXY, a 10-year-old Chinese girl, was referred for orthodontic management of a partially erupted and dilacerated upper right central incisor (UR1). She had history of trauma to her upper deciduous incisors at age 1 ½ years old. GXY presented in mixed dentition with a Class III malocclusion on a Class I skeletal pattern complicated by a partially erupted and dilacerated UR1 with the crown rotated 180°. In addition, the upper permanent canines were congenitally missing. Radiographs showed UR1 was severely dilacerated and malformed, with defective root formation.

**Discussion:** Although the prognosis of UR1 was poor, but due to her young age and hypodontia, the UR1 was surgically exposed and gold chain traction was carried out to allow the alveolar bone development. This case report illustrates the benefit of early surgical and orthodontic intervention of a severely dilacerated incisor and interceptive measures taken to maintain the upper deciduous canines whilst further facial growth and dental development occurs.

**Conclusion:** Timely interceptive multidisciplinary approach is crucial in the management of complex malocclusions involving dental anomalies. Early exposure and gold chain traction of a tooth of poor prognosis into the arch will encourage proper dento-alveolar development and function which will facilitate implant supported prosthesis later on.

**Lesson Learnt:** Multidisciplinary management of a complex malocclusion if carried out at the optimal timing can benefit the patient and improve her self-esteem during her growing years whilst optimising the dento-alveolar development and function at the same time.