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Case Report
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Diabetes Insipidus Associated with Localized Periodontitis in 10 Year Old Child: a Case Report

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ABSTRACT

Hereditary diabetes insipidus is a rare endocrine disorder caused by a deficiency of the anti-diuretic hormone vasopressin. The disease is characterized by polyuria, extreme thirst and polydipsia. In the present case report, a 10 year old child presented with moderate pain in the molar area. Medical history revealed that the patient was under medication for diabetes insipidus. The patient also had a history of recurrent infection of the middle ear. Intra-oral examination revealed an early loss of primary molars with early eruption of upper first premolars. The patient had poor oral hygiene with marginal gingivitis and recession. While probing, there was severe pocketing related to the first permanent molars, especially the upper. Fluorosis of the permanent teeth was also evident.

Keywords - *Periodontitis; Diabetes inspidus; Langerhans cell histiocytosis; Fluorosis.*

INTRODUCTION

Pituitary diabetes insipidus is an endocrine disorder that results from a deficiency of the antidiuretic hormone (ADH), vasopressin.[1] This hormone is synthesized in the supra optic and the paraventricular nuclei of the posterior pituitary gland (neurohypophysis), and the granules containing the packaged neurohormones are transported via the axons to their storage site in the neurohypophysis.[2] The release of vasopressin is regulated by a complex system that, under physiologic conditions, controls the osmotic pressure of plasma.[3]

Pituitary diabetes insipidus may arise from pathological conditions of the neurohypophyseal unit. Common causes include head trauma, hemorrhage, histiocytosis and tumors of the basilar surfaces of the brain.[4] In rare cases, diabetes insipidus is inherited, usually as an autosomal dominant manner.[5-7] Oral features include dry mouth, lips and tongue. There is also evidence that mild to moderate fluorosis in optimally fluoridated communities is increasing.[8,9]

Diabetes insipidus can also be seen as a manifestation of a more complex disease known as Langerhans' cell histiocytosis, a disorder of the reticuloendothelial system, which is characterized by an abnormal proliferation of histiocytes and eosinophilic leukocytes.[10] Lichtenstein described the nomenclature -histiocytosis X - in 1953[11] to account for three clinical varieties, which showed some histological characteristics in common: eosinophilic granuloma, Letterer-Siwe syndrome and Hand-Schüller-Christian syndrome.

The term "histiocytosis" refers to a proliferation of histiocytes and other inflammatory cells, whereas the letter "X" was added to denote the unknown etiology of the disease.[10] The recent adoption of the terminology "Langerhans' cell histiocytosis" is due to the fact that the histiocytes involved in the disease present a phenotype which is similar to that of Langerhans' cells found in normal mucosa and skin.[12,13]

The Letterer-Siwe syndrome is considered to be the acute disseminated form of the disease, characterized by cutaneous lesions, hepatomegalies, splenomegalies and ganglionic hypertrophies, usually occurring in infants and newborns. Bone lesions occur in the skull, long bones and mandible. Lesions in the mandible show a definite radiolucent image that may mimic both juvenile and severe periodontal disease.

The prognosis is not favorable and most of the patients die within a short time.[10] The Hand-Schüller-Christian syndrome is considered to be the chronic disseminated form of histiocytosis X, characterized by a triad of symptoms which include exophthalmos, diabetes insipidus and osteolytic lesions in the skull. Systemic signs are frequent and include fever, skin rash, otitis, mastoiditis and upper respiratory infection. Lymphadenopathy, hepatomegaly and splenomegaly may also occur. Nearly half patients present a granulomatous involvement of the posterior pituitary gland or hypothalamus. Increased gingival volume and bleeding, deep pockets, alveolar bone loss and dental mobility, resembling periodontitis, characterize oral involvement. The earliest signs of this disease usually manifest during childhood. The prognosis for this clinical variety is better than that for the Letterer-Siwe syndrome, nevertheless, death occurs in some cases within a few years.[10,12]

Eosinophilic granuloma is the most frequently reported and mildest form of the disease.[10] This variety is considered to be a chronic, localized form, characterized

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by single or multiple osseous lesions, usually affecting children and young adults. Any bone in the skeletal system, including the mandible, may be affected. Roentgenographic characteristics are similar to those for Letterer-Siwe syndrome, resembling juvenile and severe periodontitis. The prognosis is excellent and the lesions may spontaneously recede within one or two years.[10,12]

Case Report

A 10 year-old male child patient was seen at the Alswani clinic, with the main complaint of moderate pain in the molar area. The past medical history revealed that the patient had a previous diagnosis of diabetes insipidus and was on antidiuretic hormone therapy. The patient also had a history of recurrent infection of the middle ear. While extra-oral examination was normal, intra-oral examination revealed early loss of primary molars with the early eruption of upper first premolars. A large amount of dental plaque, spontaneous gingival bleeding, gingival recession and increased gingival volume was evident (Figures1 and 2).

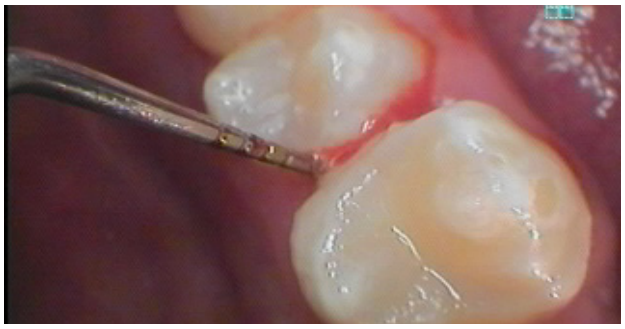

Figure 1: Intra-oral photograph showing early eruption of maxillary premolars with mild fluorosis and bleeding on probing.

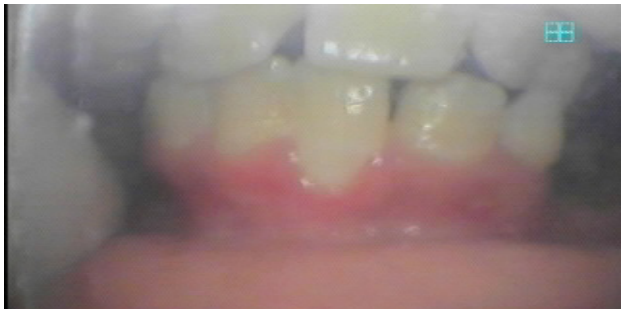

Figure 2: Intra-oral photograph showing a large amount of dental plaque on the incisors, spontaneous gingival bleeding, gingival recession and increased gingival volume. Note the moderate fluorotic stain on these teeth.

While probing deep periodontal pockets were observed in all sextants related to the first permanent molars, especially the upper, ranging from 5-10 mm in depth with mobility ranging from grade I to grade II (Figure 3). A mild to moderate dental fluorosis-like defect affected most of the permanent teeth; the fluoride level in the area where the patient lived was not known. In addition, no dental caries was detected. The parent of the patient was further questioned about the disease occurrence in his family. No familial occurrence was reported.

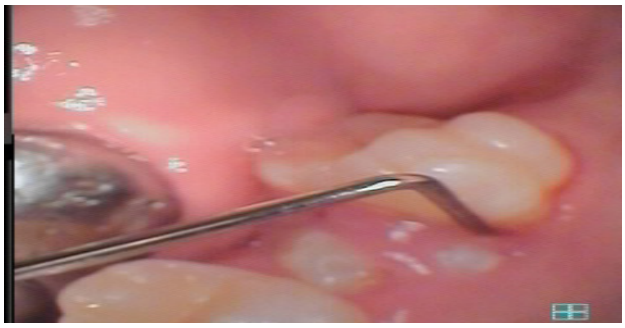

Figure 3: Intra-oral photograph showing deep periodontal pockets related to the first permanent molars.

Radiographic examination with O.P.G revealed that all permanent teeth were formed except for the right mandibular second premolar which was congenitally missing. The crown and root of the teeth appeared to be developing normally with the lower left second premolar ectopically positioned with distal inclination, while the lower first permanent were mesially inclined (Figure 4). The periapical radiograph in (Figure 5) shows how deep the periodontal pockets were on the upper right first permanent molar, where the periodontal probe was observed, almost completely, to enter the pocket.

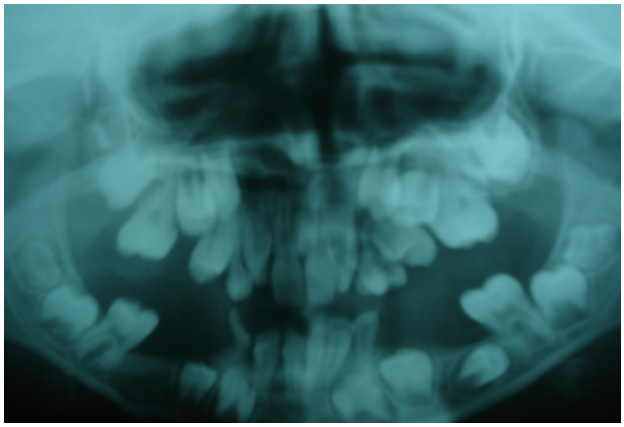

Figure 4: An orthopantamograph showing the missing lower right second premolar, ectopic position of lower left second premolar and mesial tilt of lower first permanent molars.

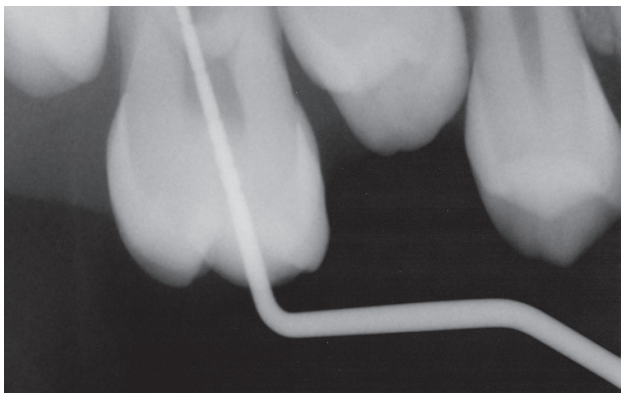

Figure 5: Periapical radiograph showing deep pocketing of first permanent molar.

DISCUSSION

Epidemiological studies indicate that gingivitis of varying severity is nearly universal in children and adolescents.[14-19] These studies also indicate that the prevalence of destructive forms of periodontal disease is lower in young individuals than in adults. Epidemiological surveys in young individuals have been performed in many parts of the world and among individuals with a widely varied background. For the most part, these surveys indicate that loss of periodontal attachment and supporting bone is relatively uncommon in the young, but that the incidence increases in adolescents aged 12 to 17 years old when compared to children aged 5 to 11 years old.[15-22]

The primary features of aggressive periodontitis include a history of rapid loss of attachment and bone loss with familial aggregation.

Secondary features include phagocyte abnormalities and a hyper responsive macrophage phenotype.[19] Aggressive periodontitis can be localized or generalized. Localized aggressive periodontitis patients (LAgP) have inter-proximal attachment loss on at least two permanent first molars and incisors, with attachment loss on no more than two teeth other than first molars and incisors. Generalized aggressive periodontitis patients (GAgP) exhibit generalized inter- proximal attachment loss including at least three teeth that are not first molars and incisors. In young individuals, the onset of these diseases is often circumpubertal. Some investigators have found that the localized form appears to be self-limiting, while others suggest that it is not.[19]

Periodontitis, as a manifestation of systemic disease in children, is a rare disease that often begins between the time of eruption of the primary teeth, up to the age of 4 or 5 years old.[22,23]

There is evidence that supports the view that there is a bidirectional relationship between diabetes and periodontal disease, that is to say, diabetes is associated with the increased occurrence and progression of periodontis and periodontal infection is associated with poor glycemic control in people with diabetes, so reinforcing the importance to monitor oral health.[8]

In this case report, the severe form of periodontitis affecting the first permanent molars in the presence of a medical history of central diabetes insipidus, has elicited the probability of the presence of other systemic diseases such as Langerhans' cell histiocytosis which also exhibits diabetes insipidus as one of it's features. Fortunately, the case was clear from Langerhans' cell histiocytosis according to the physician's report.

It is important to emphasize that certain signs and symptoms of life-threatening diseases may appear in the mouth long before they manifest themselves in other parts of the body. This report places oral health in the perspective of systemic health and suggests that the dental and medical professions need to develop closer ties in the future. The presence of fluorosis in the permanent teeth of patients with diabetes insipidus has been documented in several reports and has been attributed to the excessive consumption of optimally fluoridated water.[9] In the present case, although definitive diagnosis of fluorosis can not be made as the fluoride level in the drinking water was not known, this enamel defect could not be attributed to other causes as there was no metabolic or other medical history detected.

The patient's complaint of moderate pain during function was attributed to periodontal pockets in the molar area as there were no other dental problems such as dental caries. Treatment of the patient's periodontitis included local debridement and antibiotic therapy (Amoxill in combination with Metrionidazol).[24]

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