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CASE REPORT

PECTUS CARINATUM DEVELOPMENT AFTER THE NUSS PROCEDURE: A CASE REPORT

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ABSTRACT

Development of pectus carinatum is a very rare complication of the Nuss procedure. This complication may lead to early bar removal, which hinders sternal protrusion but induces the recurrence of pectus excavatum. We report a case of pectus carinatum development following a Nuss procedure and pectus excavatum recurrence after bar removal to discuss what could have been done better from today's perspective.

Keywords: Complications, funnel chest, minimally invasive surgery, pectus carinatum

INTRODUCTION

Pectus deformities include excavatum, carinatum, mixed-types and arcuatum. Those deformities are characterized by protrusion or depression and with or without rotation of the sternum due to the deformities of the costal cartilages or the sternum itself (1).

Pectus excavatum (PE), also called funnel chest, is accepted as the most common pectus deformity and characterized by sternal depression. Deformity may be congenital, up to 0.8% of newborns with PE deformity are noted but this ratio may be underreported (2). However, it should be considered that this deformity is not purely congenital. The majority of patients present with sudden depression of the sternum during the growth period or complain about the worsening of mild depression to severe depression (3).

Pectus carinatum (PC) is characterized by protrusion of the sternum and accepted as the second most common pectus deformity, whereas a study found that PC (0.86%) was more prevalent than PE (0.54%) in Turkish children (4).

Open surgery and reconstruction of the chest wall were mainstays for years, but the introduction of minimally invasive repair of PE, the Nuss procedure, has changed the era. The Nuss procedure uses pectus bar(s) inserted into the thorax to support the, thus correcting the deformity. Those bars are withdrawn after 2-3 years (5). Minimally invasive repair of PC, the Abramson procedure, has been developed upon this idea after all (6). Orthosis and vacuum bell treatments are new and effective treatment options for selected patients (7, 8). Surgery decisions are made jointly with patients' concerns and clinical judgments. While mostly body image concerns due to the appearance of their chest and psychosocial anxiety are motivating factors, in severe cases cardiac compression, mitral valve prolapse, and pulmonary function impairment may warrant the procedure as well (9).

Most of the complications related to the Nuss procedure, such as pneumothorax, pneumonia, and bar displacements, are well managed without causing serious comorbidities, but fatal cases due to cardiac perforation and lung injury were also reported in the literature (10-12). As bars are placed posterior to the sternum and anterior to the pericardium, excellent technique and maximum attention are required intraoperatively (13).

Development of PC, which is at the other side of the pectus spectrum compared to PE, after a Nuss procedure is very rarely reported in the literature. This case report presents an eight-year-old patient with PC development after a Nuss procedure and the recurrence of PE following bar removal.

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CASE REPORT

An eight-year-old female patient was brought to our clinic with a funnel chest. Our detailed physical examination and anamnesis revealed symmetrical and severe PE (Figure 1A). No accompanying diseases, family history, or previous surgeries were noted. The patient underwent a Nuss procedure with thoracoscopy in June 2011. A 220 mm-long bar was implanted in the patient using one stabilizer on the left end. No steel wires were used. The operation was performed successfully in 90 minutes without any perioperative complications (Figure 1B).

Excellent correction of the deformity was seen in the early postoperative period. No bulging area was noted within the postoperative three months. A significant sternal bump was noted in the postoperative sixth month. The patient was invited to the clinic in the postoperative seventh month for further evaluation of the chest. PC was noted in the seventh month, and bar removal surgery was planned (Figure 1C).

The bar removal was performed in the seventh month, which caused an indentation of the chest after 15 days. Ten months after bar removal (postoperative 17th month), significant PE was noted. The postoperative 36th month examination revealed worsened PE. The patient was seen and re-evaluated in the 55th and 120th months regarding PE (Figure 1D). The X-ray examination revealed the deformity of the chest during the same periods (Figure 2A-D).

DISCUSSION

Hereby, we have reported a case of a PE patient who had a very flexible sternum that was bent by the Nuss bars to a degree that is enough to cause PC. This flexibility would have likely induced the recurrence of PE after bar removal.

This clinical case is not similar to what Swanson and Colombani. (14) noted, where the development of PC was attributed to fibroelastic genetic disorders, as fibroelastic deficiencies were not noted in our patient. As Paya et al. (15) postulated in 2003, bar removal due to PC development hindered sternal protrusion, but early removal itself induced the recurrence of PE. Zhou et al. (16) reported one case of PC development after the Nuss procedure, and they suggested the patient use chest strap fixation, which is a kind of carinatum bracing, and did not remove the bar.

Donald Nuss found in his series that approximately 0.3% of patients developed PC after the Nuss procedure, and he suggested using carinatum bracing (5, 17).

A high-volume study found 0.8% overcorrection in their Nuss series, where most of those patients underwent premature bar removal just as our patient did (18).

We have reasoned that the patient's condition could have been managed better if it had been investigated from today's perspective.

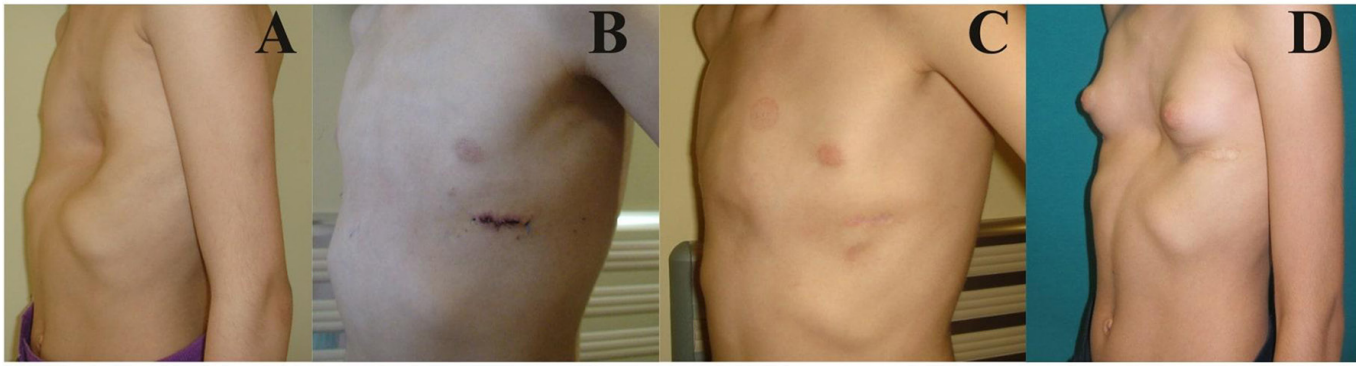

Figure 1: The patient's chest before the Nuss procedure (A). The patient's chest after the Nuss procedure (B). The patient's chest seven months after the Nuss procedure, before bar removal due to the development of pectus carinatum (C). The patient's chest four years after bar removal (55th month) due to the development of pectus carinatum (D).

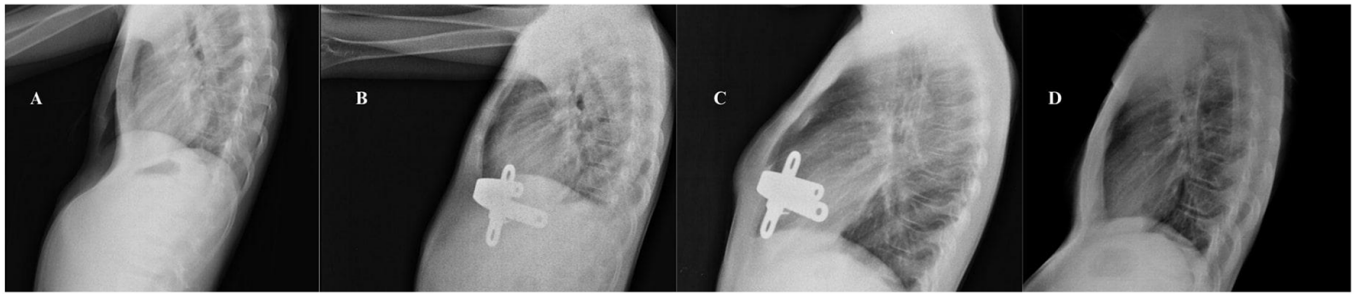

Figure 2: The patient's lateral chest X-ray before the Nuss procedure (A). The patient's lateral chest X-ray after the Nuss procedure (B). The patient's lateral chest X-ray seven months after the Nuss procedure, before bar removal due to the development of pectus carinatum (C). The patient's lateral chest X-ray four years after bar removal (55th month) due to the development of pectus carinatum (D).

In those times, Nuss bars were mostly retained for two years in our clinical practice, and this patient developed overcorrection at seven months (5). It was hypothesized that overcorrection would reverse, and the patient would be both excavatum and carinatum free after bar removal, but the patient unfortunately ended up having a recurrence of PE. Recently, pectus bars were retained up to three years. Considering that, a revision procedure with repositioning and less bending would have been a sensible option for this case.

Non-invasive techniques such as vacuum therapy could have been tried before the Nuss procedure, and success would have been likely when the patient's flexibility and age were taken into account (8, 19). Another point to consider is whether sternal protrusion could have been managed with external bracing orthoses like Donald Nuss suggested and Zhou et al. (16) tried (5). The patient could be given an external bracing orthosis while bars are still in situ if it was tolerable. The procedure would have also prevented the recurrence of PE, as early bar removal would not have been performed. If the patient could not tolerate bars and external bracing concomitantly, waiting up to two years and performing bar removal and then trying to control PC would have been another option.

Given both the rarity and reporting of this complication, there is no consensus on the management, and even guidelines do not mention this complication (1). Our clinical experience and output from this case were to offer vacuum bell therapy before the Nuss procedure for cooperative, willing, and flexible patients, where we measure the flexibility of the chest wall with a vacuum bell in the first examination. However, as the Nuss procedure is being performed frequently, it is important to be aware of this rare complication, and a careful revision procedure or orthotic support may be considered. The development of PC after a Nuss procedure is a very rare but possible complication that can be observed in flexible patients. Early bar removal due to this complication may lead to the recurrence of PE. Revision procedures, vacuums and external bracing orthoses may be used to manage this complication.

Ethics

Informed Consent: *An informed oral consent was obtained from the patient's legal guardians.*

Footnotes

Conflict of Interest: *The authors declared no conflict of interest.*

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