Clinical Case Report

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Thoracic outlet syndrome (TOS)

A case report of a rare complication after Nuss procedure for pectus excavatum

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Abstract

Rationale: The Nuss procedure has become a major alternative operation for patients with pectus excavatum (PE).

Patient concerns: We report a case of 27-year-old man with PE who developed thoracic outlet syndrome (TOS) after the Nuss procedure. The patient showed clinical symptoms of brachial plexus compression.

Diagnoses: Further evaluation demonstrated a narrowed space between the first rib and the anterior scalene muscle and compressing the brachial plexus and vessels.

Interventions: Nerve nourishing medicine and rehabilitation exercising were taken to restore the muscle strength.

Outcomes: Several months later, the clinical symptoms disappeared.

Lessons: Medicine and rehabilitation exercising may benefit the functional recovery of impaired nerve in TOS in the early stage of TOS.

Abbreviations: CT = computer tomograph, EMG = electromyogram, PE = pectus excavatum, TOS = thoracic outlet syndrome.

Keywords: Nuss procedure, pectus excavatum, thoracic outlet syndrome

1. Introduction

Pectus excavatum (PE) is a congenital chest deformity in which several ribs and the sternum develop abnormally to produce a concave or caved-in appearance of the anterior chest. It has known as the most common chest malformation and is more common in men. Rivatch operation had been the common surgery for PE before Nuss introduced a minimally invasive technique to correct PE in 1987.[1,2] He placed a steel bar in the desired chest shape under the sternum by video-assisted thoracoscopic surgery. The bar was removed after a minimum of 2 years. Because this approach has shown excellent functional and cosmetic results and so it has been accepted by more and more patients. However, several common complications such as heart perforation, pneumothorax and pleural effusion, arteriovenous fistula, have been reported after this minimally invasive operation.[2] Herein, we describe a case of 27-year-old man who presented thoracic outlet syndrome (TOS) after Nuss procedure.

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The patient had provided informed consent and the informed consent was obtained from the patient for publication of this case report and accompanying images.

The authors have no conflicts of interest to disclose.

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2. Case presentation

A27-year-old male with pectus excavatum, who had no hypoesthesia and weakness in both upper limbs, had the Nuss procedure performed in the Department of Thoracic Surgery, PLA Army General Hospital, Beijing. The preoperative Haller index was 4.7 measured on thoracic CT film. The patient signed informed consent and gave his permission to perform the Nuss procedure.

After general anaesthesia, patients were placed in a supine position with both arms abducted and the chest elevated. A correction bar of appropriate length and shape was selected according to the distance between both mid-axillary lines on the surface of the thorax. The transverse lines were drawn from the deepest point of the sternum to mid-axillary lines at both sides. Two skin incisions of 3 cm length were made along the intersections of the transverse and mid-axillary lines. The introducer was slowly inserted from the right incision, advanced across the mediastinum and toward the left incision, and raised the sternum and the anterior chest wall to the desired position. Then the correction bar was introduced to its proper position and turned over so that the convexity faced anteriorly. A stainless-steel stabilizer was placed on both sides of the metal bar. The postoperative chest X-ray was normal, and Haller index was 2.0 on CT film. The metal bar discharged on the 7th day (Figs. 1 and 2). The patient was satisfied with the cosmetic appearance. One month later, the patient had pain in both upper limbs and need somed onto reduce his pain. After additional two month, the patient presented with weakness, numbness as well as pain in both arms. The signs seemed to be caused by compressions of the brachial plexus, subclavian arteries and veins as they exited the chest. The radial pulses were still touchable with abduction maneuver.

We performed electromyogram (EMG) and CT arteriogram of subclavian arteries on patient to support the diagnosis. The CT arteriogram of subclavian artery demonstrated that the subclavian artery was partially compressed during right arm abduction and external rotation between the clavicle and the first rib (Figs. 3

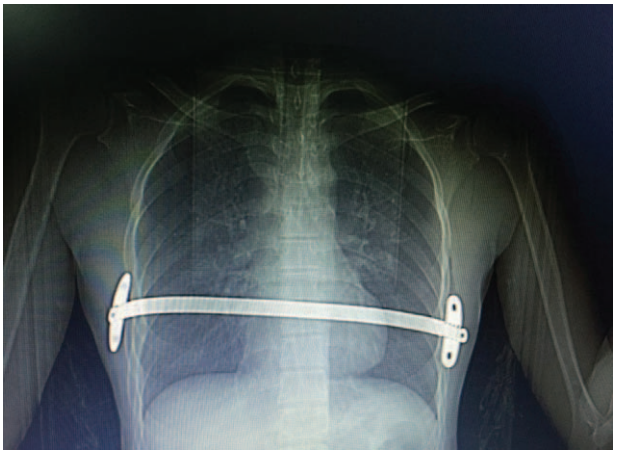

Figure 1. A steel bar was inserted through the 5th intercoastal muscles.

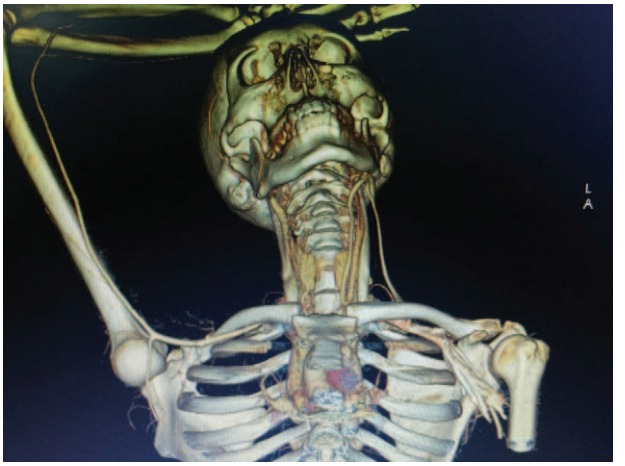

Figure 3. CT scanning showed that the narrowed subclavian space between the right first rib and clavicle. CT = computed tomography.

and 4). EMG showed that the brachial plexus was injured at the position of thoracic outlet. We recognized the narrowed space between the first rib and the anterior scalene muscle after the correction of chest deformity. The force from the metal bar elevated the first ribs. In order to treat this complication, we first advised the patient to remove the metal bar. The patient rejected our advice due to the cosmetic reason. We then gave patient nonsteroidal anti-inflammatory drugs to alleviate pain. Vitamin 12 and vitamin B1 were given to nourish the brachial plexus. Meanwhile, the patient was encouraged to do (arm/upper limb) exercises to restore the muscle strength. One month later, his muscle strength was restored to level 4. Hypoesthesia and pain of upper arms were alleviated. He did well and had no complaint during the following 1 year.

3. Discussion

Nuss introduced a minimally invasive surgical procedure to correct the PE in 1987.[1]

This surgical procedure has practically improved functional as well as cosmetic consequences of the PE patients. Several complications associated with the procedure has been reported, including cardiac injury, bar displacement, bar rejection reaction, hemothorax, and so on.[2,3]

TOS causes pain of upper limbs, shoulder, and neck, compression of the nerves and blood vessels, by which hands may show weak, pale, bluish, and even become sensitive to cold, swollen and tired easily.[4,5]

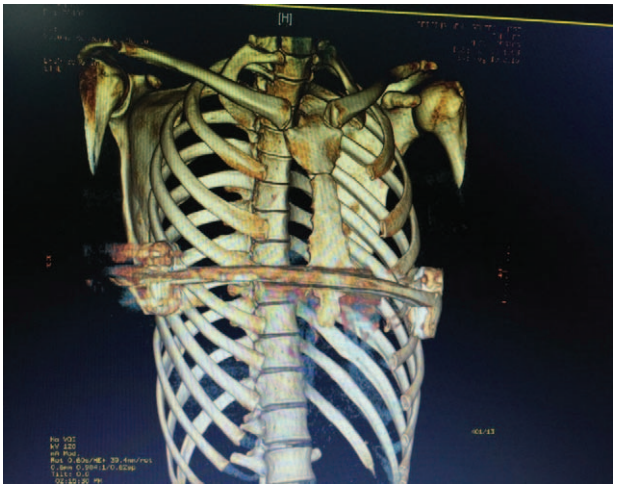

Figure 2. 3D reconstruction imaging by CT showed a better configuration after Nuss surgery. CT = computed tomography.

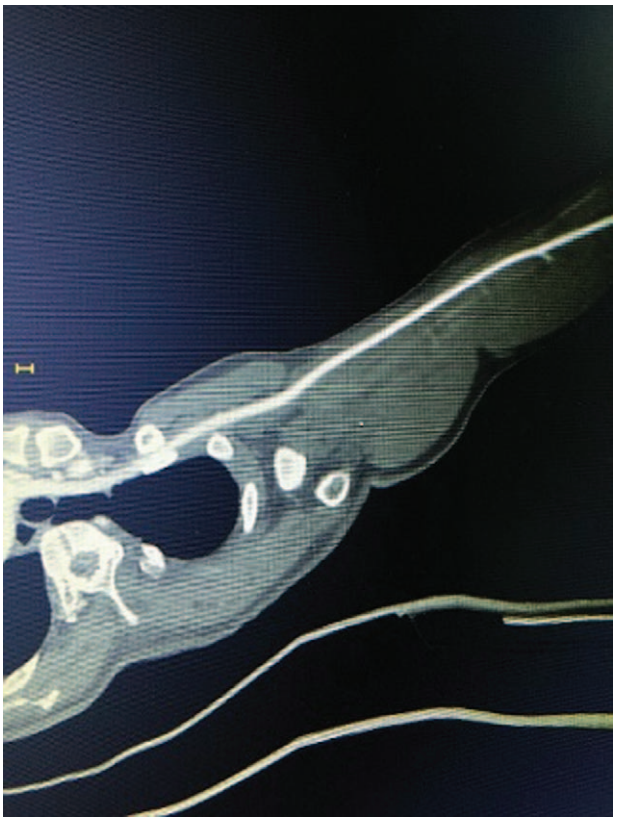

Figure 4. CT arteriogram showed the subclavian artery was partly compressed with right arm abduction and external rotation. CT = computed tomography.

Donders and Geelen in 1988 reported the first case developed TOS after corrective surgery for PE Later on, about 90 patients developed TOS after the Nuss operation have been reported.

Lee et al[6] reported a TOS case developed after the Nuss procedure. The patient showed clinical signs of brachial plexus compression postoperatively on day 3. The claw-hand deformity was developed and not improved even after the metal bar was removed for 2 months.

Lee et al thought that TOS was caused by insertion of the bar, by which the mechanical forces created the compression within the interscalene triangle among the anterior and middle scalene muscles and the first rib. In the Nuss procedure, the deformed costal cartilages and sternum were lifted by the metal bar without any resection like Rivatch operation in which elevating the deformed costal cartilages and sternum was proportional to the depth of the chest deformity.[7] Since the more the chest malformation is in need of more force to reform it, change in the anterior chest might be harmful to the brachial plexus by altering the adjacent space. They thought that the depth of the chest deformity could be a factor for TOS after PE operation.

BurcuKilic[8] report a 22-year-old man with deep pectus excavatum who developed vascular TOS after the Nuss procedure. The patient showed clinical features of subclavian artery compression, including pain, weak and cold in right arm. There was no right-sided radial pulse in the costoclavicular position and with the hyperabduction maneuver. Further evaluation demonstrated that the first rib pressed on the right subclavian artery without any nerve-compressive symptoms. The first rib resection and the anterior scalene muscle and fibrous bands relieving were performed instead of removing the metal bar.

Donders and Geelen[9] reported 2 cases who developed TOS after the Ravitch's operation for correction of PE. Although removal of deformed costal cartilages was not necessary and the force lifting the sternum was smaller in the Ravitch's procedure, TOS still developed. We speculate those patients with TOS after PE correction may have symptomless anatomical abnormalities in the interscalene triangle. The correction surgery deteriorated the anatomical anomaly. Furthermore, the patients were usually asked to hold a position of shoulder hyperabduction to consolidate the postsurgical cosmetic results. Sustained posture might change the anatomical structure of the interscalene triangle.[10] Reducing the space between first rib and scalene muscle compressed brachial plexus and vessels. Preoperative anatomical abnormalities of interscalene triangle could be considered as a critical factor for TOS after PE operation.

Redman thought PE patients with neurogenic TOS usually had identifiable anatomical anomalies, including soft-tissue anomaly (58%), bone anomaly (27%), and other abnormalities (3%).[11]

In our report, TOS occurred in a case reformed front chest with one steel bar, Haller index 4.6. TOS cases reported by Kilic et al and Lee et al were inserted 2 steel bars, Haller index 4.7. TOS could appear postoperatively in both Nuss procedure and Ravitch procedure. As known, most PE patients did not develop TOS after the operation, no matter the severity of malformation of chest. It appears that the chest malformation is not a significant factor to be concerned for TOS after PE correction. Nevertheless, some anatomical defects of interscalene triangle could be the underlying mechanism for TOS. The lifting force from steel bar deteriorates the narrowness of interscalene triangle to cause TOS. We suggest that the anatomical abnormalities of interscalene triangle should be evaluated preoperatively to prevent TOS occurrence from PE operation.

We recommend that prior to correct PE, all patients should be examined to detect latent TOS due to neurovascular compression. This complication after the Nuss procedure should be actively treated to avoid damage of the cervical-brachial plexus. We prevented one case from permanent impairment by nourishing nerve to ameliorate the function of impaired nerve plexus. The resection of first rib could be an alternative to treat the TOS if the patient is reluctant to remove the steel bar.

Author contributions

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Methodology: Jingbo Ma.
Resources: Jingbo Ma.
Supervision: Jing Zhao.
Writing – original draft: Weiqiang Zhang.

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