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Case Report

Dentistry Section

Management of Dentoalveolar Fracture in a Patient with Wilson's Disease: A Case Report

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ABSTRACT

Dental trauma is most common in children as they are involved in a lot of physical activities as they grow up. Most commonly dental trauma is seen in front teeth in the upper jaw. Children with disabilities have to be given more attention as they are more prone to dental injuries and infections. Several genetic disorders affect an individual mentally and physically and make them disabled. One such autosomal recessive inherited disorder is Wilson's disease. Wilson's disease is characterised by excessive deposition of copper in the liver, brain and other tissues. It is usually diagnosed between ages 5 to 35 years of age. Dentoalveolar fracture is seen in 17% of children. Dentoalveolar fracture involves the alveolar bone and may extend beyond the alveolus. This article presents a case report of a 10-year-old male, who presented with a dentoalveolar fracture of the lower jaw and was also diagnosed with Wilson's disease. In this case, dentoalveolar fracture of the lower mandibular anterior region is managed by ribbond splinting as it is free of any metal/copper contents and is most compatible in this case. As paediatric dentists, it is essential to have the skills and techniques to manage patients with special abilities and make the right choice of treatment and materials which adds benefits to patients' well-being. Patients of such conditions are of utmost importance as they are prone to trauma and infections.

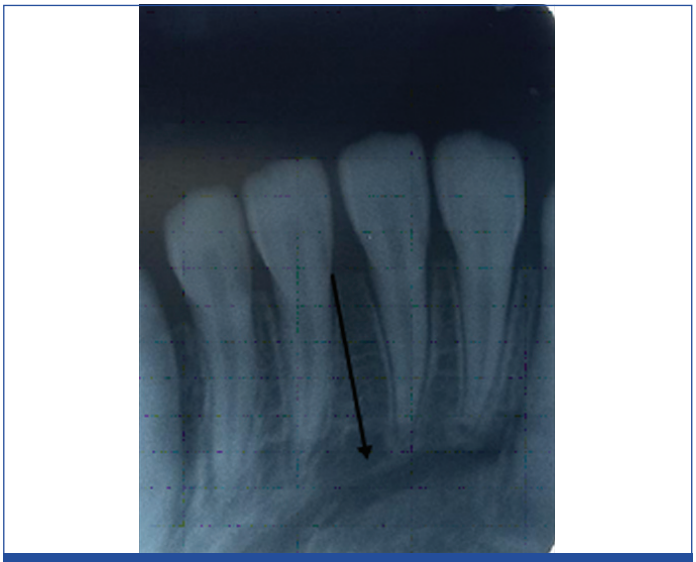
Keywords: Dental trauma, Mandibular fracture, Ribbon splint

CASE REPORT

A 10-year-old patient reported to the Department of Paedodontics and Preventive Dentistry, with wounds and lacerations on his lower lips. Patient's parent informed about history of fall while undergoing physiotherapy sessions. The medical history of the patient disclosed that he is a known case of Wilson's disease and is under medication. The patient was normal till the age of six years. Slowly patient slowed down in academics and his intellectual, physical and mental abilities were low. He was soon diagnosed with Wilson's disease. Wilson's disease is characterised by excessive deposition of copper in the liver, brain, and other tissues. The patient had an abnormal gait, and problems with speech, swallowing, and physical coordination.

On extraoral examination, the patient was not able to keep his lips competent and was constantly drooling. Swelling and laceration of the lower lip were also seen. On intraoral examination, there was segmental mobility in the lower front tooth region of the jaw. Pool of saliva on the floor of the mouth. There was mobility (Grade II) in 43, 42, 41, 31, 32, 33 along with alveolus. The patient was unable to occlude. Patient's oral hygiene was fair. Segmental fracture of the lower anterior region was suspected. A radiographic evaluation was advised for the patient.

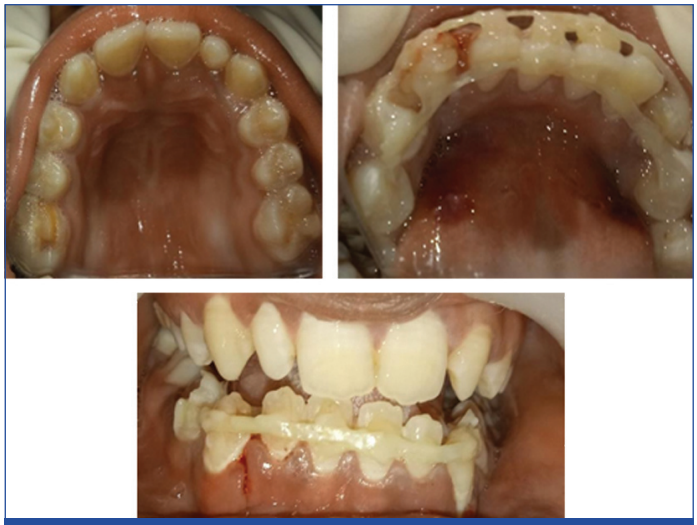
An intraoral periapical radiograph revealed a dentoalveolar fracture of the mandibular anterior region of the jaw [Table/Fig-1]. OPG was attempted, but not successfully taken as the patient was not able to stabilise in the required single position because of pain. As the patient was already diagnosed with Wilson's disease, any dental material containing copper like dental amalgams, porcelain fused crowns, or partial denture attachments were contraindicated in the patient. The daily copper intake in a patient was strictly restricted to 1.5 mg. The segment was repositioned and splinting was planned for the patient, to stabilise the lower anterior segment. As other materials were restricted in this case, Ribbond splinting of the teeth for four weeks following the guidelines of International Association of dental traumatology [1,2] as considered to be the best option. No other prosthetic appliance was used. Ribbond splint (INTERIOR) strips (8.5 cm × 2.0 mm × 0.2 mm) along with the fibre-reinforced flowable composite (Ivoclar Vivadent Tetric N-cream syringe) was used to stabilise the lower anterior teeth facially and lingually. The procedure



[Table/Fig-1]: Pre-operative radiograph indicating the fracture line.

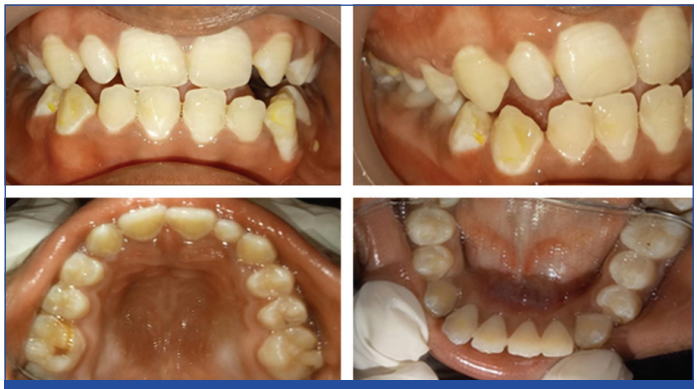
was carried out with the utmost care and isolation as much as possible [Table/Fig-2]. The patient was instructed to shift to a soft and liquid diet for at least two weeks. Rinsing with chlorhexidine daily after each meal was instructed to patient's parents to maintain oral hygiene. The patient was recalled the next day to ensure that he had no high points or difficulties with the splint. A splint was placed for four weeks until the teeth, alveolus, and segment were stabilised.

After four weeks, the patient was recalled. He exhibited significant clinical improvement, with notable healing of lower anterior region. The splint was removed, occlusion was checked. Oral prophylaxis was carried out on both the arches [Table/Fig-3]. Fluoride application was done on both maxillary and mandibular arches. Radiograph was not taken at the follow-up as the patient was not co-operative for any Orthopantomogram (OPG)/occlusal radiography procedures. However, the patient's oral health showed marked improvement, stable occlusion and complete resolution of pain and discomfort. Clinical evaluation at three months revealed excellent healing, optimal oral hygiene, and intact occlusion.



[Table/Fig-2]: Immediate postoperative pictures after Ribbond splinting on buccal and lingual aspect.

Follow-up:



[Table/Fig-3]: Four weeks follow-up- pictures right after splint removal showcasing satisfactory treatment results.

DISCUSSION

Wilson's disease was described by Kinnier Wilson in 1912, who named it "hepatolenticular degeneration" [3]. A prevalence rate of 30 cases per million (or one per 30,000) and a birth incidence rate of one per 30,000 to 40,000 are often quoted [4]. A mutation in the ATP7B gene, located on chromosome 13, is responsible for Wilson's disease [4,5].

Copper is an essential element for cellular function, but free copper is extremely toxic and can produce irreversible damage. In normal conditions, both the ATP7B protein and ceruloplasmin are involved with copper transport. Under high copper conditions however ATP7B is also redistributed to cytoplasmic vesicles where it transports excess copper across the hepatocyte apical membrane into the bile canaliculus for subsequent biliary excretion. In individuals with Wilson's disease, the mutation in the ATP7B gene results in defective ATP7B protein that cannot perform these functions [4,5]. Not only does this progressive copper accumulation ultimately compromise hepatic function, but the hepatic storage capacity is also eventually exceeded and unbound copper spills out of the liver and is deposited in other organs and tissues, where it also provokes damage and dysfunction. As the excess copper escapes from the liver, urinary copper excretion rises dramatically but is unable to compensate fully for the defect in biliary excretion. It has been assumed that the cellular damage characteristic of Wilson's disease is due to a direct toxic effect of excess copper [1,6].

The onset of Wilson's disease as early as four years of age and as late as the fifth decade of life has been reported. The initial sign of nervous system involvement is typically a decline in cognitive function. Patients may experience difficulty focusing on tasks, though their overall cognitive abilities are not usually severely impaired. Over time, patients might develop depression, become withdrawn, and show signs of motor slowing, reduced facial expression, and slurred speech. A noticeable tendency to keep the mouth slightly open may also be observed. In cases where symptoms begin in childhood, dystonic and athetoid movements are common, whereas those who experience symptoms later in life typically develop dysarthria and hand tremors. As the condition progresses, muscular rigidity and bradykinesia may emerge, eventually leading to a clinical picture similar to Parkinson's disease. Wilson's disease is a significant cause of juvenile Parkinsonism. A characteristic feature seen in patients with neurological involvement is the presence of a Kayser-Fleischer ring, a golden brown pigmentation around the outer edge of the cornea [1,6].

Medical management generally involves having a restricted diet copper removal using chelating agents such as penicillamine [7]. Lab investigations like complete blood counts, standard biochemical profiles, and urinalysis should be performed at weekly intervals for a month, followed by two-week intervals for two to three months, then at monthly intervals for three to four months, and at four- to six-month intervals thereafter [7].

Dental management also involves lab investigations such as complete blood counts, liver function tests, and kidney function tests should be carried out before general anaesthesia or surgical procedures with proper physician consent [7]. Most of the amide local anaesthetics used in dental practice undergo biotransformation in the liver. Agents, such as sedatives and general anaesthetics, are potentially dangerous in liver disease mainly because of impairment of detoxification. Therefore, less hepatotoxic agents, e.g., enflurane, sevoflurane can be used. The drugs metabolised by the liver, e.g., fluconazole and paracetamol should be used in decreased dosage as at higher doses these drugs are hepatotoxic [7]. Antibiotics and nonsteroidal anti-inflammatory drugs and tetracycline are avoided if possible. Orthostatic hypotension and rigidity are common in these patients. To reduce the likelihood of a fall from the dental chair, the patient should be assisted to and from the dental chair. Because of dysphasia and an altered gag reflex, the patient must be treated in an upright position. Special precautions must be taken to avoid the aspiration of water or materials used during dental procedures. At the end of each appointment, the chair should be inclined slowly to allow for re-equilibration [7]. Wilson's disease is a form of copper poisoning and therefore the role of the dentist is to minimise the use of copper containing dental materials and medicines. In these patients, there is impaired healing and if the treatment involves the use of orthodontic appliances, removable appliances should be preferred than fixed. Use of nickel-titanium (Ni-Ti) wires should be especially avoided because although in traces one of the components of these wires is copper [7].

In the present case, the complete evaluation of the patient was done by a physician and consent was taken for the splinting procedure. Patient was appointed in the morning with all his medication taken one hour prior to the procedure. Ribbond splint was used labially and lingually with flowable composite as the patient had restrictions of copper content materials. In the present case, segment mobility was seen in the lower anterior region of the jaw. So, the first step is carried out to reduce the segment and bring it into occlusion under local anaesthesia. Further, as the patient was on a restricted diet, ribbond splitting was carried out and was under keen observation for four weeks. Following four weeks of stabilisation, once the lower anterior segment was stabilised, the splitting was removed, preventive therapy like oral prophylaxis was carried out and fluoride application was done. In conclusion, a splinting technique in our patient proved successful, with the resolution of the dentoalveolar fractures and recovery of good aesthetics in the anterior sector. An overview of cases of dental management in Wilson's diseases from the literature has been tabulated in [Table/Fig-4] [8-10].

Author name	Chief complaint	Clinical presentation	Diagnostic investigations	Dental management and material used
da Silva Santos PS et al., [8]	The patient had been referred to the department of oral surgery for the planned extraction of teeth and to enhance overall oral health	poor oral hygiene, periodontal disease, missing teeth, and several carious teeth	Lab work including complete blood count, comprehensive metabolic panel test, thyroid stimulating hormone, Vitamin 6 and Vitamin B12	Oral prophylaxis, filling of carious tooth with glass ionomer cement No teeth were replaced
Pandyan DA et al., [9]	Referred to the department of oral and maxillofacial surgery for the extraction of upper right third molar	Decayed upper right 3rd molar	Liver function test, complete blood count, magnetic resonance imaging and ultrasound	Extraction was carried out under strict aseptic condition with a minimal dose of 1:200,000 of lignocaine with adrenaline
Green Mw et al., [10]	Oroantral fistula following the removal of an impacted maxillary third molar	After extraction of impacted third molar oroantral fistula was developed	Nil	Antibiotics, decongestants, irrigation, and surgical closure

[Table/Fig-4]: Overview of management of Wilson's disease with other dental conditions [8-10].

CONCLUSION(S)

The management of dental trauma in special children is a challenge to all pedodontists. In the present case report, the patient presented with the known condition of Wilson's disease, which is extremely rare one in 30 million population. The proper knowledge of the rare medical condition, the trauma condition, keen observation and the ability to make the right decisions' for the benefit of the patients is necessary.

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