JHS(E)

Journal of Hand Surgery
(European Volume)
0(0) 1–2
journals.sagepub.com/home/jhs
SAGE

Distal sensory nerve transfer for self-mutilation in obstetric brachial plexus palsy: a case report

Dear Editor,

Self-mutilation by excessive mouthing or biting of an upper extremity affected by obstetric brachial palsy (OBP) may result in infection and loss of part of the affected limb (McCann et al., 2004). The incidence of this behaviour is 4 to 5% (Al-Qattan, 1999; McCann et al., 2004). Usually, it is self-limiting or at least decreases in severity over time. The duration averaged 6 months in the series of McCann et al. (2004), but lasted until 4 years of age in one patient and 8 years in another. Treatment is usually non-surgical, including strapping, gloving, psychiatric counselling, wound care, pain-clinic evaluation and neuroprotective medications like pregabalin (Al-Qattan, 1999). Leechavengvongs et al. (2011) reported recovery of sensation and pain relief on the dorsal–radial aspect of the hand in adults with traumatic C5 and C6 root avulsion injuries. They cut the superficial radial nerve distally and sutured the distal stump end-to-side to the median nerve. They suggested that recovery of some sensation in the area with root avulsion relieved the deafferentation pain. We thought that the same principle could be applied to children with OBP in whom sensory deficit and neuropathic pain could be contributing to aggressive mouthing and self-mutilation.

A 3-month-old first-born girl presented with right-sided OBP. Shoulder abduction and wrist extension had spontaneously recovered, but elbow flexion was absent. When she was 9 months, exploration and neurolysis of C5 and C6 roots and the infraclavicular plexus was done. The C5 root was normal. The C6 root was partly fibrotic and formed a neuroma with the distal portion of the C5 root. The suprascapular, phrenic and spinal accessory nerves were functional on electrostimulation. Dense fibrosis around the lower roots precluded their exploration; however, electrical stimulation of the fibrotic mass comprising C7-T1 caused good wrist and finger movements. At the axillary level, the median and ulnar nerves were functional but weak. On stimulation of the radial nerve, elbow and wrist extension were strong, but the finger extension was weak. The child was followed every 6 months and improved in elbow and hand function.

When she was 27 months old, she presented with excessive mouthing of her right little finger. Bite wounds were present over the fingertip and proximal phalanx dorsum with trophic changes and loss of the nail (Figure 1(a)). This behaviour continued despite conservative measures, including wrapping the hand in cloth and applying bitter oil. Sensation for deep pinch was present in the radial and median nerve distributions and absent in the ulnar nerve distribution. No objective sensory measuring of the hand could be done due to the patient's young age. However, the parents had repeatedly observed that the child responded to pinching in the median and radial nerve distribution during sleep, whereas she did not react to pinching in the ulnar nerve territory.

Because of the parents' concerns about biting and repeated episodes of bleeding from the wounds, the patient was treated surgically when she was 33 months old. A palmar longitudinal incision was made overlying Guyon's canal and extended across the wrist to the distal third of the forearm. Using a microscope, the sensory portion of the ulnar nerve was traced proximally to include the dorsal branch and was divided, leaving the motor portion intact. The superficial radial nerve was identified through a dorsoradial incision and divided as distally as possible. The distal portion of the ulnar sensory nerve was transferred subcutaneously and sutured end-to-end to the proximal cut end of the superficial radial nerve with 10-0 sutures. The limb was immobilized in a splint for 3 weeks. The child stopped biting by 6 months, and the bite wounds healed completely. There were no bites in the radial territory. At follow-up, 8 years after nerve transfer, the child perceived touch and temperature over the little finger and had a normal-looking hand (Figure 1(b)).

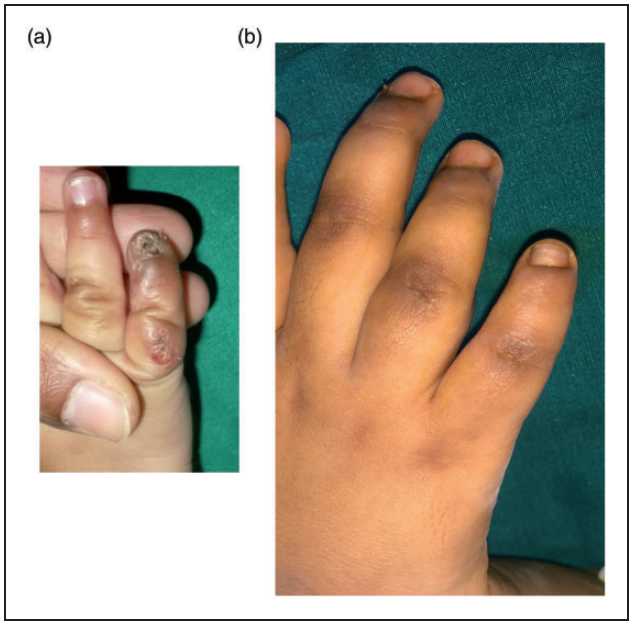

Figure 1. (a) Bite wounds on the dorsum of the little finger's proximal phalanx with loss of the nail at age 27 months. (b) Normal-looking hand 8 years after sensory nerve transfer.

It is uncertain whether this self-mutilating behaviour in OBP represents a response to pain, an exploratory response to an insensate limb or a response to non-painful dysaesthesias (McCann et al., 2004). Rossitch et al. (1992) noted that human self-mutilation following the loss of sensory input into the central nervous system (deafferentation) has features in common with animal autotomy in experimental setups. McCann et al. (2004) found it plausible 'that the self-mutilation behaviour is a manifestation of either a chronic pain syndrome or dysaesthesias'. In their series, the behaviour was more common in children who had undergone surgical procedures on or around the brachial plexus, and the median onset was 8 months after surgery, which they found 'consistent with the time frame expected for dysaesthesias to occur secondary to nerve regeneration'. In our patient, the behaviour started 18 months after the initial neurolysis, which was more than the maximum delay of 11 months reported by McCann et al. (2004). We have no explanation for this delay. Al-Qattan (1999) noted that self-mutilation by biting may be simply a consequence of habitual biting in analgesic areas and may not be motivated by subjective pain, dysaesthesia or stress.

We conclude that peripheral sensory nerve transfer may be an option to restore sensation in digits and to prevent self-mutilating injuries in children with OBP in whom conservative measures fail.

Acknowledgements Dr V Vigneswaran, MS, FNB Hand surgery, for assisting in the patient records collection and artwork.

Declaration of conflicting interests The authors declare no potential conflicts of interest with respect to the research, authorship, and/or publication of this article.

Informed consent Written informed consent was obtained from the parents for patient's anonymized information to be published in this article.

References

Al-Qattan MM. Self-mutilation in children with obstetric brachial plexus palsy. J Hand Surg Br. 1999, 24: 547–9.

Leechavengvongs S, Ngamlamiat K, Malungpaishrope K, Uerpairotkit C, Witoonchart K, Kulkittiya S. End-to-side radial sensory to median nerve transfer to restore sensation and relieve pain in C5 and C6 nerve root avulsion. J Hand Surg Am. 2011, 36: 209–15.

McCann ME, Waters P, Goumnerova LC, Berde C. Self-mutilation in young children following brachial plexus birth injury. Pain. 2004, 110: 123–9.

Rossitch E Jr, Oakes JW, Ovelmen-Levitt J, Nashold BS Jr. Self-mutilation following brachial plexus injury sustained at birth. Pain. 1992, 50: 209–11.

Praveen Bhardwaj[1]*, V. Durga Prasad[2] and S. Raja Sabapathy[1]

[1]The Department of Hand and Microsurgery, Ganga Hospital, Coimbatore, India

[2]The Department of Pediatric Orthopaedics, Ganga Hospital, Coimbatore, India

*Corresponding author: drpb23@gmail.com

doi: 10.1177/17531934211011572 available online at http://jhs.sagepub.com