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Lamellar Ichthyosis's Oral And Craniofacial Features In A Dental Setting

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Abstract

Background: *Hyperkeratosis and skin scaling are hallmarks of the uncommon autosomal recessive genodermatosis known as lamellar ichthyosis (LI). The oral and craniofacial manifestations are frequently disregarded, despite the prevalence of dermatological symptoms. Dental professionals might be the first to notice these symptoms, particularly in people who are undiagnosed or only slightly afflicted.*

Case Presentation: *We report a case of a young child who presented to the dental outpatient department with complaints of dry lips, bleeding gums, and limited mouth opening. Extraoral and intraoral examination revealed features consistent with lamellar ichthyosis, including angular cheilitis, ectropion, eclabium, gingival inflammation, and trismus. The patient had a prior diagnosis of LI confirmed by genetic testing (TGM1 mutation). A multidisciplinary care plan involving dermatology and dentistry was initiated.*

Outcome and Follow-Up : *Supportive dental care, such as emollients, scaling, and customized oral hygiene instructions, was well received by the patient. He still receives routine evaluations with coordinated dermatology assistance.*

Conclusion: *Dentists are essential in recognizing how systemic diseases like lamellar ichthyosis manifest orally. Improving quality of life and averting complications require early detection and interdisciplinary cooperation.*

Background

One of the main autosomal recessive congenital ichthyoses (ARCIs), lamellar ichthyosis usually manifests as a collodion membrane at birth and progresses to generalized hyperkeratotic scaling. One common cause is mutations in the TGM1 gene.[1] LI primarily affects the skin, but it can also cause oral problems like cheilitis, xerostomia, and limited oral opening because of skin tightness.[2]

Although these symptoms are rarely documented in dental literature, they may affect oral hygiene and function. The significance of identifying intraoral and craniofacial indicators of LI in dental practice is demonstrated by this case.

Case Presentation

A young child complained of persistently dry lips, bleeding gums, and trouble opening his mouth when he arrived at the dental outpatient department. The symptoms, which interfered with eating and oral hygiene, had gotten worse over time.

Medical history:

After a confirmatory skin biopsy and dermatological evaluation, the patient was diagnosed with lamellar ichthyosis during infancy. A pathogenic TGM1 mutation was discovered through genetic analysis. There were no other systemic conditions mentioned. He had never had dental treatment for his condition and was receiving sporadic dermatological follow-up.

Clinical findings:

Extraoral: Bilateral ectropion, everted dry lips with angular cheilitis, mild contractures, and generalized dry,





thickened skin with scaling.
Intraoral: mild trismus, xerostomia, a coated tongue, and widespread marginal gingivitis with a maximum interincisal opening of 28 mm.Hypoplastic incisiors with decayed 54.
Radiography: An orthopantomogram revealed no notable abnormalities in the osseous structure.

Investigations

No new investigations were performed at the time of dental consultation, as the diagnosis of lamellar ichthyosis had already been established via dermatological evaluation and genetic testing.

Differential Diagnosis

- Non-bullous congenital ichthyosiform erythroderma
- Harlequin ichthyosis
- Sjögren's syndrome (excluded based on clinical history and age)
- Nutritional deficiency cheilitis

Treatment

- Full-mouth ultrasonic scaling with caution due to fragile oral mucosa
- Lanolin-based emollient prescribed for lip hydration
- Personalized oral hygiene education using non-alcoholic mouth rinses and soft-bristled brushes

- Referral to dermatology for ongoing management and evaluation of systemic retinoid therapy
- Dietary recommendations and a focus on hydration to manage xerostomia

Outcome and Follow-Up

At the two-week follow-up, the patient's gingival health and oral comfort had improved. Dental checkups were scheduled every three months. A follow-up dermatological review was scheduled. Education regarding the condition's lifelong nature and the necessity of integrated care was given to the patient and his family.

DISCUSSION

Lamellar ichthyosis is mainly a dermatological condition, but it can also have serious oral and craniofacial symptoms. Ectropion and trismus can make facial function and oral hygiene even more difficult, while cheilitis and eclabium are caused by chronic dryness and skin tightening around the lips.[3]

A frequently disregarded sign of ichthyosis, xerostomia raises the risk of gingival disease and contributes to plaque buildup. Routine dental care may be difficult due to skin sensitivity and limited mouth opening. Preventive dental care, careful handling, and coordination with dermatology and other specialists are necessary for these patients.[4]

This case highlights the importance of dentists in detecting systemic conditions that show up in the oral cavity, especially when patients may not report subtle skin findings.

Learning Points

• Cheilitis, xerostomia, and trismus are oral manifestations of lamellar ichthyosis that can have a major impact on oral health and quality of life.

• It's possible that dentists will be the first medical professionals to spot symptoms of systemic diseases like ichthyosis.

• To manage the dental and dermatological issues associated with ichthyosis, multidisciplinary care is crucial. For patients with LI, long-term care should include regular dental monitoring and preventive measures.

Patient Perspective

Eating and brushing my teeth were always painful due to the dryness and tightness around my mouth. My lips feel much better and brushing is much easier now that I've been to the dental clinic and received the right care. I didn't know this skin condition could affect my mouth too.

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