

NICKEL ALLERGY AFTER A NEGATIVE TEST RESULT FOLLOWING NUSS PROCEDURE FOR PECTUS EXCAVATUM: A CASE REPORT

José Fontoura-Matias¹, Joana Pereira-Nunes¹, Sofia Vasconcelos-Castro², Nuno Gomes³, Ana Paula Cunha^{3,4}, Miguel Soares-Oliveira²

¹ Department of Pediatrics, Centro Hospitalar Universitário São João, Porto, Portugal

² Department of Pediatric Surgery, Centro Hospitalar Universitário São João, Porto, Portugal

³ Department of Dermatology, Centro Hospitalar Universitário São João, Porto, Portugal

⁴ Department of Medicine, Faculty of Medicine, Universidade do Porto, Porto, Portugal

* Corresponding author: zpfmatias@gmail.com

CASE REPORT

The Nuss procedure is the most common approach to pectus excavatum repair. It is a minimally invasive technique in which a steel bar is placed posteriorly to the sternum to correct its position and correct the excavatum deformity.¹

The standard practice in most of the centers is to perform a preoperative skin testing to exclude metal allergies only in patients with personal or familial known metal allergy, with around 1 percent of people developing allergy to the steel bar after its insertion, with potential serious consequences.²

Due to recent data showing that 6.4% of patients present clinical or patch test evidence of metal allergy prior to the surgery³, it is our department's protocol to test all patients before the Nuss procedure and those with positive allergy receive a titanium bar instead of a stainless steel one.

We present a rare case of development of metal allergy after Nuss procedure, despite preoperative negative skin test and alert to this potential complication.

We present the case of a 15-year-old male who was referred to the pediatric surgery clinic due to severe pectus excavatum that caused exercise intolerance (Haller index of 6.6). After clinical and radiological evaluation, surgery was recommended.

He denied any atopy history, including asthma, rhinitis and conjunctivitis, and any known allergy or contact dermatitis. He had no other relevant medical history.

Before surgery, he performed a metal allergy patch-testing with baseline series from Portuguese Contact Dermatitis Group and with metal series, as according to international standards. The allergens were applied under occlusion on the

patient's dorsum, under standardized conditions. IQ Ultra™ Patch Test Units (Chemotechnique Diagnostics) and Mefix™ adhesive tape were used. The test was read on the 3rd and 7th days and interpreted to be all negative.

The Nuss procedure was performed using one stainless steel bar with a left-sided stabilizer (Figure 1). The bar was secured to the chest wall with nonabsorbable sutures. The patient presented a bilateral pneumothorax on the day after surgery, and a chest tube was placed on the right (large volume pneumothorax, without mediastinum deviation). The patient was discharged on the seventh postoperative day, after a negative provocative clamping test and chest tube removal, with good pain control under oral medication. One month after surgery, the patient reported no complaints.

Several weeks later however, he presented occasional redness and pruritus in both lateral skin incisions and over the anterior-lateral subcutaneous bar's location. He was again observed in a Dermatology appointment and a metal allergy was suspected. He repeated the patch-testing and, this time, presented a positive result for nickel. As his complaints were sporadic and mild, he was medicated with antihistamines when needed, and it was decided that the bar would not be removed before the recommended 3-year period time. The patient's situation was further complicated with two episodes of left-sided spontaneous pneumothorax nine months after Nuss procedure (one month apart). On the CT scan the retrosternal bar showed no complications, namely displacement. Thoracoscopic apical resection of emphysematous lung and apical pleural abrasion were performed, without complications.

One month after the second pneumothorax (11 months after the Nuss procedure), he was again evaluated in

the Emergency Department due to chest discomfort. Pneumothorax was immediately excluded by chest radiography, and the complaints were attributed to the nickel allergy. He was medicated with oral corticosteroids and antihistamine, with resolution of the symptoms and, to the present-day, with more than one year of follow-up, no more symptoms due to nickel allergy have been reported, with nickel contact eviction in his daily life.

DISCUSSION

This case describes the onset of nickel allergy after a Nuss procedure in an adolescent with a negative preoperative test for allergies, and no relevant past medical and family history. His symptoms resolved with oral medication, without the need for bar removal.

Similar cases are very seldom reported. In a published series with 932 patients, 90 presented positive preoperative skin testing and for that reason received a titanium bar. From the 277 with negative testing, five (1.8%) presented, however, allergic symptoms after Nuss procedure, with the need for premature bar removal in three of them.⁴

Contact allergy is frequent in the general population, especially for nickel, with 8-10% of children and adolescents affected.⁵

A review including 5680 patients subjected to the Nuss procedure showed allergy presenting in 2.7% of them. The clinical symptoms vary in intensity, from local dermatological symptoms as rash, oedema and erythema, to generalized exfoliative dermatitis, pleuritis and pericarditis, which occur rarely. Corticosteroids and histamine antagonists can be effective, although, in some cases the metal bar has to be replaced with a titanium substitute.⁶

In this case, even though the patient presented a pneumothorax as a postoperative complication, he also presented two spontaneous pneumothorax nine months after surgery. This complication was never associated with nickel allergy to a Nuss metal bar before. YB Wang et al reported a case of a pneumothorax in an adult secondary to idiopathic hypereosinophilic syndrome and postulated that pneumothorax could be in the origin of eosinophilic direct pleural injury⁷. Know BI et al also suggested that a mechanism mediated by eosinophils may play a role in the development of pneumothorax in allergic diseases⁸. Therefore, we hypothesize that this complication could have been secondary to the metal allergy.

Given the potentially severe consequences of metal allergy after a Nuss procedure, at our center we test every patient beforehand, regardless of personal history, as suggested by other centers.³ Even though false negative cases may appear, as happened with our patient, several of the existent allergies will be detected pre-surgery with this approach, and this way we could spare patients with positive tests from possible complications.

Doctors must be aware, however, that despite its rarity, a metal allergy can appear after surgery regardless of a preoperative negative test. Symptoms must be early recognized, and adequate treatment implemented as soon as possible.



Figure 1

Chest x-ray after bar placement.

REFERENCES

1. Nuss D, Kelly RE, Jr., Croitoru DP, Katz ME. A 10-year review of a minimally invasive technique for the correction of pectus excavatum. *J Pediatr Surg.* 1998;33(4):545-52.
2. Kelly RE, Goretsky MJ, Obermeyer R, Kuhn MA, Redlinger R, Haney TS, Moskowitz A, Nuss D. Twenty-one years of experience with minimally invasive repair of pectus excavatum by the Nuss procedure in 1215 patients. *Ann Surg.* 2010 Dec;252(6):1072-81.
3. Shah B, Cohee A, Deyerle A, Kelly CS, Frantz F, Kelly RE, et al. High rates of metal allergy amongst Nuss procedure patients dictate broader pre-operative testing. *J Pediatr Surg.* 2014;49(3):451-4.
4. Obermeyer RJ, Gaffar S, Kelly RE Jr, Kuhn MA, Frantz FW, et al. Selective versus routine patch metal allergy testing to select bar material for the Nuss procedure in 932 patients over 10 years. *J Pediatr Surg.* 2018 Feb;53(2):260-264.
5. Ahlström MG, Thyssen JP, Wennervaldt M, Menné T, Johansen JD. Nickel allergy and allergic contact dermatitis: A clinical review of immunology, epidemiology, exposure, and treatment. *Contact Dermatitis.* 2019;81(4):227-41.
6. Gałazka P, Leis K, Kroczyk K, Baska A, Kazik J, Czajkowski R. Metal allergy after the Nuss procedure for pectus excavatum: a review. *Postepy Dermatol Alergol.* 2020 Dec;37(6):848-852. doi: 10.5114/ada.2020.102094.
7. Wang YB, Han YJ, Uchida K, Zhao BC, Chen KB, Ma WY, Xie FJ, Liu TL, Zhang LX. Pneumothorax as the initial manifestation of idiopathic hypereosinophilic syndrome. *Ann Thorac Surg.* 2014 Nov;98(5):1838-41.
8. Kwon BI, Hong S, Shin K, Choi EH, Hwang JJ, Lee SH. Innate type 2 immunity is associated with eosinophilic pleural effusion in primary spontaneous pneumothorax. *Am J Respir Crit Care Med.* 2013 Sep 1;188(5):577-85.