Case Report

Phrenic Nerve Stimulation for Diaphragmatic Pacing in a Patient with High Cervical Spinal Cord Injury

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Background: Phrenic nerve stimulation is a therapeutic option for patients with central hypoventilation syndrome due to brain stem and high cervical spinal cord dysfunction.

Case Report: A 28-year old woman with high cervical spinal cord injury at the level of C2 had chronic central hypoventilation syndrome, requiring long-term use of a home ventilator. Preoperative end tidal CO\textsubscript{2} and tidal volume during spontaneous breathing indicated hypoventilation syndrome. Bilateral phrenic nerve stimulation for diaphragmatic pacing was performed with spinal cord stimulators used for chronic pain. The end tidal CO\textsubscript{2} pressure (ETCO\textsubscript{2}), tidal volume, and spontaneous breathing time have improved up to 29 months of postoperative follow-up period.

Conclusion: Phrenic nerve stimulation for diaphragmatic pacing can reduce all-time requirement of ventilatory support in patients with high cervical spinal cord injury.

Keywords: Central hypoventilation syndrome, Phrenic nerve stimulation

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Some patients with central hypoventilation syndrome caused by brain stem or high cervical spinal cord lesion require all-time ventilatory support due to insufficiently spontaneous respiration. Phrenic nerve stimulation for diaphragmatic pacing is a therapeutic option to reduce the need of a respirator\textsuperscript{(1-3)}. Because a lack of the specific device of phrenic nerve stimulation for diaphragmatic pacing in Thailand, the authors adapted to use spinal cord stimulator for the operation in a patient with central hypoventilation syndrome caused by high cervical spinal cord injury\textsuperscript{(4,5)}.

Case Report

A 28-year-old woman had a car accident five years ago. She was diagnosed as unstable fracture of the axis with cervical spinal cord injury. An occiput-C1-C2 fusion was performed. She had respiratory insufficiency requiring long-term ventilator use, alternating with spontaneous breathing. Thereafter, the patient could breathe spontaneously and be discharged.

One year later, she developed spontaneous peritonitis caused by small bowel gangrene necessitating surgical resection and end-to-end anastomosis. After the operation, the patient could not breathe spontaneously and required all-time home ventilator probably due to weakness of the incised abdominal muscle, which is a group of accessory muscles for respiration. Subsequently, the authors decided to use phrenic nerve stimulation to restore respiration for the patient. Preoperative spontaneous tidal volume (TV) was less than 50 ml. End tidal CO\textsubscript{2} (ETCO\textsubscript{2}) during 1-minute spontaneous breathing was progressively elevated more than 50 mmHg with symptoms of hypoventilation and hypercarbia.
Since the specific device for phrenic nerve stimulation was not available in Thailand, the patient was operated by using the surgical technique, phrenic nerve stimulation for diaphragmatic pacing with a spinal cord stimulator, innovated by Taira T et al. The cervical and chest incisions (Fig. 1) and 1-cm nerve sheath exposure were demonstrated (Fig. 2). The phrenic nerve was identified with an intraoperative electrical nerve stimulator. Muscle relaxant was not used to observe contractions of the ipsilateral diaphragm and corresponding chest movement. A quadripolar lead-type electrode for deep brain stimulation (DBS) was placed obliquely along the site of exposed nerve and fixed to surrounding connective tissue (Fig. 3). A stimulation device (Itrel-3-Medtronic used for spinal cord stimulator in patient with chronic intractable pain) was implanted into the subcutaneous pocket performed on the anterior chest wall (Fig. 4). The electrode and stimulation device were connected together. The contralateral side was operated in the same manner.

The devices were used in cyclic mode (on 2 seconds, off 3 seconds, soft start 1 second), pulse width 150 μsec, frequency 21 Hz. The amplitude (voltage) was gradually adjusted to achieve the optimal setting (3.3 v on the right side, 3.7 v on the left side). This brought about maximal tidal volume, longer time, and absent adverse effect. The post-operative parameters were improved (ETCO2, 35-45 mmHg, TV 300-350 ml). Postoperative radiographs revealed good position of the stimulation devices and electrodes (Fig. 5). Realtime fluoroscopic study of the chest during turning on the device showed movement of the ipsilateral diaphragm (Fig. 6).

Twenty-nine months postoperatively, she could breathe approximately 45-75 minutes on each side. Total spontaneous breathing time was 5-10 hours per day depending on her demand and daily activity.

Discussion
Phrenic nerve stimulation for diaphragmatic pacing is an alternative option to restore the respiration in patients with central hypoventilation syndrome caused by high cervical spinal cord or brain stem lesions. An animal study of electrophrenic respiration was proposed in 1948. There were many publications and worldwide uses of the operation. Glenn et al used radiofrequency electrophrenic respiration in four patients with central hypoventilation. They reported an effectiveness of one-sided phrenic nerve stimulation for 8-10 hours nightly. They recommended
The presented patient could breathe spontaneously for many hours per day and all-time requirement of the ventilator apparently reduced. Minor adverse effects, including undesired local muscle contraction, spastic movement of the ipsilateral shoulder and upper extremity occurred sometimes. These adverse effects could be corrected by adjusting some parameters of the device setting. Good ipsilateral diaphragmatic movement while turning on the device was demonstrated with real-time fluoroscopy, as purposed by McCauley et al[17]. Of 29 months follow-up period, the device was persistently effective. Many reports also showed long-term effectiveness of the operation[2,5].

Currently, this technique is a new option for patients with central hypoventilation syndrome caused by various etiologies. Postoperative long-term follow-up and adjustment of the device setting are very crucial to achieve the maximal benefit for the patient.

Conclusion

Phrenic nerve stimulation using spinal cord stimulator is effective and can reduce all-time requirement of ventilatory support in patients with central hypoventilation syndrome caused by high cervical spinal cord injury. However, long-term follow-up and adjustment of the stimulator setting are necessary to achieve the maximal benefit.

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References

การผ่าตัดฝังอุปกรณ์กระตุ้นเส้นประสาทฟรีนิคเพื่อช่วยการทำงานของกระปุกลมปอดในผู้ป่วยหนึ่งราย ซึ่งมีการบาดเจ็บของไขสันหลังส่วนคอระดับสูง

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ภูมิหลัง: การผ่าตัดกระตุ้นเส้นประสาทฟรีนิคเป็นทางเลือกหนึ่งในการรักษาสำหรับผู้ป่วยที่มีภาวะหายใจไม่เพียงพอ ซึ่งเกิดจากระบบประสาทตอนกลางอันมีสาเหตุมาจากความมีดีปกติของความสามารถหนึ่งหรือไขสันหลังส่วนคอระดับสูง

รายงานผู้ป่วย: ผู้ป่วยหญิงอายุ 28 ปีมีไขสันหลังบาดเจ็บที่ระดับคอส่วนบนส่วนกลางที่ 2 มีภาวะหายใจไม่เพียงพอเรื้อรังซึ่งมีอาการหายใจต้องการใช้เครื่องช่วยหายใจตลอดเวลาในขณะที่ผู้ป่วยต้องการหายใจเพียงพอ ผู้ป่วยได้รับการผ่าตัดฝังอุปกรณ์กระตุ้นเส้นประสาทฟรีนิคเพื่อช่วยการทำงานของกระปุกลมปอดในขณะที่ผู้ป่วยหายใจตนเอง อาการหายใจต่าง ๆ ดีขึ้นอย่างมากในช่วงติดตามผลการรักษาหลังผ่าตัด

สรุป: การผ่าตัดกระตุ้นเส้นประสาทฟรีนิคเพื่อช่วยการทำงานของกระปุกลมปอดสามารถลดความสามารถในการหายใจของผู้ป่วยได้