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Congenital insensitivity to pain with anhidrosis, dental considerations and management: A case report

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Background: Congenital insensitivity to pain with anhidrosis (CIPA), also known as hereditary sensory and autonomic neuropathy type IV (HSAN IV) is an autosomal recessive disorder characterized by insensitivity to pain, anhidrosis, intellectual disability and unconscious self-mutilation of fingers, lips, oral mucosa and tongue. Mutations in the neurotrophic tyrosine kinase receptor type 1 gene (NTRK1), have been reported to be associated with CIPA. No specific treatment for CIPA, however, early diagnosis and management of systemic and oral complications may be useful in the reduction of frequency and severity of these complications.

Aim: To report oral manifestations associated with CIPA and its management.

Case Report: 1.5 years old Saudi girl attended with her parents to the department of pediatric dentistry at Prince Sultan Military Medical City. The parents requested extractions of the erupted teeth. Oral examination revealed multiple mouth ulcers affected her buccal mucosa, tongue and lips as a result of continuous biting her oral soft tissues. Upper central incisors were exfoliated following eruption. Primary molars erupted prematurely. The child had recurrent episodes of hyperthermia and anhidrosis. Genetic analysis confirmed diagnosis of CIPA. The parents are consanguineous. All teeth were extracted on the dental chair in subsequent visits. All ulcers healed nicely at the follow up appointment at age 2 however, the patient had hip dislocation and unable to walk.

Conclusion: There are no guidelines available to treat this rare condition. However, early diagnosis and dental management of patients with CIPA are vital for prevention of orofacial manifestations. Mouth guard is useful to prevent oral injuries in older children.

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