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Case Report

Dental Care in Children with Dystrophic Epidermolysis Bullosa: Case Report and Evidence-Based Management

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Abstract

Epidermolysis bullosa (EB) is a group of rare, genetically determined disorders characterized by blister formation, skin fragility, and mucocutaneous blisters and erosions following minimal trauma. Oral manifestations and dental involvement of EB vary in frequency and severity depending on the subtype. Dental management of children with EB is complex due to the high risk of iatrogenic soft-tissue injury and requires strict adherence to evidence-based preventive and minimally invasive protocols. Published clinical practice guidelines recommend a trauma-free approach, reinforced preventive strategies, careful behavioral management, and modifications of dental instruments and procedures to minimize friction and pressure on oral tissues. However, detailed clinical implementation of these recommendations in complex pediatric cases remains limited in the literature. A 6-year-old girl with dystrophic epidermolysis bullosa and growth retardation was referred to the Pediatric Dentistry Department. Clinical examination revealed microstomia, multiple oral bullous lesions, gingival inflammation, ankyloglossia, enamel hypomineralization, and multiple carious lesions. Radiographic assessment was performed using panoramic imaging due to limited mouth opening. Dental management was carried out under local anesthesia only, using 2% lidocaine with epinephrine 1:80,000, without sedation or general anesthesia. Behavioral management

included the Tell-Show-Do technique and progressive chair acclimatization. Protective measures were applied during all procedures, including lubrication of lips with petroleum jelly and minimal-pressure suction and air usage. Treatment included multiple staged extractions of severely decayed primary molars, atraumatic restorative treatment of permanent and primary teeth using hand excavation and glass ionomer cement, and preventive care consisting of oral hygiene instruction and dietary counseling. A customized toothbrush adapted to the patient's hand deformity was also provided. The patient was followed over multiple visits with good tolerance and no major complications. This case reinforces that with strict adherence to minimally traumatic, evidence-based protocols, necessary dental treatment can be safely performed in children with dystrophic EB.

Keywords
Epidermolysis bullosa; dystrophic epidermolysis bullosa; oral manifestations; pediatric dentistry; dental management; case report

1. Introduction

The "Epidermolysis bullosa disease (EB)" refers to a group of vesicular bullous diseases dominantly or recessively inherited and characterized by a temperament that develops vesicles in ectodermal structures, naturally or after trauma [1]. It appears that EB is associated with abnormal enzyme activity and collagen degradation, but the exact pathogenesis and etiology remain obscure. In fact, Dystrophic Epidermolysis Bullosa (DEB) occurs when blisters appear in the upper dermis due to attachment problems between the basement membrane and the upper dermis. In a mild case, blisters tend to develop only on the hands, feet, knees, and elbows. Blistering can also be widespread, causing severe DEB. A baby born with severe DEB can have widespread blistering and areas of missing skin. Blisters may develop in the mouth and esophagus, making swallowing painful. When this happens, the child may need a feeding tube because the tube is missing or doesn't function, and the layers of the skin won't join properly. It can cause skin that relates to a flaw in the gene that helps produce a protein that glues the skin layers together if this protein looks thin. Mucous membrane disease can cause constipation and make eating difficult [2, 3].

Actually, EB as a group of conditions is rare [4]. The disease's phenotypes range from mild to life-threatening, with a prevalence of only about 500,000 people worldwide [5].

Patients suffer from psychological, general, and oral repercussions. Oral repercussions, frequent in children with EB, include hypoplastic teeth, gingivitis of the periodontium, bullous mucosal changes, hyposalivation, and xerostomia [2]. These latter require special precautions during their care [2]. Thus, a comprehensive oral care plan and conservative dental care are essential to eliminate pain and infection [6]. Dental treatment may improve aesthetics and self-esteem in patients with EB [7].

We aimed, through this case report, to illustrate the general and oral manifestations of hereditary DEB and to provide recommendations for appropriate precautions during oral care of these patients.

2. Case Report

A 6-year-old girl was referred to the Pediatric Dentistry and Prevention Department at the University Hospital XX for dental care. Since birth, she had skin detachments at the extremities. Most of her skin was affected by generalized body blisters. She had been diagnosed with Dystrophic Epidermolysis Bullosa with growth retardation. The Dermatology Department had confirmed the diagnosis before referral to our service. The family tree showed that it has a recessive mode of transmission linked to the X chromosome.

The general examination showed ocular and dermatological lesions associated with right- and left-hand syndactyly (Figure 1).

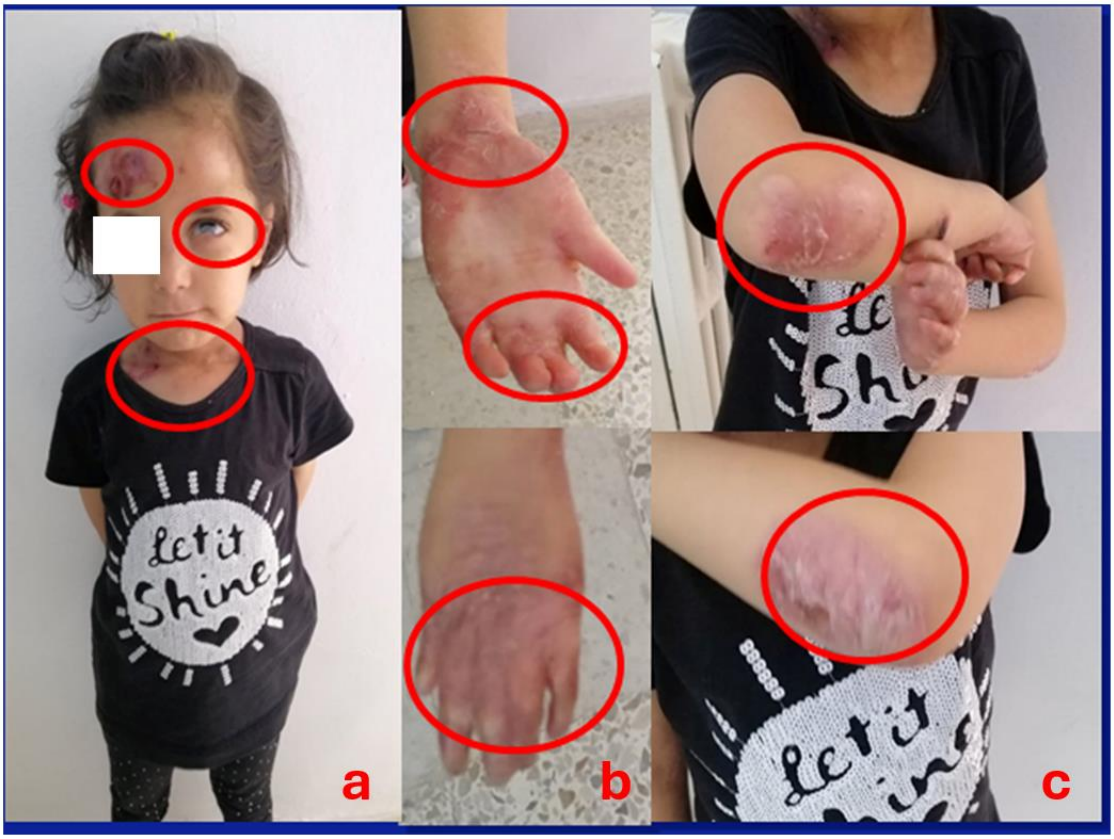

Figure 1 The front view of the patient showing: (a) Post bullous erosion on the forehead, neck, wrist, and bullous lesion on the left eye; (b) view of both left and right hands of the patient showing syndactyly of the 2nd and 3rd fingers; (c) examination of both right and left elbows shows post bullous erosion.

A written consent has been drawn up for the publication of these identity-revealing photos.

The oral examination revealed microstomia. Indeed, the size of the mouth isn't equidistant to the pupillary distance and is inferior to 1.5 times the alar width [8] (Figure 2a). There was also general gingivitis with irritation, redness, and swelling (Figure 2a) [9].

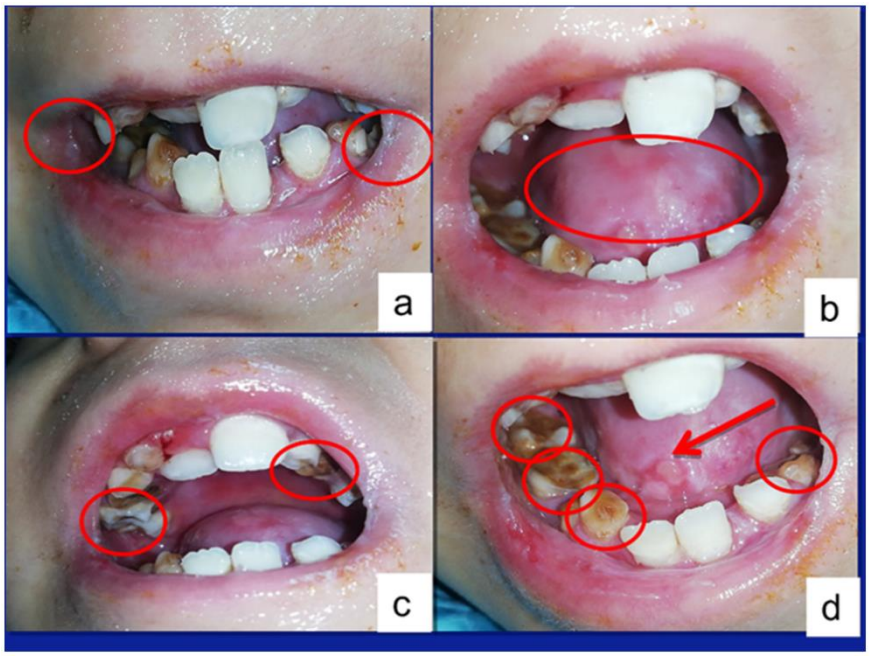

Figure 2 Dental status of the child showing: (a) microstomia, multiple bullous, general gingivitis, and anterior cross bite; (b) complete ankyloglossia, lingual pseudomembranous candidiasis; (c) multiple dental caries; (d) vestibular obliteration,enamel hypoplasia at the level of all first and second primary molars, and primary canine.

We also noted a vestibular obliteration: thus, the vestibular depth measured from the gingiva to the mucobuccal fold was less than 2.5 mm; and when measured from the crest of the lip to the mucobuccal fold, it was less than 10 mm [10] (Figures 2c, 2d), multiple bullous were found in the tongue and in the inner side of the lips and cheeks (Figure 3), aphthous-like lesions in the tongue(Figure 2d), complete ankyloglossia (Class IV according to Kotlow) [11] (Figure 2b), and multiple dental caries (Figures 2c, 2d).

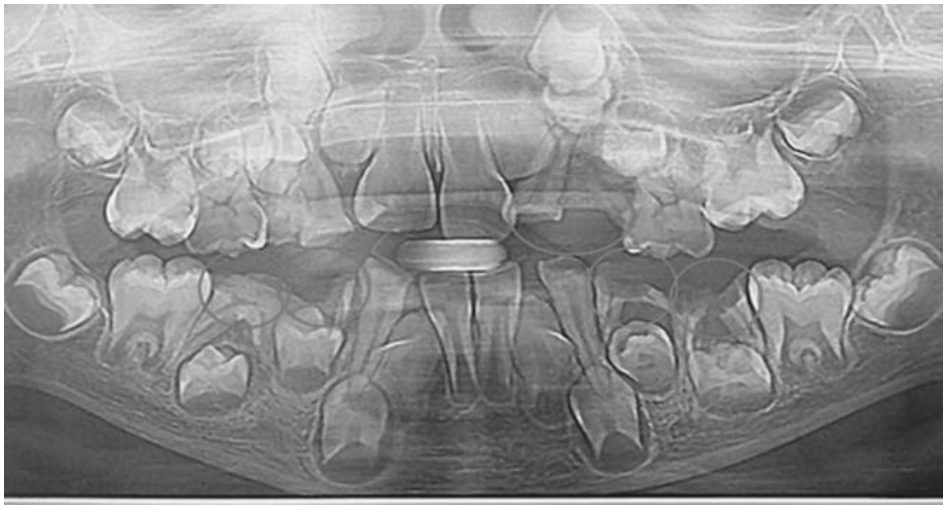

Figure 3 Panoramic view: dentomaxillary disharmony signs, pathological root resorption of the first and second temporary molars, and early loss of deciduous maxillary second incisors.

According to (DMFT/DMFTS) and (dmft/dmfts) classification, the clinical examination revealed:

#11: score 9

#21; #31; #41: score 0

#16; #26; #36; #46: score 6

#73; #53; #74; #75; #84; #85; #64; #65; #83; #63; #54; #55: score 1 (d)

#62; #52: score 4 (m)

We also noted lingual pseudoexhibitsous candidiasis with whitish-yellow plaques of soft, gelatinous consistency that exhibit a centrifugal growth pattern. These plaques detach upon rasping, leaving an erythematous zone. Symptoms were scarce (burning and itching sensation) [12] (Figure 2b), enamel hypomineralization with hypoplasia at the level of all first and second primary molars, and a primary canine with brownish or yellow coloration #73 (Figures 2c, 2d) with anterior crossbite (Figure 2a).

According to the recently proposed standardized scoring method for MIH/HSPM observations recording by Ghanim et al. [13].

#85; #75; #65; #55: score 3 (post eruptive enamel breakdown)

#16; #26; #36; #46: score 2 (white creamy demarcated opacities)

#11; #21; #31; #41: score 0 (no visible enamel defect)

For X-ray examination, the periapical technique wasn't suitable in this case because of microstomy and blisters in the sublingual region. A panoramic view was the only and the best choice. We noted signs of dentomaxillary disharmony, pathological root resorption of the first and second temporary molars, and early loss of deciduous maxillary second incisors (Figure 3).

The child benefited from chair initiation and dental equipment (Anthos A3 chair, Italy), motivation for oral hygiene, and dietary instruction. We replaced the brushing tools with a toothbrush adapted to the patient's hand, made of heavy silicone (Protesil, putty, VANNINI DENTAL, Italy) and molded to the patient's hand as the syndactyly worsened (Figure 4a).

Figure 4 Dental management of the child living with epidermolysis bullosa. (a) the personalized toothbrush, made with the heavy silicone modeled by the patient's hand, (b) the results of the curative and preventive treatments carried out.

On the first visit, we used the "Tell Show" Approach [14] to reduce fear. Lips smeared with petroleum jelly (Vaseline Blanche, Sfax, Tunisia) during the session to reduce blisters. The suction tip rests on the occlusal table of the molar to prevent lesion formation. Air syringes were used very carefully at the lowest possible pressure to avoid air bubble formation. In addition, the plaque removal was gentle.

At the second, third, and fourth visits, which were spaced 10 days apart, we extracted the left deciduous mandibular, first and second molars (#74; #75); the right deciduous mandibular, first and second molars (#84; #85), the left deciduous maxillary, and the first and second molars (#64; #65).

All treatments were performed in a dental chair with pre- and post-antibiotic prophylaxis and recall systems in place. The child was treated under local anesthesia, which can avoid the side effects of general anesthesia. Surface anesthesia by refrigeration (Ice spray, Pic solution, Italy) was used to relieve injection pain. Local Anesthesia based on 2% lidocaine with 1:80,000 epinephrine was administered deeply and slowly.

At the fifth visit which occurred 10 days later, all first permanent molars (#16; #26; #36; #46), the first right upper (#11), the first right lower incisor (#41), and the lower left deciduous canine (#73) and the right upper canine (#53) and the second upper deciduous molar (#55) were examined. The teeth were vital with no persistent pain response to the cold test. This led us to recognize reversible pulpitis. Thus, we proceeded with atraumatic restorative treatment. In fact, the carious tissue was removed manually using a low-speed round bur with a 14 mm diameter. The restorative material

was the glass ionomer (IMICRYL, R&D Series, Nova glass, F, Turkey). In the last session, we plan to extract the right lower deciduous canine (#83). We also performed the extraction of the left maxillary deciduous canine (#63) and the right deciduous maxillary first molar (#54). Restorative treatment was handled with care, adjusted, and polished to reduce the risk of iatrogenic cheek blisters and ulcers.

Thanks to the cooperation of the patient and her parents, the results of the curative and preventive treatments, as summarized in Table 1, were satisfactory (Figure 4b).

Table 1 Checklist of the procedures followed in the present report.

Procedure	Technique
Psychological approach	Introduction to the chair and dental equipment. Tell show do technique.
Motivation for oral hygiene, and dietary instruction	The making of a toothbrush adapted to the patient's hand.
Prevention of bubble and lesion formation	-Lips should be smeared with petroleum jelly. -The suction tip rests on the occlusal table of the molar. -Air syringes were used very carefully at the lowest possible pressure.
Periodental treatment	The plaque was gently removed.
Anesthesia	(1) Surface anesthesia by refrigeration. (2) Local anesthesia.
Surgical treatment	Extraction of (#74; #75; #84; #85; #64; #65). Under pre-and post-antiobioprophylaxy and recall system.
No traumatic restorative treatment	The removal carious tissue was made manually and with low speed round burs diameter 14. (#16; #26; #36; #46; #11; #41; #73). The restorative material was the glass ionomer.
In the last visit	

2.1 Ethics Approval and Consent to Participate

This case report was conducted in accordance with the ethical standards of CERB. Written informed consent to participate was obtained from the legal guardian.

3. Discussion

EB refers to a group of rare and complex diseases that require extensive knowledge of their complications and the precautions required to manage patients [2]. The frequency and severity of oral manifestations of EB vary by subtype, with the simplest form not affecting teeth while the dystrophic form (DEB) often results in enamel hypoplasia, early caries development, and gingivitis due to plaque buildup, leading to frequent tooth extractions [15, 16]. Studies have shown a significantly higher prevalence of dental caries, plaque, and gingivitis among individuals with DEB than among healthy people [16, 17].

Enamel defects in dystrophic epidermolysis bullosa (DEB) are closely related to the underlying genetic defect affecting epithelial integrity. As reported by Parushetti et al. [16], the fragility of the oral epithelium and the structural alterations of basement membrane components interfere with normal odontogenesis, particularly during the secretory and maturation stages of amelogenesis. This disruption may lead to enamel hypoplasia and hypomineralization, increasing tooth susceptibility to caries. In addition, recurrent oral blistering, chronic inflammation, and feeding difficulties, which are commonly observed in DEB patients, further aggravate nutritional deficiencies and may indirectly impair enamel mineralization, thereby contributing to the severity of dental involvement.

DEB also causes perioral and intraoral blisters, which can lead to microstomy, ankyloglossia, and occlusion of the oral vestibule. The normal vestibular depth, measured from the gingiva to the mucobuccal fold, varies from 2.5 to 11.5 mm, while the distance measured from the crest of the lip to the mucobuccal fold ranges from 10 to 29 mm [11].

In the present case, the child had DEB with blisters on the lips, tongue, and buccal mucosa, which were associated with massive and diffuse plaque accumulation. A diagnosis of gingivitis was established. The gingival condition improved progressively during follow-up visits following the implementation of reinforced preventive measures and improved oral hygiene practices.

There are many enabling factors of plaque buildup and gingivitis in DEB; such as painful mouth, blisters, cavities lesions, and tongue ankylosis; approved by the measuring of “free tongue” length between the frenulum attachment to the tongue and the tongue’s tip which is inferior than 3 mm [11], and microstomy which approved by the fact that the size of the mouth isn’t equidistant to the pupillary distance or inferior than 1.5 times the alar width [8].

Patients with EBH require specialized oral care due to the complexity of mouth opening and epithelial fragility [17, 18]. Moreover, the risk of morbidity and mortality among EB patients is high [19]. Indeed, there are many general complications of Bullous Epidermolysis such as a hypermetabolic state, High-calorie requirement for growth disorders, and uniform [2]. According to EB clinical practice guidelines, dental management should prioritize trauma prevention, minimally invasive procedures, and reinforced preventive care to reduce disease burden [6].

It’s recommended to prescribe pre-and postoperative antibiotic therapy to prevent infection of any blisters that arise during dental care [14]. Antibiotherapy is also recommended if the patient suffers from dilated cardiomyopathy to prevent infective endocarditis. Indeed, children with recessive dystrophic epidermolysis bullosa (RDEB) and Junctional EB non-Herlitz (JEB-nH) may develop dilated cardiomyopathy [2]. In our case, we prescribed pre-and postoperative antibiotherapy based on amoxicillin at a rate of 50 mg/kg, a single dose one hour before the act, then 50 mg/kg/day for 6 days. Although antibiotic prophylaxis was used in this case, current EB guidelines generally reserve antibiotic therapy for cases with a clear risk of infection or specific systemic indications, underscoring the importance of individualized risk assessment.

Restorative treatment is possible either under local analgesia or general anesthesia, but must be applied with extreme caution and with pre- and post-operative antibiotherapy [14].

General anesthesia is mostly contraindicated for patients with extensible junctional and dystrophic epidermolysis bullosa with affected lower airways. Scarring of these airways can lead to death from respiratory failure. In this case, all dental procedures were performed under local anesthesia only (2% lidocaine with epinephrine 1:80,000), using a slow and atraumatic injection

technique to minimize tissue distension and blister formation, and no sedation or general anesthesia was required [2, 20].

Children living with EB are chronically scarred on their skin. They may also present restricted mouth opening, esophageal strictures, and oral, pharyngeal, and laryngotracheal scarring [2]. Thus, they are prone to anemia, stunting, malnutrition, and infection. Patients require many precautions to protect the skin and oral mucosal surfaces from trauma using lubrication [21].

During dental procedures, a strictly atraumatic protocol was followed, including avoidance of excessive soft tissue stretching, continuous lubrication of the lips and oral mucosa with petroleum jelly to reduce frictional trauma, and the use of modified suction and low-pressure air delivery to prevent blister formation, a rubber dam could not be placed due to microstomy; effectively, clamps could injure the gum, our patient benefited from lubrication of the skin and surrounding mucous membranes and gingiva by the application of petroleum jelly (Vaseline) to prevent trauma because of the adhesion of the cotton rolls and mirrors to avoid traumatic labial or cheek traction [2].

Caries management followed a minimally invasive approach in accordance with EB guidelines, using atraumatic restorative treatment (ART) with hand excavation and selective low-speed instrumentation, followed by a glass ionomer cement [6]. A staged treatment plan was implemented, combining selective extractions of non-restorable primary molars with restorative care for teeth with reversible pulp involvement to control infection risk while minimizing trauma exposure per visit.

Treatment options for oral mucosal areas, other than Vaseline, are available, especially for the jugal area, such as applying aloe vera gel (Bright Sparkling, Forever Living Products, Scottsdale, Arizona, USA), which can diminish the subdermal temperature, provide a refreshed sensation, reduce the healing period, and promote antimicrobial activity [22]. In addition, Biotene mouthwash (GlaxoSmithkline, USA) can be prescribed, which reduces blister formation by moisturizing and stimulating saliva. This product has buffering capacity, immunological activity, antimicrobial activity, and self-cleaning effect [23].

Treatment of epidermolysis bullosa is generally supportive. Blister perforation accelerates healing and prevents lateral spread [23].

Currently, researchers are concentrating on gene and cell therapy, recombinant protein infusion, intradermal injections of allogenic fibroblasts, and stem cell transplantation. The development of new therapies focuses on enhancing wound healing and improving the quality of life for EB patients [24]. The use of petroleum jelly must be carried out carefully, taking into account the effects that may result from the ingestion of a large quantity of this product, such as abdominal pain, coughing, diarrhea, irritation of the throat, and shortness of breath [24].

The use of petroleum jelly must be carried out carefully, taking into account the effects that may result from the ingestion of a large quantity of this product, such as Abdominal pain, coughing, diarrhea, irritation of the throat, and shortness of breath [25].

The prosthetic dental rehabilitation of EB patients depends on the specific manifestations and the subtype of the disease [26]. It is essential to preserve the natural dentition; that’s why a removable space maintainer is usually contraindicated. In fact, scarring of normal oral tissues can render prosthesis retention impossible [2]. With the increasing longevity of patients with DEB, maintaining oral health and dental structure becomes increasingly important.

Oral rehabilitation can be fixed or removable as needed. Fixed rehabilitation is used whenever possible with devices. The use of stainless-steel crowns has been reported as a successful approach

for children with RDEB and JEB [27]. In the "Clinical practice guidelines: Oral health care for children and adults living with Epidermolysis bullosa" published in 2020, the authors report that a successfully removable prosthesis is tolerated by patients with EBS, JEB, DDEB, and pretibial RDEB [17]. Overdentures have been described as a practical, economical, non-surgical treatment option for patients with JEB [27].

Maintenance is essential for patients to BE through regular checkups to make sure that the patient presents good oral hygiene with regular use of topical/systemic fluoride and regular check-ups [2]. Early dental management, daily topical and systemic fluoride application, oral hygiene instructions, and preventive care measures help minimize caries development and improve oral health [15, 20].

Aphthous-like lesions can be related to multiple potential causes. Aphthous-like lesions can develop in patients with COVID-19 [28]. Also, many viruses, including cytomegalovirus, herpes simplex virus, and Zika virus, can infect the oral mucosa and cause very painful oral ulcers.

New compounds have recently been introduced and have been shown to have a significant impact on the oral environment. Probiotics, lysates, and postbiotics can alter clinical and microbiological parameters in patients with periodontal disease [29]. These products should also be considered as adjuvants in future clinical trials in patients with epidermolysis bullosa. However, it might not be possible to conduct such trials because many of these patients do not receive dental care early enough, as their parents pay for their child's health condition [2].

Despite the importance of our case, we faced certain limitations during visits to the dentist. We could not use the rubber dam to avoid mucosal damage, and we could not use the handpiece due to a microstomy. In addition, the periapical technique was unsuitable for radiological examination due to microstomia and sublingual blisters, and prosthetic treatment was not possible due to gingival blisters.

Epidermolysis bullosa is a rare disease that presents several oral and general manifestations, as well as significant psychological repercussions, making any therapeutic action difficult. Through this case report, we have highlighted the specific oral care needs of children with hereditary epidermolysis bullosa, ranging from disinfection of the oral cavity to application of petroleum jelly during any skin contact or mucous membrane, through the avoidance of traumatic manipulations and the prescription of pre- and postoperative antibiotic therapy, even for anesthesia.

Author Contributions

S.K. and MA.C. contributed to the conception and design of the study and data collection. R.J. contributed to the clinical management and writing of the manuscript. M.C. contributed to the manuscript revision. All authors read and approved the final manuscript.

Competing Interests

The authors declare that they have no competing interests.

Data Availability Statement

All relevant data generated during this study are included in this published article.

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