Self-biting behavior in patients with neonatal brachial plexus palsy

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Abstract
Purpose Self-biting behavior in patients with neonatal brachial plexus palsy (NBPP) has been associated with finger amputation. Our objective is to describe the incidence of this complication, risk factors, and clinical management.
Methods We retrospectively analyzed 612 patients with NBPP. There were 303 males and 309 females. 51.8% of patients had C5-C6 lesions, 28.9% had C5-C7, 18.9% had C5-T1, and 0.3 had C7-T1 involvement.
Results We identified 15 patients with self-biting behavior (2.5%). Ten patients had C5-T1 lesions, and five had C5-C7 lesions. Eight patients were submitted to brachial plexus surgery and seven were not. This behavior appeared between 8 and 46 months of life (mean 23.5), and it was always temporary. There was no difference between operated and non-operated patients (p > 0.05), and no correlation between age at surgery and age of appearance of self-biting behavior (p > 0.05). Physical restriction was effective in treating this complication and we had no case of finger amputation.
Conclusion Self-biting behavior is a rare complication of NBPP, and it is usually associated with severe motor involvement. The behavior duration is limited to a few months. This condition can be effectively treated with physical restriction to prevent hand biting.

Keywords Brachial plexus · Neonatal brachial plexus palsy · Pain · Self-mutilation

Introduction
Neonatal brachial plexus paralysis (NBPP) has an incidence of 0.5–3 per 1,000 live births [1]. Most of the patients have spontaneous recovery, but some may require surgery for functional improvement of the limb [2]. Motor impairment has been the center of attention of most articles dealing with NBPP, since there is a general concept that sensory outcome is good [3–5]. Some studies questioned this concept [6, 7], and sensory involvement in NBPP may be underreported [8].

The self-mutilation due to biting behavior has been rarely described in patients with NBPP [9–15], which may lead to finger amputation and other serious lesions [13–15]. The pathophysiology is poorly understood. This is extremely rare in adults or older children with late onset brachial plexus lesions.

We could identify only two retrospective series dealing with this problem [9, 13]. We conducted a retrospective analysis of our cases to better understand the clinical picture, risk factors, and prognosis of this disturbing complication.

Methods
We retrospectively analyzed 612 children with NBPP referred to the Outpatient Unit of Peripheral Nerve Surgery of Neurosurgery Department of Hospital das Clinicas of University of Sao Paulo from 2000 to 2020. All patients were seen by the same pediatric neurologist specialized in NBPP and clinical data of every visit was digitally recorded using a fixed protocol. The follow-up period varied from 3 to 82 months (mean 28.4). There were 303 males and 309 females. The right side was involved in 375 patients and four patients had bilateral lesions, always asymmetric. For analysis purpose, only the most severe side was considered. Three hundred seventeen patients (51.8%) had pure C5-C6 involvement (Narakas 1),
177 (28.9%) had C5-C7 lesions (Narakas 2), 116 (18.9%) had C5-T1 lesions (Narakas 3 and 4), and two patients (0.3%) had C7-T1 (Klumpke) palsy. One hundred twelve patients were submitted to brachial plexus surgery.

Self-biting behavior was considered present if there were clinical objective hand lesions that were caused by hand biting. Patients with occasional report of hand biting without objective lesions were not considered for analysis purpose.

Statistical analysis was done with Fisher exact test for categorical variables. Comparison of numerical values was done with Mann–Whitney test and correlation between variables was done with Spearman’s rho test. Statistical significance was considered if \( p < 0.05 \).

Results

We identified 15 children with self-biting behavior among 612 patients. This corresponds to 2.5% of the patients, with a 95% confidence interval from 1.5 to 4.0%. There were six males and nine females. The right side was involved in eight patients and the left side in seven. Ten patients had C5-T1 lesions (Narakas groups 3 and 4) and five had C5-C7 lesions (Narakas group 2). The incidence was 2.8% in Narakas group 2 and 8.6% in Narakas groups 3 and 4. There were no patients with pure C5-C6 lesions (Narakas group 1 or Erb’s palsy). Eight patients had brachial plexus surgery and seven did not. Brachial plexus surgery was performed between 3 and 8 months of age. Possible risk factors for self-biting behavior are listed in Table 1.

The self-biting behavior was noted between 8 and 46 months of life (mean 23.5). Only two patients started self-biting after 36 months. Operated patients started self-biting after 26.3 months while non-operated patients started self-biting after 20.3 months. There was no difference between operated and non-operated patients \( (p > 0.05) \). No correlation between age at surgery and age of appearance of self-biting behavior was observed \( (p > 0.05) \).

| Feature                  | Self-biting patients | Other NBPP patients | Statistical significance |
|--------------------------|----------------------|---------------------|-------------------------|
| Sex: female              | 9/15                 | 300/597             | NS                      |
| Affected side: right     | 8/15                 | 367/597             | NS                      |
| Brachial plexus surgery  | 8/15                 | 104/597             | \( p < 0.01 \)          |
| Motor involvement: C5-C6 | 0/15                 | 317/597             | \( p < 0.01 \)          |
| Motor involvement: C5-C7 | 5/15                 | 172/597             | NS                      |
| Motor involvement: C5-T1 | 10/15                | 106/597             | \( p < 0.01 \)          |

\( NS \) not significant, \( NBPP \) neonatal brachial plexus palsy

The finger and hand regions involved are shown in Table 2. The C6 dermatome (thumb with or without index finger) was involved in six cases, C7 dermatome (middle finger, dorsum of hand, or pure index finger lesions) in seven cases, and C8 dermatome (little or ring fingers) in two cases.

There were no cases of finger amputation in our sample. Patients were treated with gloves or splints to prevent biting while the self-mutilating behavior persisted. The lesions were treated with local antibiotics, and in severe cases such as Fig. 1 also with systemic antibiotics. In only three cases, the behavior lasted more than 6 months, and in none it lasted more than a year. No patient required surgical treatment of the lesion.

Discussion

This is probably the largest sample of patients with NBPP dealing with self-biting behavior. It occurred in 2.5% of our patients and could be related to C6, C7, or C8 dermatomes. This occurred only in patients with severe lesions, particularly in those with C5-T1 involvement. There were no cases of pure Erb’s palsy (Narakas 1), despite that they composed more than half of our sample. There was no sex or affected side preference. The incidence of self-biting behavior was lower in our sample than reported by Al-Qattan [9] or McCann [13] (respectively 4.7% and 3.9%). Smaller samples have a greater 95% confidence interval, so these numbers are not significantly different. It is surprising to see so few studies dealing with this problem.

Most of our patients with self-biting injuries were submitted to brachial plexus surgery; however, seven patients were not. Five were late referrals and two patients with C5-C7 lesions recovered biceps function before 6 months of age and were not considered candidates for surgery. There was no difference between operated and non-operated groups. McCann and cols. [13] reported a higher incidence of self-biting behavior in patients submitted to brachial plexus surgery, but also suggested that this could be related to more severe primary injuries. Surgery may not be a real
risk factor for self-biting behavior, but frequently seen in the context of brachial plexus severe lesions that require nerve surgery. Our findings confirmed this hypothesis.

This behavior can have mutilating consequences, such as finger amputation [13–15]. We also have seen severe lesions (see Fig. 1). However, it seems to be a transient problem that can be properly managed with physical restriction preventing the child from hand biting. It usually lasts for a few months and in no case it lasted for more than a year. McCann and cols. [13] reported two patients (out of eleven) with self-biting behavior lasting for more than 24 months. Even so, it was still a transient problem. We had no case of finger amputation.

The pathophysiology of this disturbance is poorly understood. The autotomy in rats with denervated paws is believed to be a possible animal model [16, 17], by there are major differences in frequency, timing, and nature of the behavior. Most animals remove the distal portion of the limb within days. Some believe that this could be related to neuropathic pain [16]. It is difficult to evaluate pain in young children, but none of our patients seemed to show other clinical signs of chronic pain, such as depression, sleep disorders, irritability, poor interaction with other children, or no interest to play. In Al-Qattan’s series [9], none of the patients complained about pain. On the other hand, most of our patients reported some local pain when inquired. This could be due
to the secondary lesion, or to parents induced concern. The lesions were usually referred by the parents as “dodói,” which in Portuguese means “painful.” The transient nature of this behavior is an argument against neuropathic pain. As suggested by McCann and cols. [13], the timeframe of self-biting behavior is compatible with dysesthesia related to sensory reinnervation. Since this is a rare behavior, other factors such as genetic predisposition could play a role.

This is a retrospective series, and the incidence of the problem could be underreported. Patients with short follow-up could have experienced this complication later in life. Most of our patients with short follow-up had mild lesions, and all patients were instructed to get back to our outpatient unit in case of possible late complications. Exclusion of cases with short follow-up would have an impact on incidence that we think would not reflect reality. Even so, a prospective large study would be the best way to obtain an accurate incidence of this condition.

It is important to early recognize this disturbance to avoid serious and irreversible lesions. Simple measures such as physical restriction to prevent hand biting are effective. The transient nature of the behavior can be assured to the parents. Surgical treatment to restore hand sensation [10] in children with self-biting behavior is exceptional and could be only considered after 12 months of continuing biting.

Author contribution COH, RSM, and MGS submitted the project. COH, MZ, and HSN reviewed patient data. COH, MZ, and RSM wrote the main manuscript. All authors reviewed the manuscript.

Data availability All data and material belong to the authors and can be shared by e-mail by request.

Declarations

Competing interests The authors declare no competing interests.

Ethics approval Ethical approval was waived by the local Ethics Committee of Hospital das Clínicas da Faculdade de Medicina da Universidade de São Paulo in view of the retrospective nature of the study and all the procedures being performed were part of the routine care. The study was conducted in accordance with the declaration of Helsinki.

Consent to participate Parents provided consent for the picture publication as long as the identity of the patient remains confident.

Conflict of interest There is no competing interest from any of the authors for this publication.

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