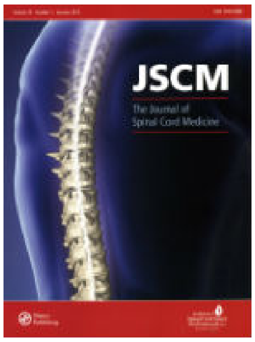

The Journal of Spinal Cord Medicine

ISSN: 1079-0268 (Print) 2045-7723 (Online) Journal homepage: https://www.tandfonline.com/loi/yscm20

Thoracoscopic intercostal to phrenic nerve transfer for diaphragmatic reanimation in a child with tetraplegia

Jacob Latreille, Erika B. Lindholm, Dan A. Zlotolow & Harsh Grewal

To cite this article: Jacob Latreille, Erika B. Lindholm, Dan A. Zlotolow & Harsh Grewal (2019): Thoracoscopic intercostal to phrenic nerve transfer for diaphragmatic reanimation in a child with tetraplegia, The Journal of Spinal Cord Medicine, DOI: 10.1080/10790268.2019.1585706

To link to this article: https://doi.org/10.1080/10790268.2019.1585706

Published online: 18 Mar 2019.

Case Report

Thoracoscopic intercostal to phrenic nerve transfer for diaphragmatic reanimation in a child with tetraplegia

Jacob Latreille[1], Erika B. Lindholm[2], Dan A. Zlotolow[3], Harsh Grewal[2,3]

[1]Drexel University College of Medicine, Philadelphia, Pennsylvania, USA, [2]Division of Pediatric General, Thoracic and Minimally Invasive Surgery, St. Christopher's Hospital for Children, Philadelphia, Pennsylvania, USA, [3]Department of Surgery, Shriners Hospitals for Children, Philadelphia, Pennsylvania, USA

Context: To describe for the first time a novel technique of thoracoscopic intercostal nerve mobilization and intercostal to phrenic nerve transfer in the setting of tetraplegia with the goal of reanimating the diaphragm and decreasing ventilator dependence.

Findings: A 5-year-old female on 24 h ventilator support secondary to traumatic tetraplegia was evaluated for possible phrenic nerve pacing. Left-sided phrenic nerve stimulation did not result in diaphragmatic contraction indicating a lower motor neuron injury. The patient underwent thoracoscopic mobilization of the left phrenic nerve and 10th intercostal nerve while positioned in the left lateral decubitus position using four 5 mm trocars. The mobilized intercostal nerve was transected close to its distal anterior termination and coapted without tension to the cut end of the terminal phrenic nerve using fibrin sealant. Lastly, phrenic nerve pacer leads and battery were implanted in the chest wall and connected to the electrode placed on the intercostal nerve. One year following the procedure, the patient was tolerating phrenic pacing during the day while requiring ventilation overnight. Currently, the patient is 2 years post-operative from this procedure and does not require ventilator support.

Conclusion/clinical relevance: We have shown for the first time a novel approach of thoracoscopic nerve mobilization and phrenic to intercostal nerve transposition to be both safe and effective for restoring innervation of the diaphragm in a child. This minimally invasive procedure is recommended as the preferred approach to reanimate the diaphragm.

Keywords: Phrenic nerve transfer, Nerve pacing, Diaphragmatic reanimation, Traumatic tetraplegia

Introduction

High cervical spine injuries can result in diaphragmatic paralysis, due to either direct damage to the anterior horn cells of the phrenic nerve or damage to the descending white matter tracts that innervate these motor neurons. Patients with diaphragm paralysis often have a shorter lifespan. The associated mortality and morbidity is high in these cases due to respiratory complications which require persistent vigilance to avoid.[1] Treatment that results in successful weaning from the ventilator is known to result in improvement in quality of life with improved mobility, speech, and eating.[2,3]

In upper motor neuron injuries where the phrenic nerve and associated spinal cord motor neurons remain intact, phrenic nerve pacing can be implemented to stimulate contractions of the diaphragm and improve respiratory function. In lower motor neuron injuries, however, direct pacing is not possible because the phrenic nerve distal to the injury has undergone Wallerian degeneration and has lost the ability to conduct an electrical signal. Instead, viable axons from a nerve with intact lower motor neurons need to be transferred to the phrenic nerve to restore the phrenic nerve's conductive properties.[1,4] *Krieger and Krieger* were the first to report that simultaneous nerve transfers with pacemaker placements were a feasible method for treating lower motor injuries via nerve transfer.[2]

Since then, studies have shown numerous ways of restoring respiratory function in mixed upper and

Correspondence to: Erika B. Lindholm, Department of Pediatric Surgery, St. Christopher's Hospital for Children, 160 East Erie Avenue, Philadelphia, PA, USA. Email: erika.lindholm@americanacademic.com
Color versions of one or more of the figures in the article can be found online at www.tandfonline.com/yscm.

DOI 10.1080/10790268.2019.1585706

lower motor neuron injuries. The many options include procedures which incorporate pacing of the intact phrenic nerve with concomitant pacing of intercostal inspiratory muscles, or of donor nerves such as the intercostal or spinal accessory nerve that are transferred to the phrenic nerve for direct reinnervation of the diaphragm.[1,4] Furthermore, in the interest of reducing complications due to the conventional thoracotomy approach and the preference for minimally invasive methods, thoracoscopy has been evaluated in adult populations with unilateral phrenic nerve injuries demonstrating that mobilization of the intercostal and phrenic nerves is possible and perhaps preferred.[5,6] However, there do not appear to be any reports of the entire nerve transfer and pacing procedure being done thoracoscopically.

This case describes for the first time the novel use of thoracoscopic intercostal to phrenic nerve transfer with concomitant phrenic nerve pacing in a tetraplegic ventilator-dependent pediatric patient, demonstrating ventilator independence following reanimation of the diaphragm bilaterally.

Case presentation

The patient involved in this report was a 5-year-old female with a traumatic spinal cord injury who presented for inpatient evaluation of possible phrenic nerve pacing. This injury was the result of a C1 spinal cord injury following being struck by a motor vehicle one year prior, resulting in tetraplegia, neurogenic bowel and bladder, autonomic dysreflexia, and 24 h ventilator dependence secondary to bilateral diaphragmatic paralysis.

Initially, testing was performed to assess phrenic nerve function by using nerve conduction studies (NCS) as well as electromyography in conjunction with ultrasound to monitor contractions of the diaphragm. Bilateral NCS showed absent compound muscle action potentials. Using M-mode ultrasonography, right-sided stimulation at 80 mA showed amplitude changes up to 0.43 cm which was able to be replicated, indicating an upper motor neuron injury. Left-sided stimulation at 80 mA showed no changes, indicating a lower motor neuron injury. A plan was devised to begin phrenic nerve pacing of the right phrenic nerve following placement of a nerve stimulator, while an intercostal nerve with attached electrode would be transferred to the left phrenic nerve.

Surgical repair began on the patient's right side while lying in supine position. A transverse incision was made along the patient's neck and the C5 nerve root in the upper trunk and phrenic nerve were visualized. A pacer (Avery Biomedical Devices, Commack, NY) was secured around the phrenic nerve and sewn into place. Intraoperative testing with a receiver placed in the lower rib cage demonstrated proper functioning with a 1.5 mA threshold. The patient was then turned to a right lateral decubitus position and four 5 mm trocars were placed in the intercostal spaces. Under direct visualization, the left phrenic nerve was dissected off the pericardium (Figure 1). The 10th intercostal nerve was identified as a candidate due to its proximity to the diaphragm and sufficient length to avoid tension. The 10th intercostal was tested using a nerve stimulator and found to respond to a physiologic 0.5 mAmp current with mid-range pulse width using a Checkpoint nerve stimulator (Checkpoint Surgical, Cleveland OH). The intercostal

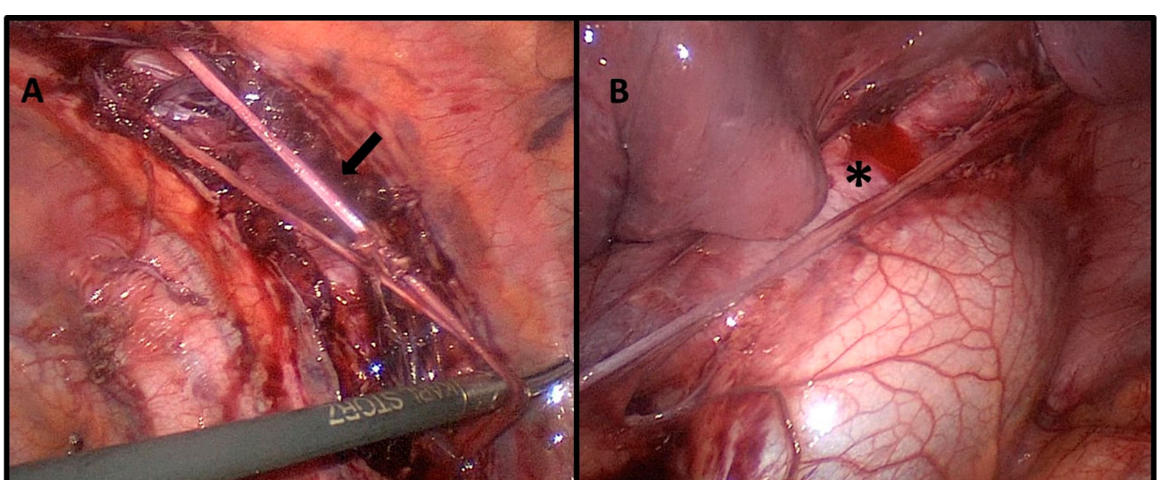

Figure 1. Thoracoscopic mobilization of (A) the 10th intercostal nerve (arrow) off of the chest wall, and (B) the phrenic nerve (asterisk) off of the pericardium.

nerve was then dissected thoracoscopically using the anteromedial portals and a phrenic pacer electrode was placed around the intercostal nerve at the medial posterior position on the chest wall (Figure 1). The intercostal nerve and attached electrode were then brought into the thorax and transferred to the left phrenic nerve. No tension was present and the coaptation was secured with TISSEEL fibrin sealent (Baxter, Deerfield IL) (Figure 2). The receiver was placed in the lateral lower chest wall subcutaneously. The wounds were then closed and the patient was awakened from anesthesia without complication.

Following surgical intervention, pacing was initiated two weeks later and no response from the left side was anticipated until 6 months later. After one year, the patient was tolerating 3.5 mA left and 2.5 mA right pacing throughout the day for up to 14 h with a final tidal volume of 250 ml. By the second year, the patient was tolerating 3.8 mA left and 2.8 mA right pacing for 24 h per day with a final tidal volume of 250 ml and had achieved ventilator independence. M-mode ultrasonography showed marked improvement with diaphragmatic excursion of 2.1 cm on the left and 3 cm on the right. Continued outpatient evaluation will focus on the ability of pacing to meet the anticipated increase in tidal volume required as the patient grows. Since this operation, three additional surgical procedures using thoracoscopic nerve transfer technique have been performed; however there is limited follow up.

Discussion

In the pediatric population, the most common causes of cervical spine injuries is blunt trauma from motor vehicle accidents or by being struck by a motor vehicle while walking.[7] Underdeveloped spinal support muscles and ligaments, incomplete ossification, overall larger head to body size, axial-oriented cervical spine articular processes, hypermobility at the C2–C3 level, and anteriorly wedged vertebral bodies are developmental characteristics of young children who are more prone to upper cervical spine injuries.[7,8] Lesions above the C3 level are amenable to phrenic nerve pacing due to preservation of the motor neurons in the medial aspect of the anterior horn of the spinal cord.[9] Multiple surgical techniques exist for this type of injury, and treatment can involve a cervical, thoracic, or laparoscopic approach for placement of a pacemaker to stimulate the phrenic nerve.[10–12]

The phrenic nerve receives input from the ventral rami of C3, C4, and C5 and descends superficially to the anterior scalene towards the diaphragm where it separates into three branches to provide innervation inferiorly.[2,13] It is the only nerve to supply motor innervation to the diaphragm. Dysfunction of the phrenic nerve therefore leads to pneumonia, sleep disorders, pulmonary effusion, atelectasis, and ventilator dependency.[9] These injuries can often be assessed by NCS and needle electromyography in conjunction with M-mode ultrasound.[14,15] If spinal cord levels C3, C4, or C5 are all within the zone of injury to the spinal cord, pacing the phrenic nerve is impossible due to Wallerian degeneration of the nerve distally and inability to propagate electrical signals.[13] To overcome the loss of axons in the phrenic nerve, multiple nerve transfers have been recommended. The goal is to achieve ventilator independency; an outcome associated

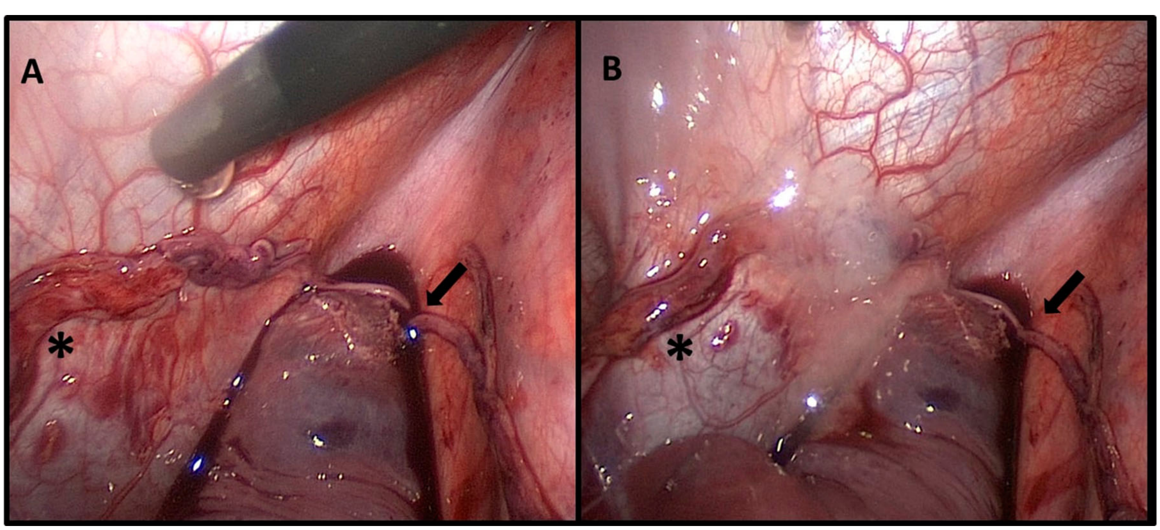

Figure 2. (A) The intercostal nerve (arrow) transferred to the left phrenic nerve (asterisk) without tension and (B) coaptation was secured with TISSEEL fibrin.

with increased mobility, speech, quality of life, and reduced health care costs.[9]

Nerve transfers work by coapting the freshly cut end of an intact nerve with viable lower motor neuron cells to a nerve that has lost axons due to Wallerian degeneration. The axons of the intact nerve grow down the preserved architecture of the recipient nerve and eventually reach the motor endplates of their target muscle. If the donor axons can reach their target before permanent motor endplate demise, the donor nerve will take over the function of the damaged recipient nerve. In cervical spine injuries, the goal of this transfer is to allow the donor nerve to be paced to activate the diaphragm, thus achieving independence from a ventilator.[1] Prognostic factors such as young age and surgical technique are believed to improve the odds of recovery, with a success rate of 75–90% for nerve transfer procedures. The length of time since the injury and whether or not there remains partial activity of the nerve are factors since both determine the viability of the motor end-plates.[1,2,9]

Similar to previous work, our work demonstrates that nerve transfers utilizing the intercostal nerve can be used for restoring diaphragm function and reducing ventilator dependence.[1,2] The technique involved was the first to implement a thoracoscopic approach in the pediatric population. This minimally invasive technique greatly decreases the morbidity of the procedure in comparison to an open thoracotomy approach, and we recommend this as our preferred approach to reanimate the diaphragm.

Disclaimer statements

Contributors None.

Funding There are no sources of financial support for the work reported in this paper.

Conflicts of interest There are no conflicts of interest for any of the authors.

References

1 Kaufman M, Elkwood A, Aboharb F, *et al.* Diaphragmatic reinnervation in ventilator-dependent patients with cervical spinal cord injury and concomitant phrenic nerve lesions using simultaneous nerve transfers and implantable neurostimulators. J Reconstr Microsurg. 2015;31:391–5.
2 L. M. Krieger, A. J. Krieger. The intercostal to phrenic nerve transfer: an effective means of reanimating the diaphragm in patients with high cervical spine injury. Plast Reconstr Surg. 2000 Apr; 105(4): 1255–61.
3 Huttmann SE, Magnet ES, Karagiannidis C, *et al.* Quality of life and life satisfaction are severely impaired in patients with long-term invasive ventilation following ICU treatment and unsuccessful weaning. Ann Intens Care. 2018 March; 8(1): 36.
4 DiMarco AF, Takaoka Y, Kowalski KE. Combined intercostal and diaphragm pacing to provide artificial ventilation in patients with tetraplegia. Arch Phys Med Rehabil. 2005;86:1200–7.
5 Shun Kawashima, Tadasu Kohno, Sakashi Fujimori, Naoya Yokomakura, Takeshi Ikeda, Takashi Harano, *et al.* Phrenic nerve reconstruction in complete video-assisted thoracic surgery. Interact CardioVasc Thorac Surg. 2015; 20(1): 54–9.
6 Lijie T, Zhenglang X. Mobilization of the phrenic nerve in the thoracic cavity by video-assisted thoracic surgery: techniques and initial experience. Surg Endoscopy. 2003;17: 1165.
7 Madura C, Johnston J. Classification and management of pediatric subaxial cervical spine injuries. Neurosurg Clin N Am. 2017;28: 91–1.
8 Leonard JC. Cervical spine injury. Pediatr Clin N Am. 2013;60: 1123–37.
9 Kaufman MR, Elkwood AI, Rose MI, *et al.* Reinnervation of the paralyzed diaphragm: application of nerve surgery techniques following unilateral phrenic nerve injury. Chest. 2011;140:191.
10 Shaul DB, Danielson PD, McComb JG, Keens TG. Thoracoscopic placement of phrenic nerve electrodes for diaphragmatic pacing in children. J Pediatr Surg. 2002; 37:974–8.
11 DiMarco AF, Onders RP, Ignagni A, Kowalski KE, Mortimer JT. Phrenic nerve pacing via intramuscular diaphragm electrodes in tetraplegic subjects. Chest. 2005;127:671–8.
12 Assouad J, Masmoudi H, Gonzalez-Bermejo J, *et al.* Diaphragm pacing after bilateral implantation of intradiaphragmatic phrenic stimulation electrodes through a transmediastinal endoscopic minimally invasive approach: pilot animal data. Eur J Cardio-Thorac Surg. 2012;42:333.
13 Tubbs RS, Pearson B, Loukas M, Shokouhi G, Shoja MM, Oakes WJ. Phrenic nerve neurotization utilizing the spinal accessory nerve: technical note with potential application in patients with high cervical tetraplegia. Child's Nerv Syst. 2008;24:1341–4.
14 Kaufman MR, Elkwood AI, Colicchio AR, *et al.* Functional restoration of diaphragmatic paralysis: an evaluation of phrenic nerve reconstruction. Ann Thorac Surg. 2014;2013;97:260.
15 Epelman M, Navarro OM, Daneman A, Miller SF. M-mode sonography of diaphragmatic motion: description of technique and experience in 278 pediatric patients. Pediatr Rad. 2005;35:661–7.