

CASE REPORT

Metal allergy as a late-onset complication of the Nuss procedure in a pediatric patient

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Abstract

We report the case of a 14-year-old boy with pectus excavatum who developed a metal allergy to stainless steel bar as a late-onset complication after the Nuss procedure. He did not have atopic diathesis. Treatment with oral steroids was effective and the metal bar was successfully removed 2 years later.

KEYWORDS

metal allergy, Nuss procedure, pectus excavatum, stainless steel, titanium

1 | CASE REPORT

A 14-year-old boy with a severe pectus excavatum and a Haller computed tomographic index (HI)¹ of 3.96 and scoliosis was treated with the Nuss procedure in the University Children's Hospital Zagreb. At the age of 10, he started to complain of chest pain and shortness of breath during exercise. Lung function measurements revealed mild restrictive ventilation dysfunction. His past medical history and family history did not reveal allergic diseases or eczema. The Nuss procedure was performed with the use of a stainless steel bar fixed with lateral stabilizers. Early postoperative period was unremarkable. Eight months after the procedure the patient was hospitalized in the Clinical Hospital Centre in Osijek due to a left-sided hemothorax and pneumonia. A chest tube was inserted and 2000 mL of blood was evacuated from the left pleural cavity. After the successful tube thoracostomy, the patient's condition was stable thus emergent thoracotomy was not performed. Empiric pneumonia antibiotic therapy was initiated and had good clinical response. The patient was discharged 10 days after the admission in good general condition. Bacterial cultures of the pleural effusion and blood were negative. After 2 weeks he was rehospitalized due to a left-sided pleural effusion and pericardial effusion without signs of cardiac tamponade. Again, blood and pleural cultures were negative. He was treated with indomethacin with no clinical improvement and finally effusions resolved after 7 days of prednisone

1 mg/kg/day. After 1 month he became highly febrile with a temperature of 39°C accompanied by a dry cough. Laboratory tests demonstrated an increased level of inflammatory parameter C-reactive protein (CRP) 307 mg/L (0–10 mg/L), and a white blood cell count (WBC) of $10 \times 10^9/L$, dominantly neutrophilia. Thoracic X-rays revealed a left-sided pleural effusion and a pericardial effusion (Figures 1 and 2). Meropenem therapy was started and the patient was referred to our hospital on day 3.

On admission he was afebrile, cardiorespiratory stable, without distension of great vessels. A chest auscultation revealed a pericardial rub, quiet heart sounds, and bilaterally decreased breath sounds in the bases. Blood test revealed increased inflammatory parameters (an erythrocyte sedimentation rate (ESR) 45 mm/h, CRP 180 mg/L, WBC $6.9 \times 10^9/L$ ($4.5\text{--}13 \times 10^9/L$), neutrophils $4.54 \times 10^9/L$ ($1.8\text{--}8 \times 10^9/L$), lymphocytes $1.73 \times 10^9/L$ ($1.1\text{--}6.5 \times 10^9/L$), monocytes $0.56 \times 10^9/L$ ($0.2\text{--}0.8 \times 10^9/L$), eosinophils $0.09 \times 10^9/L$ ($<0.81 \times 10^9/L$), basophils $0.05 \times 10^9/L$ ($<0.11 \times 10^9/L$). The electrocardiogram (ECG) showed a sinus rhythm of 98 beats/min, low voltage of QRS complexes, an incomplete right bundle branch block and a negative T-wave in leads V1 to V2. A thoracic ultrasonography showed a pericardial effusion all around the heart, up to 4 cm in diameter and bilateral pleural effusions (left 32 mm and right 30 mm). We performed blood and pleural cultures and no diagnostic pathogens grew from any cultures taken. Immunological work-up revealed increased antistreptolysin O titer (AST-O) 1942 IU/mL (<200 IU/mL), immunoglobulin G (IgG) 21.79 g/L ($7.0\text{--}16.0$ g/L) and IgM 3.16 g/L ($0.4\text{--}2.3$ g/L). Immunoglobulin E (IgE), eosinophil cationic protein (ECP), rheumatoid factor (RF), and complement component C3 and C4 were in the normal range. Skin

Abbreviations: AST-O, antistreptolysin O titer; C, complement component; CRP, C-reactive protein; ECG, electrocardiogram; ECP, eosinophil cationic protein; ESR, erythrocyte sedimentation rate; Ig, immunoglobulin; RF, rheumatoid factor; WBC, white blood cell count.

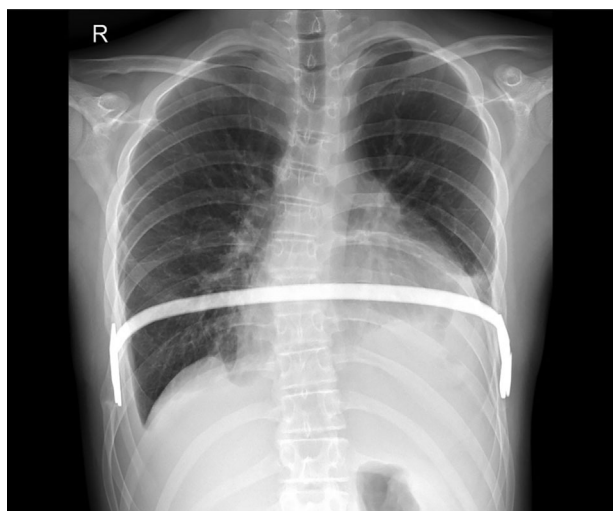


FIGURE 1 Thoracic x-rays showing pericardial effusion and left-sided pleural effusion after Nuss procedure

patch-tests to potassium dichromate and nickel sulfate were positive. With suspicion of metal allergy to the stainless steel bar, oral steroid therapy (prednisone 32 mg/day; 0.5 mg/kg/day) was initiated. The steroid therapy resolved his symptoms immediately. The oral steroids were decreased to 4 mg/day during the next 2 weeks, and continued until the bars were removed 2 years later. The allergic reaction did not recur, and no side effects of the steroids were observed.

2 | DISCUSSION

The Nuss procedure is a minimally invasive procedure which consists of a metal bar placement in the chest cage to repair pectus excavatum. The Nuss bar is typically removed after about two to 4 years because earlier removals are associated with a higher rate of pectus excavatum

recurrence.¹ Metal allergies to stainless steel bar are one of the complications associated with this procedure.²

Allergy to nickel, cobalt, or chromium occurs in approximately 13% of the population.³ The stainless steel bar used in the Nuss procedure contains various metals, such as iron, chromium, nickel, molybdenum. These metals are not antigens themselves, but they become ionized in the body, and, thereafter bind with natural proteins and cause allergic reactions.⁴ Metal allergies are frequently misdiagnosed as surgical infections although the management of these two conditions is quite different.² In our patient, the previous good clinical response to steroids and negative blood and pleural effusion cultures were suggestive of the diagnosis of metal allergy.

Before the Nuss procedure, patients should be evaluated for allergies to jewelry, orthodontic braces, metal buttons/snaps on clothing, food, and also signs and symptoms of atopy (allergic rhinitis, eczema, asthma). If any of these indications is found in the patients' history, a patch test to metals should be performed.⁴ Titanium is a biocompatible metal and is rarely a cause of allergy symptoms. Patients with a prior history of metal allergy, those with a family history of metal allergy, those who's test is positive for nickel, cobalt or some other constituent of stainless steel and patients who suffer from eczema should receive a titanium bar.² However, titanium bar placement is much more expensive (4-5 times more) and more complicated than the steel one.⁵ Furthermore, titanium can also cause metal allergy in patients treated with the Nuss procedure even in patch test negative patients.⁴ Our case clearly shows that conversion from non-allergic to allergic status is possible even after the Nuss procedure. According to Rushing et al² in patients who presented with metal allergy after the Nuss procedure, the presentation time of symptoms averaged 40 days (range 5-140 days). In our patient the metal allergy was presented as pleural and pericardial effusions 8 months after the Nuss procedure. According to a positive patch test to metals, we can only hypothesize that our

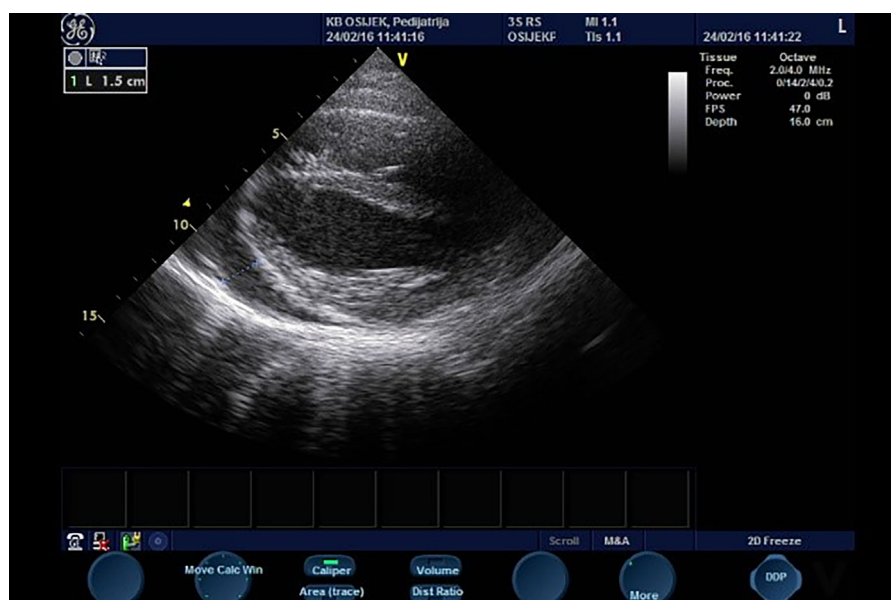


FIGURE 2 Echocardiographic image of pericardial effusion

patient evolved a type IV delayed hypersensitivity reaction (normal level of total IgE and eosinophils count) to stainless steel bars what was manifested as a late onset complication of the Nuss procedure. We can not explain an earlier development of a metal allergy manifestation in literature cases except that the complications occurred in already atopic patients whereas in our case more time was needed for a metal to become ionized and a conversion from non-allergic to allergic phenotype. A potential trigger for the development of an allergic phenotype and sensitization to a metal is a matter of debate (surgery trauma, foreign body, infection, puberty, etc.).

It is of great importance to send patients to an allergy specialist before performing the Nuss procedure and during the follow-up due to the high prevalence of metal allergy in the general population and the possibility of conversion from non-allergic to metal-allergic status following the implantation of a metal bar as it is shown in our case.

ACKNOWLEDGMENT

No financial support provided.

CONFLICTS OF INTEREST

The authors have no conflicts of interest to declare.

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How to cite this article: Navratil M, Batinica M, Ivković-Jureković I. Metal allergy as a late-onset complication of the Nuss procedure in a pediatric patient. *Pediatric Pulmonology*. 2018;1–3. <https://doi.org/10.1002/ppul.24072>