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Oral self-injury. An update

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Abstract – *Background*: Self-inflicted oral injuries of organic origin are particularly common in certain diseases, syndromes, and systemic disorders. In this article, we discuss the characteristics of these oral lesions and their treatment. *Literature Search*: The authors have reviewed the most relevant literature relating to oral self-injury through a search in textbooks and published articles included in the Medline database for the years 1970–2010, and selected published cases from the last two decades. *Results*: The majority of the literature on oral self-injury is in the form of case reports. Self-injury is particularly prevalent in patients with Lesch–Nyhan syndrome, a heterogeneous group of neurological disorders, congenital insensitivity to pain with anhidrosis, and mental retardation. It is most common in males in the early years of life, and the sites most frequently involved are the lower lip and the tongue. Therapeutic approaches in these patients have included psychological and pharmacological treatment, intraoral devices, and surgical procedures. *Clinical Implications*: Intraoral devices are the best therapeutic option for self-injury of organic origin, although this approach is not free of complications. The current lack of standardized treatment protocols for oral self-injury means that therapy must be individualized.

Definition and prevalence

Self-injury or self-mutilation is defined as a behavioral disturbance that consists of deliberate destruction of or damage to body tissues that is not associated with a conscious intent to commit suicide (1). For a lesion to be classified as a non-suicidal self-inflicted injury, it must satisfy the following characteristics: it must be considered socially unacceptable (in order to differentiate it from other behaviors such as piercing), direct (to differentiate it from lesions induced by indirect methods such as the deliberate ingestion of a drug), repetitive, and have produced mild or moderate damage (2).

The most common forms of self-injury include cuts, burns, scratches, blunt injury, bites, and interference with wound healing (3). The anatomical regions most frequently affected are the head, particularly the oral and perioral tissues, the hands, and the neck (4).

Although the prevalence of this behavior in the general population has not been fully defined, it has been suggested that it may affect around 750 individuals per 100 000 population (5). However, other studies have indicated much higher figures, of up to 4% in adults (2), 17% to 38% in university students (6, 7), and up to 69% among young people considered to be of high risk (homeless and runaway youths, substance abusers, victims of sexual abuse) (8). The reported prevalence among groups of psychiatric patients varies between 21% and 82% (2, 9), and the figure in institutionalized individuals with mental retardation are estimated to be between 7.7% and 22.8%(10); although the prevalence in those living in the community is unknown, it has been suggested that it does not exceed 2% (11).

Risk factors

Self-injury is an important health problem that can affect individuals of any age, sex, or ethnic group, and its frequency is increasing among adolescents and young adults (12). Although it has been suggested that any cause of discomfort, such as sinusitis, toothache, or headache, can precipitate self-injurious behavior (13), it is particularly common in certain diseases, syndromes, and disorders (Table 1).

The prevalence of lesions varies between the disorders and remains to be determined in many of these conditions. However, it is generally agreed that self-injury is very common among patients with Lesch–Nyhan syndrome, congenital insensitivity to pain with anhidrosis, Cornelia de Lange syndrome, and mental retardation (the most frequent form of self-injury in this last disorder is biting) (10).

Etiology and pathogenesis

Oral self-injury may be classified as functional or organic (14). In functional self-injury, the individual deliberately provokes the injury using a method aimed at attracting

Table 1. Principal syndromes and conditions that favor the appearance of self-mutilation

Lesch–Nyhan syndrome	Mental disorders (depression, obsessive compulsive disorder)
Mental retardation	Congenital insensitivity to pain with anhidrosis (CIPA)
Moebius syndrome	Infectious diseases (encephalitis)
Munchausen syndrome	Individuals with damage to the cerebral cortex, hypothalamus, reticular or pyramidal system, coma, etc.
Riga–Fede disease	Pharmacological or recreational drug reactions
XXY syndrome	Individuals who cannot manifest their stress or discomfort to their parents or carers
Gilles de la Tourette syndrome	Others: persons under great stress, prisoners, homeless, etc.
XXXXXY syndrome	
Cerebral palsy	
Rett syndrome	
Autism	
Epilepsy	
Cornelia de Lange syndrome	

attention. In cases of organic origin, individuals inflict the injury unconsciously, in a compulsive manner and with no specific intent.

Functional self-injury may represent a method of manipulation or a true means to seek help (1, 10). In general, it is considered that the onset and persistence of self-injurious behavior is attributable to automatic and social reinforcement that may be positive (e.g., to achieve a certain result) or negative (e.g., to avoid a specific situation). Lloyd-Richardson et al. (15), in an anonymous survey performed in 2007 on 633 randomly selected adolescents (mean age, 15.3 years) in the United States, found that the most common forms of negative reinforcement reported by individuals with self-injury were to interruption unpleasant feelings or lethargy, to avoid school or work, or to avoid having to perform an unpleasant task; the principal positive reinforcements detected were to attract attention, to achieve control of a situation, and to pass the time when the individuals were alone.

A number of etiologic and pathogenic models have been proposed to explain organic self-injury, the most widely known being the dopaminergic, opioid, and serotoninergic models.

In studies performed in rats, it was found that the frequency of self-injury increased after the administration of dopaminergic drugs (10) and that dopamine antagonists helped to control this behavior (4, 10). This led to the proposal of the hypothesis that patients presenting self-injurious behavior, particularly those with Lesch–Nyhan or Gilles de la Tourette syndrome, expressed marked hypersensitivity of dopaminergic receptors (16–18).

In another theory, it was proposed that pain provoked by self-mutilation induces a high level of endogenous opiate release, creating a form of addiction to the phenomenon in affected individuals (10). Another possible opiate-mediated mechanism involves an alteration of opiate metabolism in which increased endogenous opiate release is required to maintain an adequate opiatergic tone (19). The increase in endogenous opiate activity may partially explain the common clinical observation that associates episodes of self-mutilation with an apparent analgesia or insensitivity to pain and with certain dissociative states (20). Moreover, it has been reported that the administration of opiate antagonists such as naloxone or naltrexone can have favorable effects in patients presenting self-injurious behavior (21).

Dysfunction of the serotonergic system has also been implicated in the development of self-injury, and a decreased serotonin receptor binding index has been detected in deliberate self-harm patients (22). In support of this concept, patients with Lesch–Nyhan syndrome and self-mutilation obtained clinical benefit from treatment with a serotonin precursor (5-hydroxytryptophan) (23). However, subsequent studies have not consistently confirmed these findings (24). Posteriorly, it was suggested that fluoxetine, a drug that blocks the reuptake of serotonin, may decrease self-injury in mentally disabled, but the authors emphasize the need for future well-controlled clinical trials (25).

Oral lesions

Biting is the most common mode of self-inflicted injury, most frequently involving the oral and perioral regions and the hands (Fig. 1a,b) (10, 11). In the literature, there are no large series on oral self-inflicted injury, with the exception of those published by Amano et al. (26) and

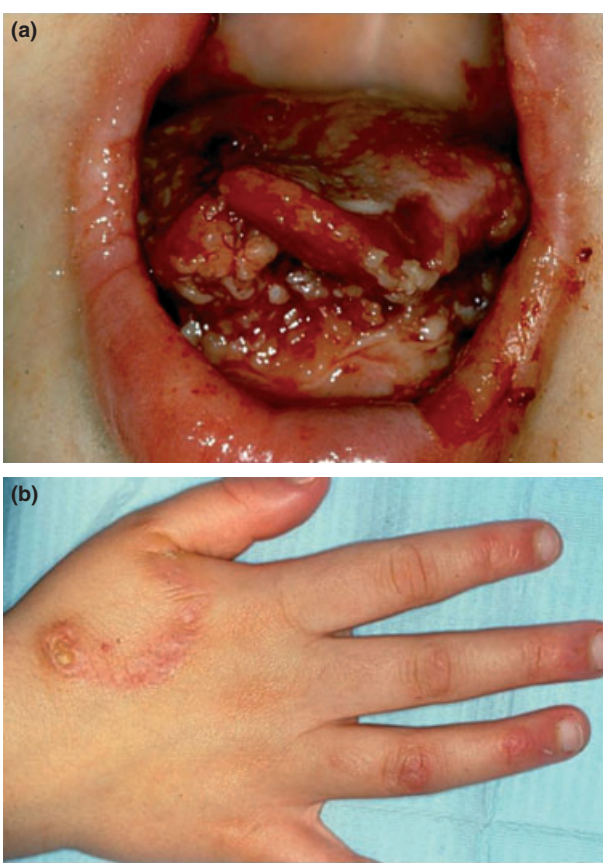

Fig. 1. Dramatic self-inflicted injury involving the tongue in a CATCH-22 syndrome patient (a), and hand biting in a patient with autism (b).

Bodner et al. (27) on patients with congenital insensitivity to pain with anhidrosis (autonomic neuropathy type IV), with 18 and 24 cases, respectively. We have therefore reviewed the most relevant case reports published in the past 20 years (a total of 30 patients) (4, 10, 17, 28–53) to analyze the characteristics of oral self-injury. The most common underlying diseases were Lesch–Nyhan syndrome, a heterogeneous group of neurological disorders (congenital, post-traumatic, and degenerative), congenital insensitivity to pain with anhidrosis, and mental retardation. Lesions may appear at any age, although the first 2 years of life are particularly important. This behavior is more common in males (ratio, 2:1). The sites most frequently affected are the lower lip and the tongue. There have been occasional case reports in which the soft tissue lesions are associated self-inflicted tooth dislocation or even avulsion. Oral self-injury is associated with lesions in other areas of the body in 20% of cases. Many of the oral findings coincide with those described by Bodner et al. (27) in their series of patients with congenital insensitivity to pain with anhidrosis, allowing us to speculate that the pattern of intraoral self-injury is not specific to the underlying disease.

Treatment

A number of therapeutic modalities have been tried in patients with self-injurious behavior. They may be classified into 4 groups: psychological treatment, pharmacological treatment, intraoral devices, and surgical procedures.

Psychological treatment

This should be one of the first therapeutic options to be considered by the health professional as it has been shown that some cases respond favorably and it does not have the adverse effects of pharmacological treatment nor is it as aggressive as intraoral devices (10, 54).

A psychological approach is essential in patients with functional self-injurious behavior, and its aim is to determine the reason why the patient behaves in this way and to design a specific therapeutic strategy. In a recently published epidemiologic study, Klonsky(3) evaluated 439 randomly selected individuals and found a prevalence of non-suicidal self-injury of 5.9%. A history of psychological therapy for emotional problems was detected in 39% of affected individuals. Lloyd-Richardson et al. (15), in a series of adolescents in the United States, found that 14.8% of participants with no history of self-injury had received psychological therapy compared with 25.2% of those who had a history of mild self-injury and 40.6% of those with moderate or severe self-injury.

The most widely used behavioral techniques are positive reinforcement, punishment, overcorrection, and alternative sensory activities (54). In individuals with mental retardation, the method usually applied is positive reinforcement (55). In patients with Lesch–Nyhan syndrome, the situation is more complex as these patients often understand what is happening to them and wish not to do it but are incapable of stopping; they are frequently afraid of their own actions and ask to be immobilized to restrict their movements, particularly at night (28, 56, 57). In general, behavioral therapy is of limited efficacy both in individuals with moderate-to-severe mental retardation and in patients with Lesch–Nyhan syndrome, and it must usually be combined with other therapeutic approaches such as pharmacological treatment or physical restraint.

Pharmacological treatment

Allopurinol, a xanthine oxidase inhibitor, has been used in patients with Lesch–Nyhan syndrome. This drug reduces uric acid levels, thereby increasing the life expectancy of these patients, although it has no effect on the neurological component of the syndrome and thus does not modify the self-injurious behavior (56, 57).

Tricyclic antidepressants such as clomipramine and desipramine have also been used. Leonard et al. (58) compared these 2 drugs and demonstrated that clomipramine (also used in the treatment of obsessive-compulsive disorder) was more effective in patients with onychophagia.

Based on the possible role of certain neuropeptides, neurotransmitters, and opiates in the etiology and pathogenesis of self-injurious behavior, it has been proposed that certain agonists, antagonists, or derivatives of these substances could be used to modify this behavior (59). Dopamine-receptor antagonist antipsychotic agents such as haloperidol, fluphenazine, and clozapine have been used in an attempt to compensate a possible deficiency of dopamine in the nucleus striatum (10). Some cases have been treated with anticonvulsants such as carbamazepine, probably for their capacity to reduce norepinephrine and dopamine turnover (60). Serotonin agonists such as fluoxetine, hydroxytryptophan and trazodone have also been used, although in some cases they did not lead to an improvement in the self-injurious behavior (61).

Tintner and Jankovic (62) observed that in cases of bruxism and other forms of oromandibular dystonia, there was a reduction in the biting activity after the injection of botulinum toxin into the masseter muscles. On this basis, botulinum toxin has been used in recent years to treat self-injurious behavior associated with biting (40). Its mechanism of action is not fully understood, as the mere inhibition of acetylcholine release from the peripheral nerve terminals would reduce the force of biting, although without altering the frequency of self-injury. It has been suggested that the toxin may inhibit the release of neuropeptides and neurotransmitters, such as substance P and glutamate (63, 64). Recently, the authors of the present report used botulinum toxin to treat a patient with Lesch–Nyhan syndrome with lesions of the tongue and lips owing to biting, but no favorable response was achieved.

In summary, the majority of drugs tested to date have only been effective in a very small number of patients and usually have minimal or no effect. In general, the benefits of their administration do not outweigh their adverse effects (such as excessive sedation of the patient), and tolerance to the drug is likely to develop (65, 66).

Restrictive oral devices

Physical restraint is the only method that enables self-injury to be prevented directly and in an individualized manner. The possible options include helmets, face masks, gloves, special clothing such as straitjackets, and, in the case of lesions owing to biting, restrictive oral devices (10).

Intraoral devices basically work through 2 mechanisms: they impede the patient from performing certain habits, helping to repress this behavior, and they represent a direct barrier that prevents the patient from being able to bite the oral tissues (e.g., maintaining the lip at a distance from the dental arches). It has been suggested that intraoral devices to prevent self-injury should satisfy the following conditions (37, 46, 67): they should maintain the injured tissues at a distance from the dental arches, so that they do not suffer further injury; they must not provoke new lesions; they should permit full movement of the mandible; they should not interfere with routine oral hygiene; they should be easy to manufacture and insert into the mouth, without causing discomfort to the patient; they should facilitate healing of the oral tissues; and they should be able to withstand the forces exerted by the oral structures without being displaced or breaking. Acrylic splints held in place by orthodontic bands have been designed to displace the lip in a vestibular direction by means of a plate also formed of acrylic material; there are also acrylic splints with oral shields that are inserted into the buccal or labial sulcus, soft buccal protectors positioned over the teeth, oral splints held in place by extraoral straps, and face masks anchored around the neck.

Literature from the past 2 decades shows that mouthguards (Fig. 2a,b) have been the most widely used devices, although with variable results. They have been reported to be effective in some cases (39, 41), to have failed in others (37, 43), and in a large number of cases, the effect was unknown or was only described over a short period (29, 42, 43, 49, 50). Another option is the acrylic splint that can be positioned directly on the teeth (Fig. 3a,b) (35), cemented to the teeth (44, 52), or held in place by extraoral straps (31) or by retention loops of orthodontic wire (30). The fixation systems for these splints increase their stability and prevent the patient from removing them; for this reason, it is widely agreed that they are more effective than mouthguards. A number of variants have been described, such as the joining of the upper and lower parts of the splint in a single unit, with a small central orifice to permit respiration; this model prevents injury both to the lip and to the tongue (45). Another device frequently used is the 'lip bumper' (Fig. 3a,b). This consists of one long element and a small acrylic shield, fixed by intermaxillary wires or elastic bands to tubes or bands anchored to the first molar teeth. These devices act by displacing the lip downwards and forwards to prevent it being bitten, and they have been used successfully in a number of cases described in the literature (10, 32, 41, 48), although in some cases follow up was not performed(48) or was of less than 3 months (41). The oral screen has the advantage that it does not have to be fixed to the teeth, and it may therefore be indicated in infants and children in whom tooth eruption is not advanced (43). Other

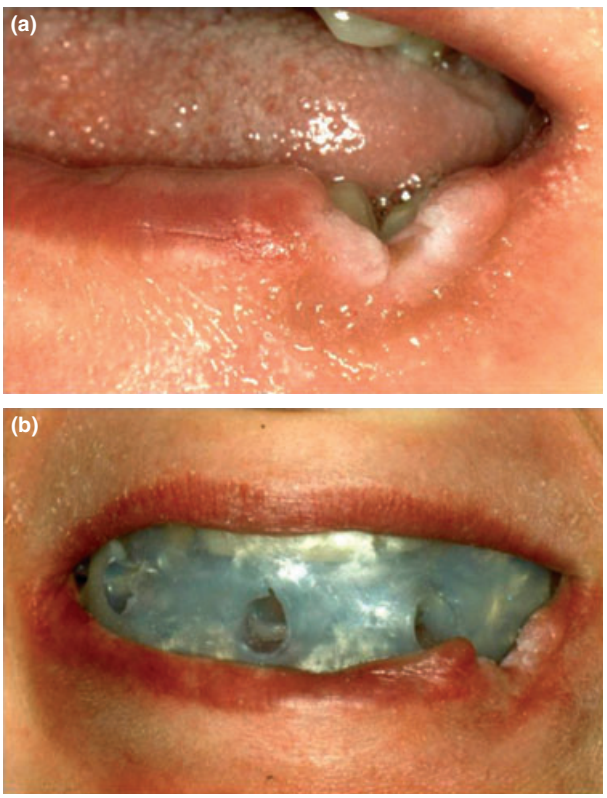

Fig. 2. Self-inflicted lip ulcer related to post-traumatic brain damage (a), treated with a mouthguard (b).

intraoral devices have been developed to treat specific situations. For example, Lucavechi et al. (4) designed a removable mandibular device with lateral acrylic shields to prevent a patient from introducing his fingers or other objects into his mouth; these actions had caused marked gingival recession. The authors suggested that this device could also be effective to prevent biting of the mucosa of the cheek.

Intraoral devices are not free of adverse effects, although they are not usually severe and are reversible. The devices interfere with oral hygiene, favor the appearance of fungal infections, and can provoke new lesions. Frequent readjustments are required, and the manufacturers require some time to produce the devices, limiting their application in urgent situations (57).

Surgical procedures

In some patients, the severity of self-injury and the failure of other treatment modalities lead to a need to consider tooth extraction as a therapeutic alternative. Although this is a radical solution, it has been shown to produce an enormous reduction in damage to the soft tissues, and some patients in whom dental extraction has been postponed have developed significant deformity of the oral and perioral tissues (68). In young patients, temporary teeth are extracted simultaneously or in sequence (16, 20, 40). Eruption of the definitive teeth requires re-evaluation and, when necessary, further extractions (57).

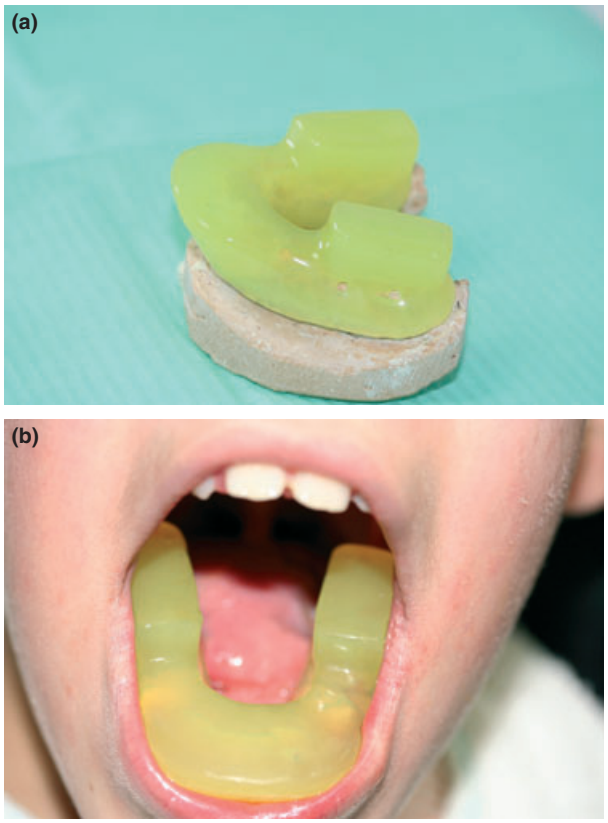

Fig. 3. Acrylic splint incorporating a 'lip bumper' (a), directly positioned on the teeth in a Lesch–Nyhan syndrome patient (b).

There was a case report in which a woman with cerebral palsy and mental retardation underwent orthognathic surgery to create an open bite and avoid self-injury owing to biting (69).

Oral self-injury in elderly patients

To date, no epidemiological studies have been published on the prevalence of self-inflicted oral lesions in elderly patients, which could suggest that the prevalence is low.

Neuropathologic mastication is caused by uncoordinated myotonic activity of the masticatory muscles and tongue following neuronal damage. Although it has been reported in the context of head injury, septic shock, and hypoxia (70), it is not uncommon in elderly patients with degenerative neurological diseases. In those patients, the pain produced by the swelling and injury of the oral mucosa may stimulate further uncontrolled, reflex, neuropathologic mastication, resulting in additional and self-perpetuating injury (71).

Degenerative neurological diseases can also favor the appearance of oral self-injury. In patients with Parkinsonism, orofacial dyskinesias such as 'flycatcher tongue' and lip pursing are side effects of levodopa and bromocriptine. These involuntary movements can lead to self-inflicted biting (72). In Alzheimer's disease, injury owing to apraxia is not uncommon and may present with traumatic oral ulcers and broken teeth; oral dyskinesia owing to antipsychotic medication can also produce self-inflicted mucosal lesions (72).

We have had a number of elderly patients with self-inflicted oral lesions owing to cheek biting, a form of self-injurious behavior that has been included in DSM-IV-TR under the category of impulse control disorders not otherwise specified (73). The true prevalence of cheek-biting disorder is not known (74). However, in a cross-sectional study conducted in Germany that included 655 subjects of 35 to 44 years of age and 1367 subjects of 65 to 74 years, lip and/or cheek biting was found to be less prevalent in the older group (1.9% vs 10.1% in the younger age group) (75).

In our experience, elderly patients tolerate removable restrictive oral devices less well than younger subjects and show a lower level of compliance. As a result, it must be ensured that removable protective mouthguards are very stable, or else fixed devices should be used. For uncooperative patients, particularly those with severe cognitive deterioration, tooth extractions are often be the treatment of choice.

Clinical guideline

Both the general state of health of patients with oral self-injury and the characteristics of the specific lesions are very variable, making it difficult to establish a common clinical protocol (48). However, some authors have attempted to standardize treatment in these patients (70). Family members and caregivers can play an important role in the early diagnosis of self-inflicted oral lesions; because of the high pain threshold and communication problems of many of these patients, lesions are only detected when they bleed, when they are large, or when they affect easily visible sites. Caregivers can be understandably distressed both by the appearance of the injury and the imagined discomfort for the patient (71). Monitoring of the lesion enables us to determine how and why it occurs, differentiating between functional and organic oral self-injury, and the information provided by caregivers is particularly useful. In some patients, it is difficult to differentiate between the two modes, and both neurological and psychiatric evaluations are necessary.

Antiseptics are usually administered to prevent or treat superinfection, although antibiotics have been used in some cases (10). It is important to maintain and, when necessary, improve oral hygiene in order to avoid superinfection and favor the healing process. Behavior control techniques prescribed by a specialist psychologist or psychiatrist are recommended if the disorder has a functional origin, and these may be combined psychoactive drug therapy when necessary. When there is an organic origin, filing down the tooth cusps and/or the incisal borders of the anterior teeth should be considered (37). In cases in which this therapeutic modality is not indicated or is ineffective, intraoral devices should be used, possibly in combination with pharmacological treatment, usually prescribed by a neurologist. The main problems associated with intraoral devices are the following: they require a degree of patient collaboration to take dental impressions and to insert retention elements (bands, tubes, or composite resin retainers), which sometimes has to be performed under pharmacological sedation or even under general anesthesia; they

may provoke new lesions, thus requiring follow up of the patient; and their stability may be compromised by movements of the oral structures and, in younger patients, by tooth eruption.

If the above methods do not work, serial tooth extraction is undertaken as a final option, with the exception of elderly patients with neurodegenerative diseases with a poor prognosis where tooth extraction represents a common therapeutic option.

Conclusion

The greater part of the literature available on oral self-mutilation is in the form of case reports, making it very difficult to establish widely applicable clinical protocols; the therapeutic approach therefore typically needs to be determined on an individual basis. In our experience, intraoral devices alone or in combination with pharmacological treatment offer the best management option for oral self-injury of organic origin. Further prospective studies must be designed with sufficient follow up, selecting patients with similar systemic conditions, to draw up guidelines for the therapeutic approach to oral self-mutilation.

Conflicts of interest

Authors declare that they have no conflicts of interest in the authorship or publication of this contribution.

References

1. Favazza AR. Why patients mutilate themselves. Hosp Community Psychiatry 1989;40:137–45.
2. Briere J, Gil E. Self-mutilation in clinical and general population samples: prevalence, correlates, and functions. Am J Orthopsychiatry 1998;68:609–20.
3. Klonsky ED. Non-suicidal self-injury in United States adults: prevalence, sociodemographics, topography and functions. Psychol Med 2011;5:1–6.
4. Lucavechi T, Barbería E. Self-injurious behavior in a patient with mental retardation: review of the literaure and a case report. Quintessence Int 2007;38:393–8.
5. Favazza AR, Conterio K. The plight of chronic self-mutilators. Community Ment Health J 1988;24:22–30.
6. Gratz KL, Conrad SD, Roemer L. Risk factors for deliberate self-harm among college students. Am J Orthopsychiatry 2002;72:128–40.
7. Whitlock J, Eckenrode J, Silverman D. Self-injurious behaviors in a college population. Pediatrics 2006;117:1939–48.
8. Whitbeck LB, Chen X, Hoyt DR, Tyler KA, Johnson KD. Mental disorder, subsistence strategies, and victimization among gay, lesbian, and bisexual homeless and runaway adolescents. J Sex Res 2004;4:329–42.
9. Nock MK, Prinstein MJ. A functional approach to the assessment of self-mutilative behavior. J Consult Clin Psychol 2004;72:885–90.
10. Saemundsson SR, Roberts MW. Oral self-injurious behavior in the developmentally disabled: review and a case. ASDC J Dent Child 1997;64:205–9.
11. Rojahn J. Self-injurious and stereotypic behavior of noninstitutionalized mentally retarded people: prevalence and classification. Am J Ment Defic 1986;91:268–76.
12. Ross S, Heath N. A study of the frequency of selfmutilation in a community sample of adolescents. J Youth Adolesc 2002;31:67–77.
13. Loschen EL, Osman OT. Self-injurious behavior in the developmentally disabled: assessment techniques. Psychopharmacol Bull 1992;28:433–8.
14. Singh P. Three Paediatric Patients with Oral Self-Mutilation-A Report. Dent Update 2008;35:280–3.
15. Lloyd-Richardson EE, Perrine N, Dierker L, Kelley ML. Characteristics and functions of non-suicidal self-injury in a community sample of adolescents. Psychol Med 2007;37:1183–92.
16. Medina AC, Sogbe R, Gómez-Rey AM, Mata M. Factitial oral lesions in an autistic paediatric patient. Int J Paediatr Dent 2003;13:130–7.
17. Silva DR. Self-injurious behavior as a challenge for the dental practice: a case report. Pediatr Dent 2003;25:62–6.
18. Tatakis DN, Milledge JT. Severe gingival recession in trisomy 18 primary dentition. A clinicopathologic case report of self-inflicted injury associated with mental retardation. J Periodontol 2000;71:1181–6.
19. Stanley B, Sher L, Wilson S, Ekman R, Huang YY, Mann JJ. Non-suicidal self-injurious behavior, endogenous opioids and monoamine neurotransmitters. J Affect Disord 2010;124:134–40.
20. Antonia S, Stanley B. An opioid deficit in borderline personality disorder: self- cutting, substance abuse, and social dysfunction. Am J Psychiatry 2010;167:882–5.
21. Agarwal LJ, Berger CE, Gill L. Naltrexone for severe self-harm behavior: a case report. Am J Psychiatry 2011;168:437–8.
22. Audenaert K, Van Laere K, Dumont F, Slegers G, Mertens J, vaan Heeringen C et al. Decreased frontal serotonin 5-HT 2a receptor binding index in deliberate self-harm patients. Eur J Nucl Med 2001;28:175–82.
23. Mizuno T, Yugari Y. Self mutilation in the Lesch-Nyhan syndrome. Lancet 1974;1:761.
24. Anderson LT, Herrmann L, Dancis J. The effect of L-5-hydroxytryptophan on self-mutilation in Lesch-Nyhan disease: a negative report. Neuropadiatrie 1976;7:439–42.
25. Ricketts RW, Goza AB, Ellis CR, Singh YN, Singh NN, Cooke JC 3rd. Fluoxetine treatment of severe self-injury in young adults with mental retardation. J Am Acad Child Adolesc Psychiatry 1993;32:865–9.
26. Amano A, Akiyama S, Ikeda M, Morisaki I. Oral manifestations of hereditary sensory and autonomic neuropathy type IV. Congenital insensitivity to pain with anhidrosis. Oral Surg Oral Med Oral Pathol Oral Radiol Endod 1998;86:425–31.
27. Bodner L, Woldenberg Y, Pinsk V, Levy J. Orofacial manifestations of congenital insensitivity to pain with anhidrosis: a report of 24 cases. J Dent Child 2002;69:293–6.
28. Evans J, Sirikumara M, Gregory M. Lesch-Nyhan syndrome and the lower lip guard. Oral Surg Oral Med Oral Pathol 1993;76:437–40.
29. Sugahara T, Mishima K, Mori Y. Lesch-Nyhan syndrome: successful prevention of lower lip ulceration caused by self-mutilation by use of mouth guard. Int J Oral Maxillofac Surg 1994;23:37–8.
30. Cehreli ZC, Olmez S. The use of a special mouthguard in the management of oral injury self-inflicted by a 4-year-old child. Int J Paediatr Dent 1996;6:277–81.
31. Chen LR. Successful treatment of self-inflicted oral mutilation using an acrylic splint retained by a head gear. Pediatr Dent 1996;18:408–10.
32. Nurko C, Errington BD, Ben Taylor W, Henry R. Lip biting in a patient with Chiari type II malformation: case report. Pediatr Dent 1999;21:209–12.
33. Pantanowitz L, Berk M. Auto-amputation of the tongue associated with flupenthixol induced extrapyramidal symptoms. Int Clin Psychopharmacol 1999;14:129–31.
34. Pigno MA, Funk JJ. Prevention of tongue biting with a removable oral device: a clinical report. J Prosthet Dent 2000;83:508–10.
35. Coyne BM, Montague T. Teeth grinding, tongue and lip biting in a 24-month-old boy with meningococcal septicaemia. Report of a case. Int J Paediatr Dent 2002;12:277–80.

36. Shimoyama T, Horie N, Kato T, Nasu D, Kaneko T. Tourette's syndrome with rapid deterioration by self-mutilation of the upper lip. J Clin Pediatr Dent 2003;27:177–80.
37. Yasui EM, Kimura RK, Kawamura A, Akiyama S, Morisaki I. A modified oral screen appliance to prevent self-inflicted oral trauma in an infant with cerebral palsy: a case report. Oral Surg Oral Med Oral Pathol Oral Radiol Endod 2004;97:471–5.
38. Diab M. Self-inflicted orodental injury in a child with Leigh disease. Int J Paediatr Dent 2004;14:73–7.
39. Kobayashi T, Ghanem H, Umezawa K, Mega J, Kawara M, Feine JS. Treatment of self-inflicted oral trauma in a comatose patient: a case report. J Can Dent Assoc 2005;71:661–4.
40. Dabrowski E, Smathers SA, Ralstrom CS, Nigro MA, Leleszi JP. Botulinum toxin as a novel treatment for self-mutilation in Lesch-Nyhan syndrome. Dev Med Child Neurol 2005;47:636–9.
41. Cauwels RG, Martens LC. Self-mutilation behaviour in Lesch-Nyhan syndrome. J Oral Pathol Med 2005;34:573–5.
42. Jeong TS, Lee JH, Kim S, Kim JH, Tootla RG. A preventive approach to oral selfmutilation in Lesch-Nyhan syndrome: a case report. Pediatr Dent 2006;28:341–4.
43. Schalka MM, Corrêa MS, Ciamponi AL. Congenital insensitivity-to-pain with anhidrosis (CIPA): a case report with 4-year follow-up. Oral Surg Oral Med Oral Pathol Oral Radiol Endod 2006;101:769–73.
44. Guimarães LF. Self-inflicted oral trauma in a baby with Moebius syndrome. J Dent Child 2007;74:224–7.
45. Romero M, Simón R, García-Recuero JI, Romance A. Dental management of oral self-mutilation in neurological patients: a case of congenital insensitivity to pain with anhidrosis. Med Oral Patol Oral Cir Bucal 2008;23:37–8.
46. Kiat-Amnuay S, Koh SH, Powner DJ. An occlusal guard for preventing and treating self-inflicted tongue trauma in a comatose patient: a clinical report. J Prosthet Dent 2008;99:421–4.
47. Neves BG, Roza RT, Castro GF. Traumatic lesions from congenital insensitivity to pain with anhidrosis in a pediatric patient: dental management. Dent Traumatol 2009;25:545–9.
48. El Bouihi M, El Fares N, Slimani F, Bennis K, Benjelloun A, Chekkoury Idrissi A. A maxillary lip bumper used to manage labial self-mutilation. Rev Stomatol Chir Maxillofac 2009;110:233–5.
49. Almeida D, da Costa KN, Castro RdeA, Almeida ML, Vianna R, Antonio AG. Selfinflicted oral injury in an infant with transverse myelitis. Spec Care Dentist 2009;29:254–8.
50. Hutton A, McKaig S. The dental management of a child with congenital insensitivity to pain. Dent Update 2010;37:180–2, 185.
51. Arhakis A, Topouzelis N, Kotsiomiti E, Kotsanos N. Effective treatment of selfinjurious oral trauma in Lesch-Nyhan syndrome: a case report. Dent Traumatol 2010;26:496–500.
52. Suchak A, Mars M. Case report: a unique presentation of severe tongue biting in 10 month-old twins with a novel approach to management. Eur Arch Paediatr Dent 2010;11:149–50.
53. Munerato MC. Self-mutilation of tongue and lip in a patient with simple schizophrenia. Clin Med Res 2011;9:42–5.
54. Winchel RM, Stanley M. Self-injurious behavior: a review of the behavior and biology of self-mutilation. Am J Psychiatry 1991;148:306–17.
55. Griffin JC, Williams DE, Stark MT. Self-injurious behavior: a statewide prevalence survey of the extent circumstances. Appl Res Met Ret 1986;24:37–46.
56. Christie R, Bay C, Kaufman IA, Bakay B, Borden M, Nyhan WL. Lesch-Nyhan disease: clinical experience with nineteen patients. Dev Med Child Neurol 1982;24:293–306.
57. Cusumano FJ. Prevention of self-mutilation in patients with Lesch-Nyahn syndrome: review of literature. ASDC J Dent Child 2001;68:175–8.
58. Leonard HL, Lenane MC, Swedo SE, Rettew DC, Rapoport JL. A double-blind comparison of clomipramine and desipramine treatment of severe onychophagia (nail biting). Arch Gen Psychiatry 1991;48:821–7.
59. Konicki PE, Schulz SC. Rationale for clinical trials of opiate antagonists in treating patients with personality disorders and self-injurious behavior. Psychopharmacol Bull 1989;25:556–63.
60. Roach ES, Delgado M, Anderson L, Iannaccone ST, Burns DK. Carbamazepine trial for Lesch-Nyhan self-mutilation. J Child Neurol 1996;11:476–8.
61. Frith CD, Johnston EC, Joseph MH, Powell RJ, Watts RW. Double-blind clinical trial of 5-hydroxytryptophan in a case of Lesch-Nyhan syndrome. J Neurol Neurosurg Psychiatry 1976;39:656–62.
62. Tintner R, Jankovic J. Botulinum toxin type A in the management of oromandibular dystonia and bruxism. In: Brin MF, Jankovic J, Hallet M, eds. Scientific and Therapeutic Aspects of Botulinum Toxin. Philadelphia: Lippincott Williams & Wilkins; 2002. p. 343–50.
63. Purkiss J, Welch M, Doward S, Foster K. Capsaicin-stimulated release of substance P from cultured dorsal root ganglion neurons: involvement of two distinct mechanisms. Biochem Pharmacol 2000;59:1403–6.
64. Aoki KR. Pharmacology and immunology of botulinum toxin serotypes. J Neurol 2001;248(Suppl. 1):3–10.
65. Osman OT, Loschen EL. Self-injurious behavior in the developmentally disabled: pharmacologic treatment. Psychopharmacol Bull 1992;28:439–49.
66. Mercugliano M. Psychopharmacology in children with developmental disabilities. Pediatr Clin North Am 1993;40:593–616.
67. Hanson GE, Ogle G, Giron L. A tongue stent for prevention of oral trauma in the comatose patient. Crit Care Med 1975;3:200–3.
68. Dicks JL. Lesch-Nyhan syndrome: a treatment planning dilemma. Pediatr Dent 1982;4:127–30.
69. Macpherson DW. Orthognathic surgery for the treatment of chronic of self mutilization of the lips. Int J Oral Maxillofac Surg 1992;21:133–6.
70. Hallett KB. Neuropathological chewing: a dental management protocol and treatment appliances for pediatric patients. Spec Care Dentist 1994;14:61–4.
71. Scully C, Diz Dios P, Kumar N. Special Care in Dentistry. London: Churchill Livingstone, Elsevier; 2007.
72. American Psychiatric Association. Diagnostic and statistical manual of mental disorders, text revision, 4th edn. Washington, DC: American Psychiatric Association; 2000.
73. Sarkhel S, Praharaj SK, Akhtar S. Cheek-biting disorder: another stereotypic movement disorder? J Anxiety Disord 2011;25:1085–6.
74. Reichart PA. Oral mucosa lesions in a representative cross-sectional study of aging Germans. Community Dent Oral Epidemiol 2000;28:390–8.
75. Milwood J, Fiske J. Lip-biting in patients with profound neurodisability. Dent Update 2001;28:105–8.