CASE REPORT

Factitious buccal lesion secondary to bruxism in a child with cerebral palsy

D Harris

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Children with cerebral palsy are at greater risk of a whole range of oral conditions than their peers. These include bruxism (tooth grinding), oral skill dysfunction, gross malocclusion due to effects of the abnormal orofacial muscle tone on tooth eruption, drooling of saliva, and poor oral hygiene.¹ A challenging case of a painful buccal lesion in a 2 year old girl with cerebral palsy (CP) that did not respond to antifungal, antiviral or antibiotic treatment is presented as a factitious lesion. The recognition and significance of self-injurious behaviour and factitious lesions in children are discussed.

A 2 year old girl with severe cerebral palsy was brought to the Emergency Department by her mother. She had been born at 24 weeks gestation and had spent 4 months in the neonatal intensive care unit, ventilated for the first 40 days. She was confined to a modified pushchair and was fully cared for by her parents in carrying out of daily activities. She did not have salivary drooling and had an adequate swallow despite evidence of facial and hypoglossal nerve dysfunction.

The previous week her mother had noticed some blood on her pillow in the mornings and felt she appeared in pain whilst eating and drinking. She initially consulted her General Practitioner who prescribed Co-Amoxiclav for what was thought to be an infected buccal ulcer.

As there was no response to the treatment after 7 days the GP referred her to the Paediatric Emergency Department. At this time a lesion on the left buccal mucosa over 2 cm in diameter was noted, with a hyperkeratotic macerated border and a hyperaemic ulcerated base that bled on contact. It was thought to be a major aphthous ulcer. Bacteriology and virology swabs were taken from the base and the edge of the lesion plus the normal area of buccal mucosa. A course of oral Nystatin was prescribed, to be administered by droplet.

Seven days later the lesion was unchanged. One virology swab was positive for herpes simplex virus type 1, although it is not documented which one, and all other swabs showed no growth. It was also noted that the girl's mother had two small "cold sores" around her mouth. The girl was prescribed acyclovir and benzydamine hydrochloride (Diffiam) spray for oral comfort.

She returned to the department 3 weeks later as the lesion had increased in size and continued to cause pain, especially on eating. Within this period the GP had prescribed a second course of acyclovir.

At this point it was noted that when distressed the girl exhibited marked bruxism. On questioning, her mother reported that she had ground her teeth since they first erupted although the degree of grinding varied with her emotional state. She was noted to have all her deciduous teeth, including the deciduous molars on both sides and that

the blood stained saliva had been first noted about 2 months after the molars appeared.

A new diagnosis of a Factitious Oral Lesion (FOL) secondary to bruxism (molar grinding) and cheek chewing was considered and after discussion with a Consultant in Paediatric Dentistry she was referred to the Out Patient Clinic at The Eastman Dental Hospital in London where the diagnosis of FOL was confirmed. The management plan was to reassure the mother, observe the lesion, and consider the use of a mouth guard if it continued to enlarge.

At A&E review 7 months after the initial consultation the lesion had almost completely healed. This improvement had occurred as the girl's speech had progressed following intensive Speech and Language Therapy. Her bruxism may have been a means of attracting attention and had now receded as her other means of communication had improved.

DISCUSSION

Cerebral palsy shows a dynamic clinical pattern as the child grows and develops. All aspects of daily living are profoundly affected, the commonest problems being speech disorders, learning difficulties, hearing loss, sensory limitations, and epilepsy.¹ Communication problems do not necessarily equate with a low IQ or understanding,² and there are many different forms of non-verbal communication, some of which can be deleterious to the child's health.

Self-injurious behaviour (SIB) is deliberate harm to the body without suicidal intent, often involving repetitive actions that cause tissue damage.⁴ SIB comes under the umbrella term of challenging behaviour, along with aggressive, stereotyped (for example, rocking) and non-person-directed behaviour (for example, temper tantrums). There is an increased incidence of self-injurious behaviour and self-mutilation in children with learning difficulties,³ physical disability,⁵ and psychiatric disorders.⁶

The underlying causes of SIB are functional or structural in origin. Functional SIB may be used as a means of escape from responsibility or attention seeking and has been found to occur more often at times of stress, hunger, anger, and frustration,¹⁵ whilst structural causes are secondary to genetic defects and abnormal neurophysiology.⁷

SIB is more prevalent in females at around 750 per 100,000 in the general population⁸ but increasing to 23–40% in severely developmentally delayed individuals.⁹ Over 70% of autistic patients show self-injury at some time in their lives. SIB has also been reported in children with Lesch-Nyhan, XXY syndrome, encephalitis, congenital malformations, trisomy 18, as well as several other syndromes plus epileptic, and even unconscious patients;⁷ the majority of reported cases involve children less than 9 years of age.⁷

The end point of most types of SIB is the production of factitious (artefactual) lesions, over 70% of which appear on the head and neck.¹⁰ Within the oral cavity, injury in children can be caused by repeated biting of tissues, as in this case, or by applying foreign objects and fingernails to the gingiva or against other oral structures. These actions result in

hyperkeratosis, maceration, ulceration, gingivitis, periodontitis, and even dental self-extraction.⁸

The common differential diagnoses of oral lesions are aphthous stomatitis, intraoral herpes, and erythema multiforme (Stevens Johnson Syndrome) all of which tend to be acute in onset and short-lived.¹¹ The diagnosis of a factitious lesion is one of exclusion and very often overlooked. The common pathologies can usually be ruled out by prodromal signs and symptoms, lesion location and appearance, plus the directed use of haematological, microbiological, and if necessary, histological investigations. A factitious diagnosis must be considered for unexplained atypical or persistent lesions especially where the irritant factor can be deduced—for example, cheek chewing or access to the right gingival margin in a right handed patient, and where possible a psychological or developmental aetiology established. Factitious illness (Munchhausen) by proxy must also be ruled out, particularly in more dependant patients.

In very young children with cerebral palsy, studies looking at oral hygiene, dental caries, malocclusion, and bruxism compared to normal age matched control children show no significant difference.¹ The differences in caries, malocclusion, and bruxism between normal children and those with cerebral palsy become more marked with age.¹² Children, as they become older, are expected to be more responsible in maintaining their own oral hygiene and their diets become more varied.

These factors, coupled with the increased incidence of pouching residual food, pica, and problems in tongue, lip, and cheek movement may predispose to mild repetitive behaviours causing structural factitious lesions.

In functional cases the treatment options include (in order of escalation): a “watch and wait” approach, behavioural modification techniques such as positive reinforcement,¹³ the use of oral splints (which are not well tolerated), pharmacological treatments (olanzapine),¹⁴ dental extractions and rarely maxillary osteotomy.⁸ The most severe lesions require a more accelerated movement through the options with early multidisciplinary input including paediatricians, child psychiatrists, and paediatric dentists or oral surgeons.

Self-induced or factitious injury of the oral mucosal tissues may present with a confusing clinical picture within the Emergency Department setting, especially in children with cerebral palsy and other causes of developmental delay for whom full examination is not always easy. In such cases SIB should always be considered as a primary diagnosis to avoid these painful and distressing lesions being diagnosed erroneously and unsuitable therapies with potentially harmful side effects being tried.

Correspondence to: Dan Harris, Department of Paediatric Emergency Medicine, St Mary's Hospital, London, UK; drdanharris@msn.com

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