



## CASE STUDY: CEREBRAL PALSY SLEEP BRUXISM

# Effect of a hyperbolide mastication apparatus for the treatment of severe sleep bruxism in a child with cerebral palsy: Long-term follow-up



Lilian Chrystiane Giannasi, PhD <sup>a,b,\*</sup>,  
Sandra Regina Freitas Batista <sup>a</sup>, Miriam Yumi Matsui, MS <sup>a</sup>,  
Camila Teixeira Hardt, MS <sup>a</sup>, Carla Paes Gomes, MS <sup>a</sup>,  
Jose Benedito Oliveira Amorim, PhD <sup>a</sup>, Claudia Santos Oliveira,  
PhD <sup>c</sup>, Luis Vicente Franco de Oliveira, PhD <sup>b</sup>,  
Monica Fernandes Gomes, PhD <sup>a</sup>

<sup>a</sup> Universidade Estadual Paulista "Julio de Mesquita Filho", Faculdade de Odontologia de São José dos Campos, UNESP, São Paulo, SP, Brazil

<sup>b</sup> Sleep Laboratory, Master and Doctoral Degree Program in Rehabilitation Sciences, Nove de Julho University, UNINOVE, São Paulo, SP, Brazil

<sup>c</sup> Movement Laboratory, Master and Doctoral Degree Program in Rehabilitation Sciences, Nove de Julho University, UNINOVE, São Paulo, SP, Brazil

Received 14 December 2012; received in revised form 3 May 2013; accepted 6 May 2013

### KEYWORDS

Sleep bruxism;  
Cerebral palsy;  
Electromyography;  
Hyperbola;  
Therapy;  
Dentistry

**Summary** *Purpose:* Sleep bruxism is common among the various oromotor alterations found in individuals with cerebral palsy (CP). Few studies have investigated the use of the mastication device denominated "hyperbola" (HB) and none was found describing the use of such a device for the treatment of bruxism in children with CP. The aim of the present study was to evaluate the effect of the HB on electromyographic (EMG) activity in the jaw-closing muscles and the reduction in sleep bruxism in a child with CP using surface EMG analysis before and after nine months of treatment. *Methods:* A seven-year-old boy with severe spastic CP and sleep bruxism was enrolled in this study. The HB was chosen as the treatment option for sleep bruxism in this case because the child did not accept an occlusal splint.

\* Corresponding author. R Esperança, 265, São Jose dos Campos, São Paulo 12243-700, Brazil. Tel./fax: +55 12 3951 0800.  
E-mail address: [odontogiannasi@uol.com.br](mailto:odontogiannasi@uol.com.br) (L.C. Giannasi).

The HB has a hyperbolic shape and is made of soft, non-toxic, odorless, tasteless silicone. There are five different sizes of HB manufactured based on the diversity of tooth sizes. This device produces proprioceptive excitation in the dentoalveolar nerve, spindles and Golgi tendon organs. HB has been employed for the treatment of temporomandibular disorder, abnormal oro-dental development, abnormal occlusion, xerostomy, halitosis and bruxism.

HB therapy was performed for 5 min six times a day over a nine-week period. Surface EMG of the mandible at rest and during maximum contraction was performed on the masseter and temporalis muscles bilaterally to evaluate electromyographic activity before and after nine months of HB usage.

**Results:** HB usage led to a visible tendency toward the reorganization of mastication dynamics, achieving a marked balance in electromyographic activity of the jaw-closing muscles and improving the child's quality of life.

**Conclusion:** Based on the findings of the present study, this noninvasive therapy may be useful for individuals with cerebral palsy due to its positive effects and low cost, which allows its use in the public health realm. Further clinical studies with a larger sample size are needed to validate these results and allow the development of a new treatment protocol for patients with spastic cerebral palsy.

© 2013 Elsevier Ltd. All rights reserved.

## Introduction

Cerebral palsy (CP) is a limiting condition involving the impairment of gait, cognitive function and fine and gross motor function as well as changes in the pattern of the muscles of mastication (Koman et al., 2004). Sleep bruxism in childhood has been widely investigated, but few studies have addressed the presence and treatment of this habit in children with CP (Rodrigues et al., 2003; Peres et al., 2007; Ortega et al., 2007; Zarowski et al., 2008; Miamoto et al., 2011).

Recent studies have shown that 15.29% of normal children exhibit sleep bruxism (Fonseca et al., 2011), while the prevalence among children with CP ranges from 25% to 32% (Zarowski et al., 2008; Miamoto et al., 2011). There is agreement that the treatment of sleep bruxism (SB) in this special population requires a combination of approaches, such as physical therapy, speech therapy and myofunctional therapy (Dougherty, 2009). Regarding the myofunctional approach, there is a safe, effective option known as the "hyperbola" (HB), which can be employed to treat temporomandibular disorders, abnormal oro-dental development, abnormal occlusion, xerostomy, halitosis and bruxism (Cheida, 1997). The HB has a hyperbolic shape with rounded apices and is made of soft, non-toxic, odorless, tasteless silicone. There are five different sizes (extra small, small, medium, large and extra large) (Fig. 1), manufactured based on the diversity of tooth sizes. After curing, the HB has 35 Shore A hardness, allowing non-traumatic exercises. Its hardness and texture are compatible with the ideal force applied during mastication. There are different Shore Hardness scales for measuring the hardness of different materials. The Shore A Hardness Scale measures the hardness of flexible mold rubbers that range in hardness from very soft and flexible (1–9), to soft (10–39), to medium and somewhat flexible (40–80), to hard with almost no flexibility at all (81–100) (Meththanandra et al., 2009).

The HB produces proprioceptive excitation in the dentoalveolar nerve, spindles and Golgi tendon organs. This

device causes jaw movements performed by a complex system of neuromuscular pathways controlled by sensory afferents of the oral tissues, muscles and joints, leading to muscle toning responses, the modulation of myoelectrical activity, stimulation of bones and adjacent structures (salivary glands) and the growth and development of the stomatognathic system (Cheida, 1997). There are no other devices scientifically designed for chewing exercises that achieve neural excitation levels with positive responses (Cheida, 1997). The HB was invented by a Brazilian dentist (patent no. 8901216-0) and is registered as a mastication apparatus at the National Institute of Intellectual Property. To facilitate the reader's understanding, the authors suggest the name "myofunctional device". This is the first study to investigate this HB.

The aim of the present study was to evaluate the effect of the HB on electromyographic activity in the jaw-closing muscles and the reduction of sleep bruxism in a child with cerebral palsy, as assessed through electromyography (EMG) before and after nine months of treatment.

## Case presentation

A seven-year-old boy with spastic quadriplegic CP caused by hypoxic-ischemic brain damage (also exhibiting kernicterus - a bilirubin-induced brain dysfunction) visited the Bioscience Center for Special Health Care Needs of the

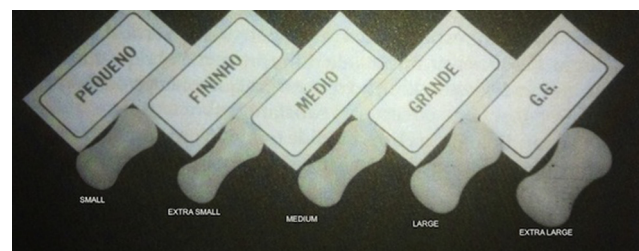


Figure 1 Different sizes of hyperbola.

State University of São Paulo (Brazil) for treatment. The patient was wheelchair-dependent, exhibited poor cognitive function, and was fed semi-solid food orally since 3 years of age. Ranitidine hydrochloride was used to treat gastroesophageal reflux, and botulinum toxin had never been used. The reasons for seeking specialized professional care were severe tooth wear and grinding noises resulting from sleep bruxism, as witnessed by his mother. The clinical examination revealed that the child was in the primary dentition phase and exhibited both the mouth-breathing pattern and severe vertical loss of the upper and lower teeth reaching the region of the dental pulp (Fig. 2).

Moderate dysphagia and involuntary tongue movements were present. The temporomandibular examination revealed no joint noises or muscle/joint pain. An individualized questionnaire assessing sleep, sleep bruxism, daytime bruxism and oral functions was filled out by the speech therapist and mother before and after nine months of treatment.

Both were instructed to provide answers of “yes”, “no”, “improved” and “reduced”, if any variable demonstrated improvement or reduction in intensity following treatment (Table 1).

This study received approval from the Brazilian National Human Research Ethics Committee (CONEP n. 007/2011) and written parental consent was obtained. The subject was enrolled at a special needs institution under the supervision of a special care team composed of a dentist, speech therapist, physiotherapist and occupational therapist for half the day (morning period). The rest of the day, the child remained with his mother. The speech therapist was recruited to the study and stated the child did not exhibit bruxism when at school. The mother also reported that bruxism occurred only during sleep. These reports were collected separately.

Prior to treatment, an attempt was made to perform polysomnography, but the child exhibited abnormal behavior during the preparation, thereby precluding the sleep study. The information given by school professionals and parents regarding the absence of daytime tooth grinding, the information given by mother regarding tooth grinding during sleep and the clinical information, such as severe tooth wear led to the diagnosis of sleep bruxism.



**Figure 2** Loss of dental structure due to severe sleep bruxism.

**Table 1** Values in RMS of electrical activity of masticatory muscles prior to treatment and after 9 months of hyberbola usage.

Muscles	Pre-treatment		After 9 months with HB	
	Rest position	Isometric position	Rest position	Isometric position
RM	537.66	1102.33	106.72	274.24
RT	257.33	414	96.53	224.13
LM	44	189.33	50.94	168.71
LT	7.4	11.23	114.72	228.20

Note: RM: right masseter; RT: right temporallis; LM: left masseter; LT: left temporallis.

The HB was chosen as the treatment option in this case because child did not accept an occlusal splint. The small size was employed (see Fig. 1).

EMG was performed prior to treatment. For such, the volunteer remained seated with the head in a comfortable position. The skin was cleaned with 70% alcohol to reduce bioimpedance and allow the proper placement of surface electrodes, as recommended by the Surface Electromyography for the Non-Invasive Assessment of Muscles (Hermens et al., 1999).

Data on electromyographic activity of the jaw-closing muscles were collected using surface EMG at rest and in the isometric position (maximum contraction). Eight-channel electromyography equipment was used (EMG-800C, EMG System of Brazil Ltda). The signal was submitted to a 20–500 Hz band filter, amplified 1000 times and converted by an A/D plate with a sampling frequency of 2000 Hz per channel and input variation of 5 mV. Four input channels were used to assess the following muscles: anterior portion of the right temporalis (channel 1); surface portion of the right masseter (channel 2); anterior portion of the left temporalis (channel 3); and surface portion of the left masseter (channel 4). Active bipolar, small, passive, circular, disposable Ag/AgC surface electrodes (Meditrace® Kendall-LTP, Chicopee, MA) were used for the evaluation and were placed separated by a distance of 20 mm (De Luca, 1997).

The surface electrodes were placed bilaterally based on anatomic references and guided by the direction of muscle fibers at three points: the anterior portion of the temporalis – 2–3 cm superioposterior to the lateral corner of the eyes in the region of greatest muscle mass parallel to the muscle fibers, but with the sensing surface perpendicularly oriented; the surface portion of the masseter – 1–2 cm above the gonial angle of the mandible in the region of greatest muscle mass, with muscle fibers parallel to the surface; and a reference electrode was positioned on the patient's right wrist to minimize undesirable interference.

The amplitude of the electromyographic signal was analyzed using root mean square (RMS) signal processing expressed in microvolts ( $\mu$ V) to quantify the signal intensity with the mandible at rest and in the isometric position. All procedures used to capture and analyze the electromyographic signals in this study were recommended by the International Society of Electrophysiology and Kinesiology (Solomonow, 1995).



EMG data collection was carried out three times with the mandible at rest and during maximal contraction. The child was verbally instructed to perform maximum voluntary bite force for 3 s, with a 3-min interval between each recording to avoid muscle fatigue. The child then performed a maximum voluntary bite for 10 s for the normalization of the data. The patient and caregiver then received the HB device for the treatment of sleep bruxism. HB therapy was performed for 5 min six times a day with an interval of two to two and a half hours between sessions over a nine-month period. Child was instructed to chewing the HB during the required time, and did it in a slow way, due to his compromised neuromotor condition. Due to the oral disabilities of the child, adaptation was necessary to allow the use of the HB and prevent the child from swallowing the device. For such, a 0.045" stainless steel wire was used to construct an orthodontic arch compatible with the perimeter of the dental arch. Each end of the orthodontic wire was inserted into the long axis of the HB and a U-shaped bend was made at the end of the wire to avoid the displacement of the HB during the mastication exercises (Fig. 3).

Due to quadriplegia, the speech therapist and mother needed to maintain the HB in the child's mouth during the exercises (Fig. 4).

As each device lasts a mean of seven days, the HB was replaced on a weekly basis. The speech therapist helped the child do the exercises when at school and the mother helped him the rest of the day. Both the therapist and mother reported that treatment was performed throughout the study in accordance with the instructions given. The child became ill twice and did not perform the exercises all six times per day during these episodes.

After nine months of treatment, the EMG exams were repeated. The data analysis demonstrated a marked balance in the activity of the jaw-closing muscles (Table 2). Although the muscular balance achieved, child continued to be feeding with semi-solid food, by physician recommendation. The speech therapist stated that the child did not exhibit daytime bruxism and both dysphagia and salivary incontinence had been significantly reduced. Furthermore, the parents reported a significant reduction in tooth grinding during sleep as well as salivary incontinence during waking hours and a considerable improvement in sucking-swallowing movements at meals, which was



**Figure 3** Mastication apparatus (hyperbola) adapted with orthodontic wire for use by child with CP.



**Figure 4** Child doing exercises with hyperbola.

interpreted as an improvement in the child's quality of life (Table 2). No side effects were observed with the use of the HB regarding the dental arch, teeth or temporomandibular joint. According to the inventor of this device, there are no contraindications for this therapy, as the HB is made of a biocompatible material (Cheida, 1997).

## Discussion

Based on the findings of the present study, HB usage led to a visible tendency toward the reorganization of mastication dynamics. The child initially exhibited marked spasticity on the right side, with greater values than those usually seen (Santos et al., 2010) and poor function on the left side. After HB usage, a marked decrease in electromyographic activity occurred in the jaw-closing muscles on the right side and an increase was observed on left side, indicating balanced muscle function.

At the nine-month follow up, electromyographic activity in the muscles of mastication exhibited similar values in the isometric position. In the resting position, the left jaw-closing muscles exhibited an improvement in function, tending toward a balance in the function of the muscles of mastication.

Thus, HB therapy seems to have positively affected the processes of chewing and swallowing, as demonstrated clinically by the reduction in sleep bruxism and salivary

**Table 2** Individualized questionnaire filled out by child's mother and speech therapist to assess quality of life.

Variables	Prior to HB	Post HB
Tooth grinding	Yes	Reduced
Body movement	Yes	Reduced
Snoring	No	No
Nightmares	No	No
Bad mood upon awakening	No	Reduced
Daytime bruxism	No	No
Dysphagia	Yes	Improved
Swallowing movement	Yes	Improved
Sucking movement	Yes	Improved
Salivary incontinence	Yes	Reduced

Note: HB = hyperbola.

incontinence, based on reports by the child's mother and speech therapist. It may be hypothesized that the repetitive movement of the jaw-closing muscles associated with HB therapy caused fatigue, resulting in a decrease in electromyographic activity in the muscles and consequent change in muscle contraction intensity. The findings demonstrate an improvement in oral function associated to a readjustment of muscle tone. The fatigue mechanism may be associated with this cycle of muscle tone readjustment. However, long-term follow up is needed to test this hypothesis.

This phenomenon likely produced hypotonia, bringing comfort to the patient and leading to an improvement in sleep bruxism. The results of the present study diverge from those reported by Santos et al. (2010), who found that the amplitude wave of the jaw-closing muscles in patients with CP was much greater than that found in normal individuals.

In the present study, the patient's initial condition demonstrated accentuated activity of the mastication muscles, which is a rare profile in the literature. In recent studies, the prevalence of sleep bruxism in children with CP is based only on subjective questions and clinical exams, as employed in the present case. No sleep studies have been carried out with this population (Zarowski et al., 2008; Miamoto et al., 2011).

Sleep bruxism often occurs concomitantly to obstructive sleep apnoea (OSA). It is possible that grinding the teeth contributes to opening the upper airways during sleep by stimulating salivation and the swallowing reflex. However, the tendency to clench can draw the mandible into a class 2 position along with the tongue. When this happens, the airway is compromised and the jaw needs to move forward. This back and forth action can lead to bruxism in this subset of patients. A previous study demonstrated a reduction in bruxism following treatment for OSA (Oksenberg and Arons, 2002). In the case cited, the mother reported that child no longer exhibited pauses in breathing at night. However, it was not possible to confirm the absence of respiratory events because the child refused to undergo a sleep study (polysomnography).

Considering the scarcity of studies in the literature, there is a need for further investigations involving different populations with varied demographic characteristics to determine whether there is a pattern to the electromyographic activity in the muscles of mastication in individuals with CP. Studies report that the prevalence of sleep bruxism in CP ranges from 25 to 32.8% (Zarowski et al., 2008; Miamoto et al., 2011). In normal children, occlusal forces from sleep bruxism have the potential to exceed the amplitude of the isometric position during the day, resulting in excessive tooth wear and an increased risk of tooth loss (Nishigawa et al., 2001). Thus, the high RMS values found prior to treatment may explain the severe tooth wear found in the primary dentition of the child in the present case.

Sleep bruxism may also lead to other problems in the primary and permanent dentition and may cause self injury, such as oral lesions (Harris, 2006; Stauffer et al., 2009; Oliveira et al., 2011). Considering the improvement in the function of the muscles of mastication, the authors support the use of the hyperbola device, which may be considered safe and effective in the reduction of both spasms and parafunctional habits as well as the prevention of tooth wear in patients with CP.

The use of an HB device may also reduce the need for invasive treatment, which can be difficult to execute in children with CP. To our knowledge, this is the first paper to address the use of an HB for the treatment of sleep bruxism in a child with CP. The results suggest that this therapeutic approach is safe and useful for patients with similar clinical characteristics. This study highlights the importance of the prevention and control of sleep/diurnal bruxism. The use of a mastication device that does not need to remain in the oral cavity for much time, in contrast to an occlusal splint, seems to be valuable, particularly for patients who do not accept intraoral devices due to involuntary tongue movements. Another advantage over the intraoral devices is the cost-benefit. The HB is commercialized in a package of 10 pieces of chosen size, costs US\$9.00 and may be used for 5 weeks. In this case, the total cost of 9 months (36 weeks) of HB treatment is US\$36.00. This value is much lower than the nightguards and more elaborated splints, which may varies from US\$500.00 to US\$5000.00. This is therapy is accessible to individuals in any socio-economic class.

## Conclusion

The results of the present study demonstrate an improvement in electromyographic activity in the muscles studied and a reduction in reported sleep bruxism following HB usage. However, the direct association between EMG and HB is not yet clear. Moreover, considerable improvement in sucking-swallowing movements at meals was observed following HB usage, but a lack of objective outcome measurement is a limitation of this study. Thus, this therapeutic option may be considered for individuals with cerebral palsy who exhibit sleep bruxism. Its promising effects and low cost allow its use in the public health realm. Further clinical studies with a larger sample size are needed to validate the results and allow the development of a new treatment protocol for patients with spastic cerebral palsy.

## Competing interests

The authors declare no financial competing interests, and any funding for this study was provided by the HB manufacturer.

## Acknowledgments

The Sleep Laboratory receives funding from the Nove de Julho University (UNINOVE) and the EMG Laboratory is supported by State University of Sao Paulo (UNESP). This project was financially supported by the Brazilian Coordination for the Improvement of Higher Education Personnel/Postdoctoral National Program (CAPES/PNPD) – grant number 02495/09-0.

## References

- Cheida, A.P., 1997. Hiperboloide: form and function. *J. Bras. Ortodontia. Ortop. Maxilar.* 11, 49–53.
- De Luca, C.J., 1997. Use of the surface electromyography in biomechanics. *J. Appl. Biomech.* 13, 135–163.

- Dougherty, N.J., 2009. A review of cerebral palsy for the oral health professional. *Dent. Clin. N. Am.* 53, 329–338.
- Fonseca, C.M.S., Santos, M.B.F., Consani, R.L.X., Santos, J.F.F., Marchini, L., 2011. Incidence of sleep bruxism among children in Itanhandu, Brazil. *Sleep Breath* 15, 215–229.
- Harris, D., 2006. Factitious buccal lesion secondary to bruxism in a child with cerebral palsy. *Emerg. Med. J.* 23, e4.
- Hermens, H.J., Frekis, B., Merletti, R., Stegman, D., Blok, J., Rau, G., Klug, G., Hagg, C.D., 1999. SENIAM – European Recommendations for Surface Electromyography. Results of the SENIAM Project. Roessingh Research and Development, Enschede, Netherlands.
- Koman, L.A., Smith, B.P., Shilt, J.S., 2004. Cerebral palsy. *Lancet* 363, 1619–1631.
- Meththanandra, I., Parker, S., Patel, M.P., Braden, M., 2009. The relationship between shore hardness of elastomeric dental materials and Young's modulus. *Dent. Mater.* 25, 956–959.
- Miamoto, C.B., Pereira, L.J., Ramos-Jorge, M.L., Marques, L.S., 2011. Prevalence and predictive factors of sleep bruxism in children with and without cognitive impairment. *Braz. Oral Res.* 25, 439–445.
- Nishigawa, K., Bando, E., Nakano, M., 2001. Quantitative study of bite force during sleep associated bruxism. *J. Oral Rehabil.* 28, 485–491.
- Oliveira, C.A., de Paula, V.A., Portela, M.B., Primo, L.S., Castro, G.F., 2011. Bruxism control in a child with cerebral palsy. *ISRN Dent.* <http://dx.doi.org/10.5402/2011/146915>.
- Ortega, A.O.L., Guimara, A.S., Ciamponi, A.L., Marie, S.K.N., 2007. Frequency of parafunctional oral habits in patients with cerebral palsy. *J. Oral Rehabil.* 34, 323–328.
- Oksenberg, A., Arons, E., 2002. Sleep bruxism related to obstructive sleep apnea: the effect of continuous positive airway pressure. *Sleep Med.* 3, 513–515.
- Peres, A.C., Juliano, Y., Ribeiro, M.O., César, M.F., Santos, R.C., 2007. Occurrence of bruxism in a sample of Brazilian children with cerebral palsy. *Spec. Care Dentist* 27, 73–76.
- Rodrigues dos Santos, M.T., Masiero, D., Novo, N.F., Simionato, M.R., 2003. Oral conditions in children with cerebral palsy. *J. Dent. Child. (Chic)* 70, 40–46.
- Santos, M.B.T.R., Manzano, F.S., Chamliam, T.R., Masiero, D., Jardim, J.R., 2010. Effect of spastic cerebral palsy on jaw-closing muscles during clenching. *Spec. Care Dentist* 30, 163–167.
- Solomonow, M.A., 1995. Practical guide to electromyography International Society of biomechanics. In: Proceedings of the 15th JyVaskyla. International Society of Biomechanics Congress, Finland.
- Stauffer, K., Hamadeh, S., Gesch, D., 2009. Failure of tooth eruption in two patients with cerebral palsy and bruxism – a 10-year follow-up: a case report. *Spec. Care Dentist* 29, 169–174.
- Zarowski, M., Mojs, E., Gajewska, E., Steinborn, B., Samborski, W., 2008. Prevalence of sleep problems in children with cerebral palsy. Preliminary study. *Ann. Acad. Med. Ste-tin.* 54, 59–64.