Complex Regional Pain Syndrome Following the Nuss Procedure for Severe Pectus Excavatum

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Complex regional pain syndrome (CRPS) is not an uncommon complication after surgery, but has never been reported after the Nuss procedure for repairing pectus excavatum. A 22-year-old man with pectus excavatum had type I CRPS that developed 2 weeks after the Nuss procedure. He complained of persistent pain, hyperalgesia, weakness, edema, and color and temperature changes on right upper extremity. Following intensive rehabilitation, the degree of pain, weakness and edema were ameliorated. He recovered 6 months after surgery and the pectus bars were removed uneventfully 3 years after the repair.

Keywords: complex regional pain syndrome, pectus excavatum, Nuss procedure

Introduction

Pectus excavatum is the most frequent congenital chest wall deformity, mostly presenting in the neonate and might be sunken at puberty. In 1998, Nuss and associates documented a minimally invasive method (the Nuss procedure) for correcting pectus excavatum with good results.1) However, several complications after this procedure have been reported, including bar flipping, pneumothorax, hemothorax, pleural or pericardial effusions and cardiac injuries.2) Neurologic complications have been reported only rarely.3,4) We described an adult patient who had type I complex regional pain syndrome (CRPS I) that developed after the Nuss procedure.

Case Report

A 22-year-old man had suffered a chest wall deformity since his childhood. The deformity was made more pronounced by puberty and he complained of progressive chest tightness, exertional dyspnea and palpitation. He had a severe pectus excavatum confirmed from the caved-in appearance of his anterior chest wall and a computerized tomographic scan of the chest in which the Haller index5) was 5.8 (Fig. 1).

A Nuss procedure was performed by Dr. Chen. The patient was placed in the supine position after inducing general anesthesia. The arms were kept abducted at 70° to the body axis. One small vertical skin incision was made in the midaxillary line on each side. After subcutaneous or submuscular dissections had been performed, the pleural cavities were entered at the hinge point (highest point of the funnel). A right thoracoscopy was done first using a 5-mm, 0° scope entering the pleural cavity via the right surgical wound for direct inspection of the mediastinal structures. A left thoracoscopy was performed later via the left surgical wound. A right-to-left mediastinal dissection was done via the introducer under direct left thoracoscopic visualization. After a substernal tunnel had been achieved, a 28 Fr. chest tube was connected to the introducer and retained in the thorax after...
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The introducer had been retracted. A pre-bent Lorenz pectus bar (Biomet Microfixation Inc., Jacksonville, Florida, USA) was connected to the chest tube and advanced across the mediastinum. After the pectus bar had been rotated and anchored into position, an additional substernal tunnel was made using direct left thoracoscopic viewing, and another pectus bar was introduced obliquely for correcting the long and pronounced deformity. These bars were fixed with a 1.0-mm stainless wire sutures at the ends of the bar to prevent displacement (Fig. 2). The patient recovered well after surgery and was discharged 6 days later.

However, hyperalgesia, edema, and color and temperature changes of the patient's right hand and forearm developed 2 weeks after the operation. Weakness of adduction of his right fingers with a muscle power grade of only 3–4/5 was also observed. Electromyography and nerve conductive velocity studies showed unremarkable findings. The blood profile including erythrocyte sedimentation rate, C-reactive protein level, complete blood counts and serum autoantibody titers were within normal ranges. Plain X-ray films of the cervical spine and the right upper limbs also showed no abnormal findings. A three-phase bone scan revealed moderately increased uptake in the right elbow joint in the bone phase and slightly increased tracer distribution in the right elbow region during the arterial and blood-pooling phase (Fig. 3). These findings were thought to be consistent with CRPS. The patient undertook a rehabilitation program including contrast baths (alternating heat and cold), and transcutaneous electrical nerve stimulation. Using a gentle range of motion, isometric strengthening exercises were also incorporated gradually into the treatment. The neurologic deficits subsided completely 6 months after surgery. The pectus bars were removed 3 years after the operation and the patient recovered uneventfully. The results of the corrective surgery were maintained after bar removal and the patient was satisfied with his appearance.

Discussion

In 1988, Nuss and colleagues developed a minimally invasive repair for pectus excavatum. The procedure introduced a curved, stainless steel bar behind the sternum for correcting the depressed chest wall without resection of the costal cartilages. Although the procedure was accepted nearly universally, the complications and learning curve for surgeons were topics of discussion. Neurologic complications have been reported rarely. In 2005, Fox, et al. demonstrated that patients were exposed to the risk of a transient brachial plexus injury during the Nuss procedure. In 2011, Lee, et al. reported a rare case of a complicated thoracic outlet syndrome after a modified Nuss procedure. Nevertheless, CRPS following the Nuss procedure for correction of pectus excavatum has not been reported.

CRPS is a neurological disorder involving the limbs, with patients presenting with disabling pain, edema, or vasomotor or sudomotor abnormality. The painful conditions are persistent and disproportionate in time or degree.
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The symptoms usually appear after an initiating noxious event such as surgery, ischemia, or nerve compression. The Consensus Conference of the International Association for the Study of Pain has subdivided CRPS into type I (formerly Reflex Sympathetic Dystrophy) and type II (formerly causalgia). Both types share some clinical symptoms, with the only distinguishing feature being the evidence of peripheral nerve injury in type II.7)

The development of CRPS is not an uncommon complication after surgery and most cases have occurred after orthopedic procedures, usually with the site of surgery being involved.8) How CRPS arises after surgery is still unclear, as no single mechanism can explain the heterogeneity of the symptoms.8) Mechanisms including central and peripheral sensitization and alterations in cutaneous innervation can alter the functions of the sympathetic nervous system and contribute to CRPS.7)

CRPS is a clinical diagnosis based on the patient’s symptoms and signs.7,8) Three-phase bone scanning is a standard imaging method used to help in making the diagnosis. The characteristic pattern is of increased tracer uptake in the affected extremity compared with the normal extremity in the first few months. Although the outcomes for patients with CRPS are very difficult to predict, an early diagnosis and treatment increase the likelihood of a successful resolution.7,8) Mild cases respond to physical therapy, whereas moderate cases may require adjuvant analgesics and/or antidepressant medications. Patients with severe pain and/or sympathetic dysfunction require regional anesthetic blockade to enable them to participate in physical therapy. A small percentage of patients with refractory, chronic pain will require long-term multidisciplinary treatment, including physical therapy, psychological support and pain-relieving measures.7)

Our patient had type I CRPS that developed 2 weeks after surgery. After rehabilitation, his neurologic deficits recovered without any sequelae. The pectus bars were removed 3 years after repair uneventfully. The patient was satisfied with the ultimate cosmetic results.

Conclusion

CRPS occurring after the Nuss procedure is very rare but possible. Early diagnosis and rehabilitation for such patients clearly can give good results.

Disclosure Statement

There is no financial or other interest in the manufacture or distribution of the device.

References


