

CASE REPORT

Oral self-inflicted accidental trauma in patients with neurological disorders: a case report of dental management in infants with cerebellar hypoplasia

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Abstract

Self-inflicted oral injuries, accidental or otherwise, can cause major consequences. Measures need to be taken to protect individuals from chronic self-injurious behaviour; however, there are no official guidelines on the subject. The purpose of this article is to show the case of a 1-year-old patient with neurological disorders who, following the eruption of deciduous teeth, had self-inflicted a traumatic ulcer on his tongue and lower lip. Following a multidisciplinary approach involving several operating units of our hospital to make a diagnosis, an oral device was designed to completely cover the dental elements to prevent recurrence of the trauma and to prevent further worsening of the injuries already caused. The purpose of this work is to demonstrate that although the surgical approach, such as extraction of the dental elements, may be the quickest solution in situations similar to the one presented, the high biological cost and irreversibility of the result lead to seeking alternatives and more conservative solutions such as the one described.

Keywords

Self-inflicted trauma; Oral cavity; Neurological

1. Introduction

Single or occasional episodes of self-induced accidental oral injury in the oral cavity are relatively benign; recurrent self-injury can result in a major sequelae and predispose the patient to develop chronic and serious complications [1]. In the oral cavity, lesions induced by self-inflicted trauma were first described clinically by a dermatologist, Dr. Schuermann, in 1958, in a textbook on diseases of the oral mucosa and lips under the term “Morsicatio Buccarum” (MB), pointing out that certain patients, even healthy ones, had compulsive neuroses and were responsible for this type of self-provoked injury [2, 3]. The cause of self-induced masticatory trauma on oral mucosa behaviour has been widely reported, especially in patients with cerebral palsy, severe neurological disabilities, Tourette syndrome, Lesch-Nyhan syndrome, Chiari malformations type II, congenital insensitivity to pain, other neuropsychiatric disorders, and cases of childhood encephalitis [4, 5]. The masticatory cycle is coordinated by a complex of acting muscles following brain inputs, and the lack of control of the chewing motion which can sometimes result in neuropathological mastication and cause the presence of accidental intraoral lesions. The management of these cases depends on the anamnesis frequency and severity of these lesions [6]. To prevent the difficulties associated with ongoing

oral trauma, measures should be taken to protect individuals from chronic self-injurious behaviours. The diagnosis and management of chronic self-induced masticatory injuries is various and nonspecific; in fact, there are no official guidelines on this issue [7, 8]. In this paper, we present an interesting clinical case of a 1-year-old girl affected by psychomotor retardation due to cerebellar hypoplasia at the diagnostic stage who experienced self-induced trauma to the oral mucosa (mainly lower lip and tongue) and describe her management.

2. Case report

A 1-year-old female infant with psychomotor retardation, hypotonia, cerebellar hypoplasia and convergent strabismus came to our attention by “The Emergency Department” of the Bambino Gesù Children's Hospital based in Rome in May 2022 for the presence of whitish-coloured lesions with ulcerated appearance and thickened consistency, particularly represented at the level of the lower lip. Her parents reported the onset of these lesions for approximately two months in association with the eruption of the deciduous incisors. Therefore, a diagnostic course involving multiple professionals was carried out to achieve an appropriate diagnosis. First, the diagnostic path involved dermatologists and dentists; later, maxillofacial surgeons, neurologists,

infectiologists, rheumatologists, and specialists in metabolic diseases and paediatricians were involved. At her first follow-up, the extraoral clinical examination (Fig. 1a) evidenced continuous solution of the lower lip tissues with loss of substance at the lower incisors, in absence of facial asymmetry but with perilabial skin maceration. At the intraoral examination (Fig. 1b,c), the deciduous incisors were fully erupted, 54, 64, 74 and 84 were midway through their eruptive course and white, nonexcisable lesions at the apex and lingual and in the cheek mucosae were evidenced.

A lip wound swab test was performed, and the results were positive for *Pseudomonas mexocina* and *Stenotrophomonas maltophilia*. Due to the inability to feed adequately, a nose-gastric tube was placed, and intravenous antibiotic therapy with levofloxacin based on the antibiogram of the lip swab and lorazepam 0.25 mg once/day were administered.

A cold-blade incisional biopsy of the lower lip lesion was performed under deep sedation. Histological examination showed necrosis and ulceration of the labial mucosa with a brisk inflammatory infiltrate composed of lymphocytes and neutrophils. The infiltrate extended to the deep tissues and involved the striated muscle. Arterioles showed vasculitic-like features and thrombosis secondary to the presence of inflammation. Moreover, the adjacent mucosa showed pseudoepitheliomatous hyperplasia with vacuolization of epithelial cells and the presence of apoptotic bodies. Bacterial colonies were also evident (Fig. 2).

Therefore, the diagnosis of self-inflicted masticatory trauma was confirmed. Blood chemistry tests, radiographic investigations, complete ultrasound of the abdomen, an MRI (magnetic resonance imaging) of the brain and specialized consultations were performed during the hospitalization period to investigate the presence of any pathology and/or syndrome. In particular, Behçet syndrome and hypoxanthine-guanine-phosphoribosyltransferase deficiency (Lesch-Nyhan syndrome) were ruled out, immunological tests for autoantibodies were negative, and karyotype examination was in progress to evaluate the diagnostic and prognostic aspects of this disease. Brain MRI showed elongated cerebellar peduncles with a “molar tooth” appearance of the midbrain (Fig. 3) and dysplastic cerebellar vermis and hemispheres: findings described in Joubert’s syndrome and related disorders.

However, genetic verification confirming the above diagnostic suspicion remains for our patient. She has no brothers or sisters, but she is the oldest born after two pregnancies that resulted in miscarriage. She performed myofunctional therapies with the speech therapist and physiotherapist daily to stimulate the functions of the neuromuscular apparatus. The evaluation of maxillofacial surgeons and dentists aimed at lower lip reconstruction was temporarily ruled out in favor of a more conservative therapy. A silicone impression (polyvinylsiloxane) of the upper dental arch was taken under narcosis during the incisional biopsy of the lower lip to fabricate a resin oral device that would anchor on the upper deciduous dental elements.

The limits of this case are represented by the fact that the material with which the device is made deteriorates over time and therefore, during the follow-up, it may be necessary to acquire dental impressions again. Taking dental impressions

in patients who have neurological disorders of this type can be difficult due to the increase in spasticity within the procedure causes.

The oral device was designed to completely cover the maxillary teeth and adapted to the occlusion with an acrylic resin of 5 mm in thickness. The device had vestibular flanges to distance the fornix mucosa from the occlusion and a central expansion screw to ensure retention. To increase the fitting of the plate, the use of fixative cream (Corega Super-Haftcreme; Procter & Gamble, Cincinnati, OH) was recommended, and nonresorbable 3.0 silk suture (without needle) was inserted through the hole in the canine region as a safety thread to avoid any risk of suffocation (Fig. 4). The appliance was prescribed to be worn full-time except during meals. Hyaluronic acid and vitamin E topic gels were applied to promote soft tissue healing two times a day for 14 days without the need for rinsing. Four days after dental device placement, the mother reported that the device was well tolerated by the child and that the bleeding episodes of the lesions decreased. At the intraoral examination, a size reduction of the lesions was shown, such as the change in ulcerated areas in favor of the presence of fibrin (Fig. 5). Then, the patient was evaluated every week for the following 6 weeks to monitor the congruence of the occlusion, the fitting and any changes in the dentition as well as the evolution of wound healing of the lesions (Fig. 6). In the following monthly visits, a progressive change in the consistency of the lesions, which began to be taut-elastic, was observed, and the areas of fibrin were replaced by scar tissue. At the level of the labial mucosa, the lesions dimensionally regressed, and the lesions of the cheek mucosa disappeared.

However, the lip shows loss of substance on the median plane with scarring outcomes and poor competence. Lesions at the level of the lingual belly have shown slower healing. Parents report decreased episodes of clenching.

During every check, the appliance was relined as needed by removing precontacts and excess resin to promote better positioning; the buccal resin flange was modified according to the continuation of the eruptive path of the upper deciduous elements. The central expansion screw was progressively activated to facilitate plate fitting and improve mucosal distancing from the occlusal plane (Fig. 7).

Thereafter, periodic checkups were performed monthly. Drug therapy with lorazepam was progressively discontinued, while the intraoral device was worn, followed by remodelling of the same and activation of the central expansion screw until December 2022. Subsequently, a new palatine plate was made in January 2023, reshaped during each control to promote eruption of the dental elements. It was recommended that the caregiver wear the palatine plate for 24 hours a day at times when the child was more restless and agitated, while at times when she was more relaxed, it was recommended that it be worn only at night. At the same time, she continued physiotherapy and speech therapy to stretch the labial scar.

3. Discussion

Accidental self-injury is most classically associated with epileptic crisis but is also found in patients with Lesch-Nyhan syndrome, cerebral palsy, congenital insensitivity to pain



FIGURE 1. Clinical observation. (1a) extraoral clinical photography of the face. (1b) intraoral clinical photography of the lower lip. (1c) intraoral clinical photography with evidence on the cheek mucosa, lower lip and lingual apex.

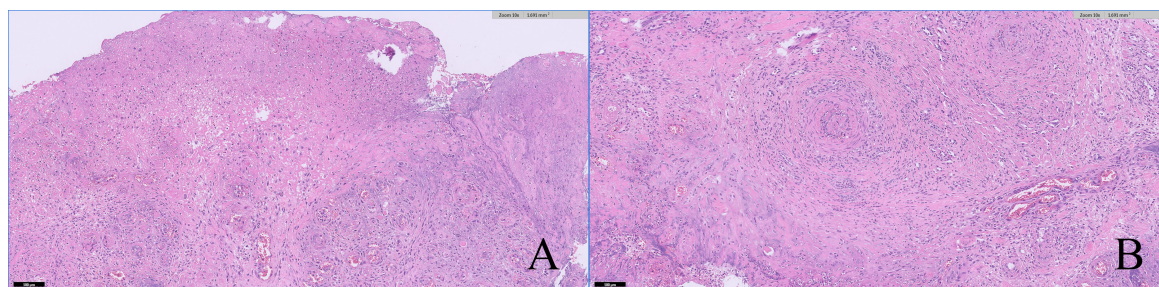


FIGURE 2. HE staining. (A) At lower magnification, the biopsy showed ulceration of the labial mucosa and diffuse necrosis extending through the deeper layers. (B) At higher magnification, a brisk chronic and acute inflammatory infiltrate was present, and involvement of arterial vessels with transmural vasculitis and thrombosis was noted.

with anhidrosis, mental disorders with mental retardation, psychiatric diseases, meningitis, and brain trauma. In addition, non-suicidal self-injury is sometimes a premeditated action aimed at seeking attention and help [9]. Joubert syndrome (SJ) is an autosomal recessive syndrome characterized by a congenital malformation of the brain stem and agenesis or hypoplasia of the cerebellar vermis that results in defects in respiration, nystagmus, hypotonia, ataxia, and delay in motor developmental stages. The pathognomonic feature of the disease is the “molar tooth sign” in brain MRI images. Searching in OMIM (On-line Mendelian Inheritance in

Man) revealed that some of the involved proteins are CEP41 (centrosomal protein 41), CC2D2A (Coiled-Coil and C2 Domain Containing 2A) and KIF7 (Kinesin Family Member 7). Patients with self-provoked intraoral injuries due to neurological deficits require very scrupulous attention from caregivers because of their lack of cooperation. Chronic tongue biting causes injury to soft, vascular and lymphatic tissues, leading to oedema while predisposing the tongue to further injury [9]. Most importantly, severe trauma may result in haematoma with subsequent traumatic macroglossia and airway compromise [9]. Self-inflicted injuries can often

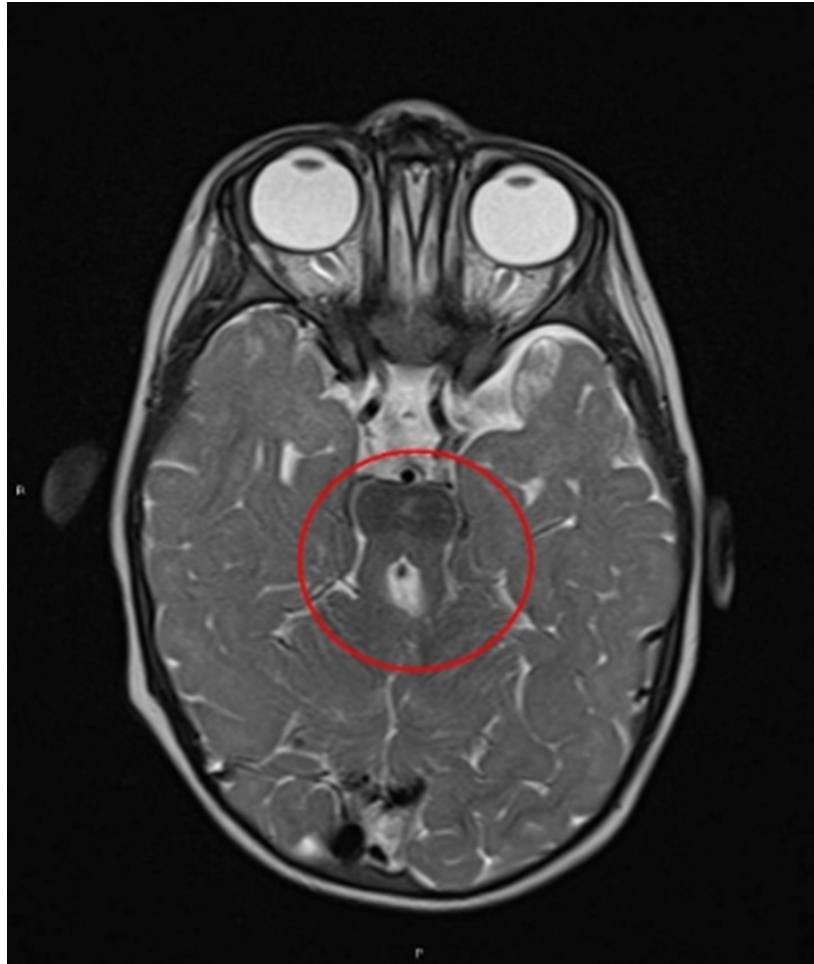


FIGURE 3. Axial T2-weighted brain MRI showing the “molar tooth” appearance of the midbrain.

generate superinfection, so to prevent it, wounds should remain cleansed, and necrotic material should be removed. The use of antibiotic therapy is recommended, as in our case, only where there is evidence of bacterial contamination, delayed healing or immunodepression. Treatment strategies involve different solutions depending on the patient's age, underlying pathology, and the extent of trauma. Alternative solutions consist of selective grinding of the erupted tooth elements, topical application of medical products for oral use (for example, in Riga Fede's disease), injection of botulinum toxin to result in paralysis of the masticatory muscles, fabrication of intraoral appliances that provide a screen to the continued recurrence of trauma, or in extremis, extraction of the tooth elements [9–11]. In the case we described, however, our patient, through an uncontrolled masticatory cycle and unthoughtful masticatory force, had generated injuries so severe that lipoplasty would be necessary in the future to make up for the loss of any substance generated. Indeed, a neurological consultation showed irritability and restlessness in the patient as well as a facial expression of pain. For this reason, we considered it appropriate to provide an oral screen to fit the entire oral vestibule built by a dental technician and made of acrylic resin with dental floss or suture [12] attached to it for security to avoid trauma, following the treatment strategy of Yasui *et al.* [13].

When wounds are deep, they should be closed by plane

suturing, which involves first suturing the muscle layers by internal resorbable stitches and then suturing the more superficial layers [14]. Having resolved the trauma, the primary goal is to prevent the recurrence of injury and the generation of complications [15]. The literature describes a variety of treatment modalities, including psychological therapy consisting of positive and negative behaviour reinforcement, overcorrection, and distraction techniques in the initial phase [12]. Associated with behavioural therapy, we find various types of treatment described in the literature that differ in method. These types of treatments consist of what we reported in Table 1.

These oral appliances serve to suppress forced habits and act as physical barriers against injury, providing better protection and control according to some clinicians. The palatine plate, in particular, has the function of generating an interposed shield between the occlusal plane and the oral mucous membranes through the use of vestibular flanges and the covering of the dental elements. The application of this device has shown various benefits, including the reactivation of sucking and swallowing, leading to better acceptance of the plaque by the patient. The plaque allows the tongue to be directed to the spot, improving deglutition dynamics and muscle competence. Being more engaged by the stimulation of the appliance, the patient will presumably be distracted by the tendency to bite [23, 24].

Proper use of these devices can allow healing of lacerations.

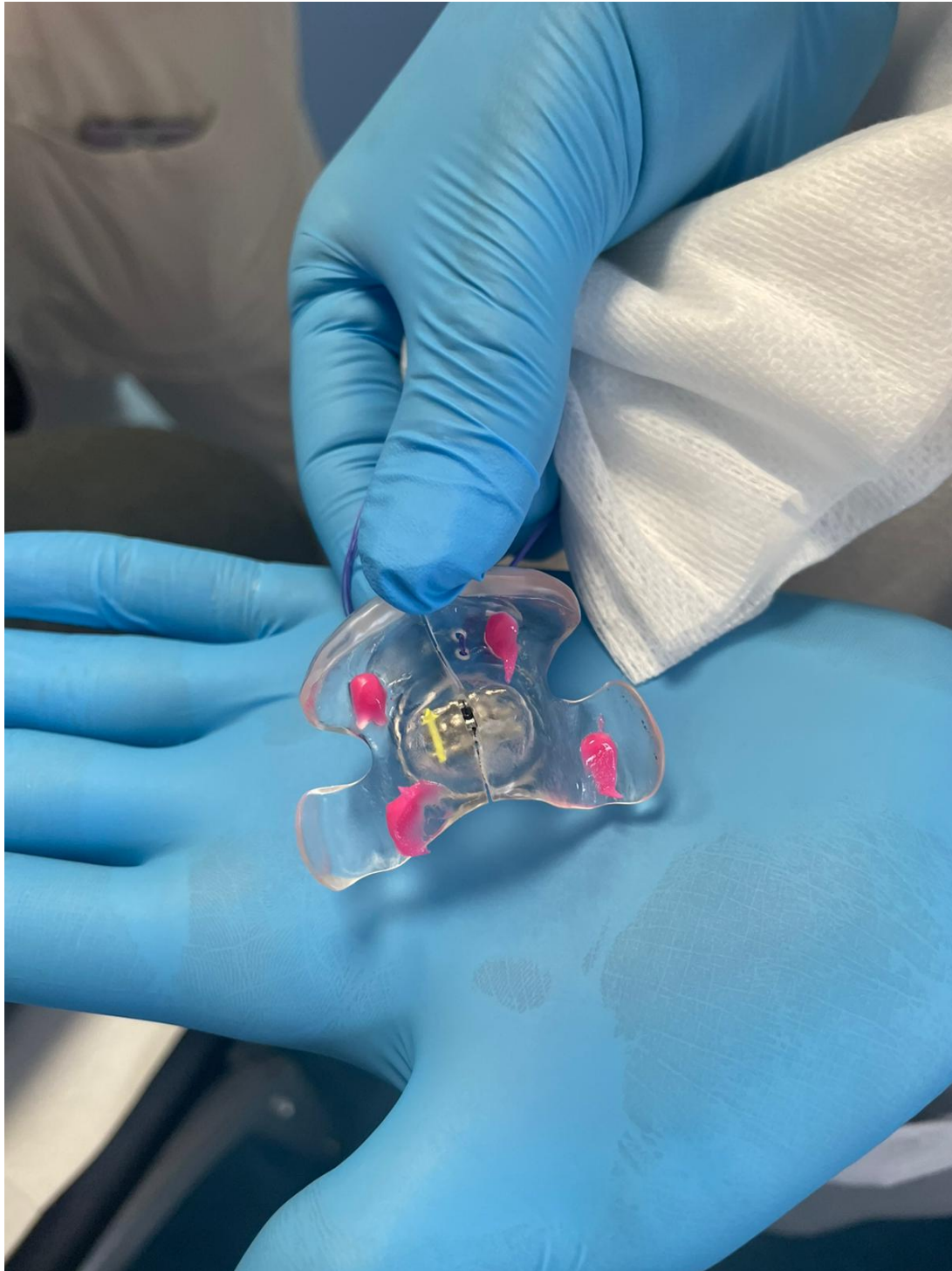


FIGURE 4. The palatal plate in addition to denture adhesive. The addition of the central screw is useful to increase the stability during the patient's growing up.

tions or ulcerated lesions within a few weeks and prevent long-term injury. Moreover, the appliance not only protects but also activates a distraction-behavioural mechanism [25]. While potentially obviating the need for invasive procedures and surgeries, oral appliances are prone to potential negative effects of infection if not properly maintained and cleaned. Dental impressions should be taken to customize the device for each individual's anatomy. In general, oral devices that are fixed within the oral cavity, and in particular devices such as the type described in this article, are best used in the comatose

or semicomatose patient population because of the potential for the device to stimulate a gag reflex or agitation secondary to the inability to fully move the tongue [26]. Removable devices allow for better hygiene but require patient collaboration with therapy; in the specific case of the paediatric patient, the real collaboration lies with the caregiver. Adequate follow-up is necessary to monitor healing and evaluate new injuries, particularly in young patients whose dentition is changing. To ensure good scar tissue quality, it is desirable to implement precise protocols to increase the elasticity of healing regions,



FIGURE 5. The healing after 4 days from the insertion of the plate. Fibrin has started to cover the ulcerated areas.



FIGURE 6. Healing 15 days after the insertion of the plate. The first signs of healing by second intention are observable.



FIGURE 7. The ulcerated areas were completely closed by second intention healing after 3 months.

TABLE 1. Types of treatment and expected effects provided for oral self-inflicted accidental trauma.

Type of treatment	Expected effect
Antibiotic therapy [16]	To prevent superinfections
Tricyclic antidepressants/neurotransmitter agonist-antagonist/anticonvulsants [17]	To treat dopaminergic dysfunction and/or serotonin system dysfunction but with the disadvantage to induce the patients in a state of chronic stupor
Intraoral devices (hard acrylic resin plates, thermoplastic plates, or mouth guards) [18]	First remedy for the management of self-inflicted injuries
Botulinum toxin an injections (BTX-A) [19–21]	Has shown promising results as a safe and effective treatment for self-induced trauma, resulting in decreased muscle strength and less frequent action, also if injected directly in masseter muscle
Surgical-resorptive approach in severe and refractory cases [22]	To prevent chronic injuries or to restore already self-inflicted deep injuries

improve their function, and decrease the risk of hypertrophic scar formation [25, 26]. Extraction of the entire dentition is an extreme but effective method to prevent mutilation. Alternatively, some surgeons have created open bites in patients to prevent contact and preserve tissue, but this treatment subjects patients to an orthognathic surgical procedure that may result in additional complications in bone healing due to chronic nutritional deficits [22].

Although the surgical approach may be the most easily resolved route, it represents a solution with a high biological and irreversible cost. Early extraction of deciduous elements, in the case of the paediatric patient, results in deficits in orthognathic growth and speech development; the use of a more conservative method allows a lower biological cost while not precluding other therapeutic approaches [27].

4. Conclusions

In conclusion, although the oral injury was caused by the underlying neurological pathology, the presence of this clinical sign created some difficulty within the dentistry role. Although there is no standardized protocol for the treatment of patients with neurological disorders who self-inflict trauma to the oral mucosa and tongue, it is extremely useful to consider the use of intraoral devices to enable healing of traumatic injuries and to prevent future trauma but more importantly to ensure a conservative approach. Indeed, this treatment, in combination with psychological therapy and/or drug treatment, prevents these patients from incurring extreme surgical treatments such as dental extractions to prevent self-mutilation. Finally, it is highly important to know that the presence of an attentive caregiver is crucial for early and rapid detection for patients with this problem.

AVAILABILITY OF DATA AND MATERIALS

The data are contained within this article.

AUTHOR CONTRIBUTIONS

AG and MC—designed the research study. AMGB and GV—performed the research. PF and AP—analysed the data. FM—

performed surgery. SV—performed histological examination. LFT—performed imaging. MC and FA—wrote the manuscript. All authors contributed to editorial changes in the manuscript. All the authors have read and approved the final manuscript.

ETHICS APPROVAL AND CONSENT TO PARTICIPATE

The study was conducted in accordance with the Declaration of Helsinki. It does not require approval by the Institutional Review Board or Ethics Committee since the current Regulation of the Ethics Committee of the Higher Institute of Health stipulates that projects with epidemiological, medico-social and evaluative contents need evaluation, approval and monitoring of trial protocols only if they contain personal data according to the legislative decrees on clinical trials and function of the ethics committees (decreto legislativo 24 giugno 2003, n.211; decreto ministeriale 8 febbraio 2013). The official definition of “personal data” is given by the National Data Protection Authority (Garante per la Protezione dei Dati Personali, <https://www.garanteprivacy.it/home/diritti/cosa-intendiamo-per-dati-personali>—Regolamento (UE) 2016/679 art.9 (accessed on 07 May 2023). The term “personal data” includes information about first and last names, images, social security codes, IP addresses and licence plate numbers. The images and radiographs in the manuscript are anonymized for this reason. The patient’s parents signed informed consent forms and were given authorization to anonymize the case documentation for scientific purposes.

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CONFLICT OF INTEREST

The authors declare no conflict of interest.

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