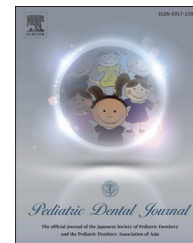


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Case Report

Conservative dental management of a patient with Epidermolysis bullosa. A case report

Khashayar Sanjari ^a, Mojtaba Bayani ^b, Hosna Ebrahimi Zadeh ^{c,*}

^a Department of Pediatric Dentistry, Dental School, Gilan University of Medical Sciences, Gilan, Iran

^b Department of Periodontics, Dental School, Arak University of Medical Sciences, Arak, Iran

^c Dental Research Center, Restorative Department, School of Dentistry, Shahid Beheshti University of Medical Sciences, Tehran, Iran

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ABSTRACT

Epidermolysis bullosa (EB) is a vesiculobullous disorder characterized by skin and mucous membrane fragility. Intubation in general anesthesia could incite formation of bullae. It is crucial to make a comprehensive dental care plan and conservative dental treatment to eliminate pain. We present a patient with EB that treated under local anesthesia. It can prevent adverse effects of general anesthesia. We applied partial pulpotomy with Biodentine to manage deep carious lesions. To manage gag reflex and anxiety, mild oral sedation was used. At 1-year follow-up, the patient was at a good oral health condition.

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1. Introduction

Epidermolysis bullosa (EB) is a genetic vesiculobullous disorder characterized by skin and mucous fragility [1]. Patients illustrate blister after trauma or even spontaneously [2]. It could be a life-threatening disease [3]. There are 4 major types of EB: simplex, junctional, dystrophic, and Kindler syndrome [4,5]. Trauma can lead to detachment of the skin and pseudosyndactyly [6]. Treatment in EB patients is based on corticosteroids [7]. Malnutrition is common due to deplete in oral

intake because of oropharyngeal lacerations. The other signs are limited mouth opening and esophageal stricture [8]. Obstruction due to blister after extubation may provoke airway complications [9]. Intubation during general anesthesia could instigate formation of bullae and airway obstruction [10]. It is imperative to make a comprehensive oral care plan and conservative dental treatment to eliminate pain and infection [11]. Dental treatment could promote esthetics and self-confidence of EB patients [12]. Functional teeth can diminish the capacity for oral injury by making mastication more effective [13,14].

* Corresponding author. Department of Restorative & Cosmetic, School of Dentistry, Shahid Beheshti University of Medical Sciences, Tehran, Iran.

E-mail addresses: khashayarsanjari@gmail.com (K. Sanjari), mbayani@mail.com (M. Bayani), ebrahimi_hosna@yahoo.com (H.E. Zadeh).

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Here, we present an EB case that treated under local anesthesia. Managing an EB patient in outpatient condition is difficult for dentist because of compromised access. But it can prevent adverse effects of general anesthesia.

2. Case report

An 11 year-old male was referred to the Department of Pediatric dentistry at Arak University of medical science, dentistry faculty Arak, Iran because of frequent dental pain (Figs. 1–4). When the boy was born, had been diagnosed with EB. He applied silver sulfadiazine ointment on the skin lesions. He had no known allergies and his diet was limited to soft food.

At referral, the patient appeared to have normal cognitive abilities His vital signs were within normal limits. Most of his skin was affected by generalized body blisters. The patient had a smooth tongue with obliteration of the buccal and lingual vestibules. His dental status was poor, with multiple dental caries. Due to the patient's poor dental status, a full dental treatment under general anesthesia was recommended at first. But intubation might compromise airway, so we decided to do dental treatments under local anesthesia. The appropriate written consent for the procedure was obtained after comprehensive consultation with the patient, his parents and a dermatologist.

For radiographic examination, Periapical technique wasn't suitable in this case because of microstomia and blisters in sublingual region. Panoramic view was the only and the best choice (Fig 5).

At first visit “Tell Show Do” approach was utilized to reduce anxiety. Lips were lubricated with Vaseline during the session to decrease bullae formation.



Fig. 1 – Frontal photograph of the EB patient.



Fig. 2 – Photography of the back and arms of the patient.



Fig. 3 – Photography of the feet of the EB patient.



Fig. 4 – Oral view of the EB patient.

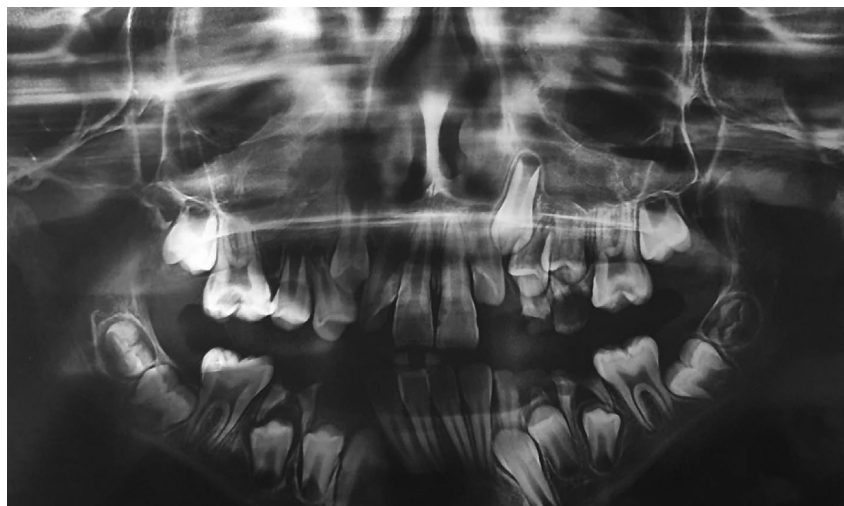


Fig. 5 – Panoramic view of the EB patient (before treatment).

Suction tip lean on occlusal table of molars to prevent lesion formation. Air syringe was used very carefully with lowest pressure to avoid bullae creation. Plaque removal was done gently. In oral examination, left permanent mandibular molar revealed stained fissures. To prevent future caries formation, Fissure sealant was programmed to the tooth. Due to difficult access to molar teeth, it was not possible to use resin sealant materials. Lubricated small cotton rolls were used to diminish mucosal trauma during restorative procedure. It was better to apply glass ionomer sealant (Fuji VII GC Tokyo, Japan) to elevate the fluoride levels in saliva and simplify the procedure.

Due to high gag reflex of patient and lack of cooperation, Hydroxyzine suspension (Kharazmi Pharma Co., Tehran, Iran) was prescribed to reduce the reflex (1.0 mg/kg). The suspension was taken 30 minutes before each session. This method also had a sedative effect on patient.

In second visit, first right maxillary molar was tested. The tooth was vital with prolonged pain reaction to cold test. This condition led us to detect a reversible pulpitis condition. In this session, Topical Procaine (Pascal International™) was used to reduce the pain of injection. Local anesthesia 2% lidocaine with 1:80,000 epinephrine (Daroupakhsh, Tehran, Iran) was deeply and slowly administered. Caries removal of first right maxillary molar was conservatively done by a hand piece with thin and small head (S-Max Pico. NSK Japan) and ultra-short shank burs (Pico burs NSK Japan) to facilitate access. But a single pinpoint exposure was observed after removing affected dentine. There was no access to use rubber dam, so isolation was performed by sterile cotton rolls. Increased bleeding on exposure site, suggests that the inflammatory response extends deeper into the pulp tissue and the treatment procedure should be modified to partial pulpotomy. The surgical removal of 2 mm of the coronal pulp was done. Bleeding was controlled by application of 2.5% hypochlorite sodium. Each time a small sterile cotton pellet soaked in 2.5% NaOCl and applied on exposed part of pulp for 3 minutes. When hemostasis achieved, Biodentine (Septodont,

Saint-Maur-des-Fosses, France) was processed according to the manufacturer's instruction by mixing a single-unit powder part and five drops of a single-unit liquid part for 30s. Biodentine was placed in the coronal pulp space with a mineral trioxide aggregate (MTA) carrier (Dentsply Maillefer, USA) and was condensed gently using hand pluggers (Dentsply Maillefer, USA). The thickness of Biodentine layer was 2 mm. The restorative material of choice was amalgam (Tytin™, Kerr Corp., CA, USA).

First right permanent molar of mandible was affected by caries. Caries removal was done conservatively. The last layer of firm caries was preserved to prevent pulp exposure. A thin layer of calcium hydroxide (Dycal, DENTSPLY) was implemented to the deepest area. Then RMGIC liner (Fuji II LC, GC Corporation, Tokyo, Japan) placed on the Dycal layer. The restorative material of choice was amalgam.

At the end of the dental treatment, all mucosal surfaces were evaluated to find blisters and drain them. The time distance between sessions were planned every 2 weeks to let the mucosa get healed.

In the third session, left maxillary molar was planned to treat. The procedure for first left maxillary molar was exactly similar to the right one.

In fourth session, dental extraction of lower left primary canine, first and second molars were done as atraumatically as possible in order to minimize laceration to the oral soft tissues. The sockets were packed with Gelfoam and suturing was not used to avoid iatrogenic soft tissue trauma. At a 1 month follow-up session, the soft tissues and extraction sockets had cured successfully with no scarring.

Sucralfate suspension was prescribed to decrease the of bullae. Because of high risk of caries in this patient, dental review scheduled every 3 months to emphasize on dietary consultation and application of topical fluoride.

At 1-year follow-up visit, the patient had no complaints of pain. The clinical and radiographic examination showed a good periodontal health (Fig. 6). Calcific barrier was observed on the radiograph below the Biodentine layer. The thickness of secondary dentin layer was about 2 mm.



Fig. 6 – Panoramic view of the EB patient (after treatment).

3. Discussion

Epidermolysis bullosa is a congenital skin and mucosal disorder with fragility in skin and mucosa [15]. EB is a rare disease, but in a dental office, it needs particular care due to the complexity in mouth opening and epithelial fragility [16,17]. The risk of morbidity and mortality among EB patients is high [18].

General anesthesia may generate complications because the intubation procedure could compromise the patient's airway [19]. Most authors declare that routine dental treatment can be provided [12,20]. A patient has been reported whom wearing dentures for many years [21].

Conservative methods are preferred to minimize oral laceration [22]. Examples of successful dental procedures which were done under local anesthesia include root canal treatment, and restorations [23]. In this case, dental procedures such as extractions, restoration and pulp therapy were done successfully with minimally invasive methods.

Hydroxyzine is one of the antihistaminic drugs used for its sedative and antiemetic capacity [24]. Its sedative effect appears quite late but lasts long enough for lengthy dental work [25]. Many articles showed high overall sedation success rates [26,27]. Some studies demonstrated some post-sedation effects such as prolonged sleep and irritability [28]. In this case, Hydroxyzine was applied successfully to control anxiety and gag reflex, during dental procedure.

Vital pulp therapy (VPT) is defined as a treatment which aims to preserve pulp vitality especially in young patients. VPT can induce root formation in these teeth [29]. Another benefit for preservation of vital pulp is the protective resistance to mastication forces compared with a root-canal-filled tooth [30]. Several studies have reported successful treatment outcome in vital teeth with curiously exposed pulp with signs and symptoms of irreversible pulpitis and periapical lesion [31].

If the bleeding on exposure site is pinpoint, normal and controllable direct pulp capping (DPC) is suggested [32]. Increased bleeding on exposure site that is difficult to stop, suggest that the inflammatory response extends deeper into

the pulp tissue. In Partial or Cvek pulpotomy The inflamed tissue is removed to the level of healthy coronal pulp tissue [33].

Partial pulpotomy has some advantages compared to direct pulp capping such as: removal of the superficially inflamed pulp tissue and providing space for the dressing material which gives the opportunity to seal the cavity [34].

In permanent dentition, mineral trioxide aggregate (MTA) is suggested in most of the pulp capping cases [35]. The main disadvantages of MTA are tooth discoloration and long setting time [36]. Also MTA is difficult to handle [37].

The other biomaterial which is used as a pulp capping agent is Biodentine. Biodentine releases more Calcium ions than MTA [38]. Biodentine has several ingredients such as tricalcium silicate, dicalcium silicate, calcium carbonate and zirconium oxide. Liquid of Biodentine includes calcium chloride as an accelerator agent [39]. Biodentine has multiple advantages. For example: low level of cytotoxicity [40] excellent biocompatibility and inducing formation of dentinal tags [41].

In this case, pulp management of deeply carious teeth were done successfully by partial pulpotomy method with Biodentine. This case report is the first case that biodentine was applied in an EB case.

Amalgam is a clinically acceptable restorative material [42]. Success of the amalgam restorations was influenced by the patient's caries experience (DMFT) and restoration size [43].

The restorative material of choice in this case report was amalgam. This material could provide an excellent coronal seal. Despite of composite resin materials, amalgam doesn't need high level of isolation and it can make the restoration procedure easy.

Louloudiadis AK [44] claimed that oral health of these patients can be improved and eliminate complications arising from the oral condition. This result is in agreement with our study.

Esfahanizade [45] and co-authors managed dental problems of an EB patient with general anesthesia, but in this case report, we managed all dental treatments, outpatiently.

The dental implant failure rate in EB patients seems to be very low [46], so it was suggested to the patient that he could receive implant in future if is indicated.

4. Conclusion

Although managing an EB patient under local anesthesia is complex for dentist and his team because of minimum mouth opening and microstomia, it can be ideal and safe and is beneficial for the patient with preventing repeated general anesthesia and its adverse effects. The main benefits of local anesthesia are that it is minimally invasive for airway and supply lengthy post-operative pain control.

Declaration of Competing Interest

There are no conflicts of interest.

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