




Nickel allergy as a probable cause of complications after closing the atrial septal defect type ostium secundum with the Amplatzer septal occluder in a 6-year-old girl

Uczulenie na nikiel jako prawdopodobna przyczyna komplikacji po zamknięciu zapinką Amplatzera ubytku w przegrodzie międzyprzedsionkowej u 6-letniej dziewczynki

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Abstract

A 6-year-old girl underwent a successful closure of a large atrial septal defects (ASD2) with an Amplatzer septal occluder (ASO). After 2 years, she required surgical removal of the implant due to a local inflammatory reaction and leakage around the occluder, closure of the ASD with an autopericardial patch and removal of the newly formed fistula between the aorta and the left atrium (Ao-LA). Fistula regeneration requiring surgical treatment was observed twice over the next two years. During this time, incidents of subfebrile body temperature with positive inflammatory markers and sterile blood cultures occurred. Interventional reclosure of the fistula was withdrawn due to its close location to the coronary artery, as well as the suspicion of an allergic reaction to nickel compounds contained in ASO, supposedly responsible for recurring complications and a very unusual course of treatment. An allergy patch test was performed, resulting positive (+++) at 96 hours. Currently, after 3 years of follow-up, there is no evidence of septal shunt or reformation of Ao-LA fistula. The patient remains asymptomatic under outpatient care, avoiding contact with nickel compounds in any form and state.

Key words: defect in the atrial septum, Amplatzer occlude, allergy to nickel, fistula

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Introduction

Atrial septal defects (ASD2) are the most common congenital heart defects [1]. In the diagnosis of ASD, the “gold standard” is transthoracic (TTE) and transoesophageal (TEE) echocardiography [2, 3]. A small ASD2 (3–4 mm)

usually remains under observation, while larger ones require closure [4]. Standard management is TEE-guided transvascular closure of the defect with the use of an occluder, while surgical treatment is reserved for atypical forms [5, 6]. Recommendations after closing the defect include prophylaxis of infective endocarditis and aspirin admission

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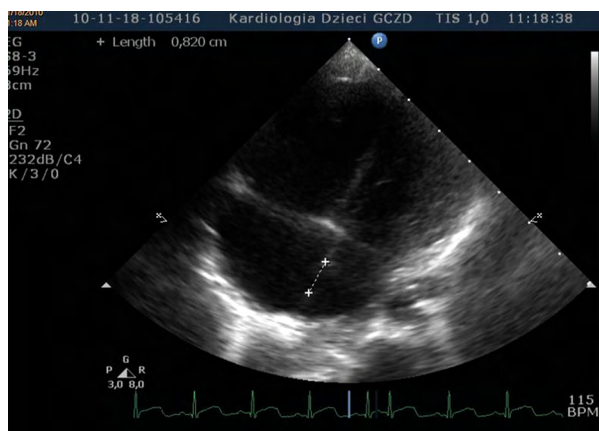


Figure 1. Transthoracic echocardiography 4-chamber view. Atrial septal defects 2 approx. 8.2 mm. The enlarged right atrium and right ventricle

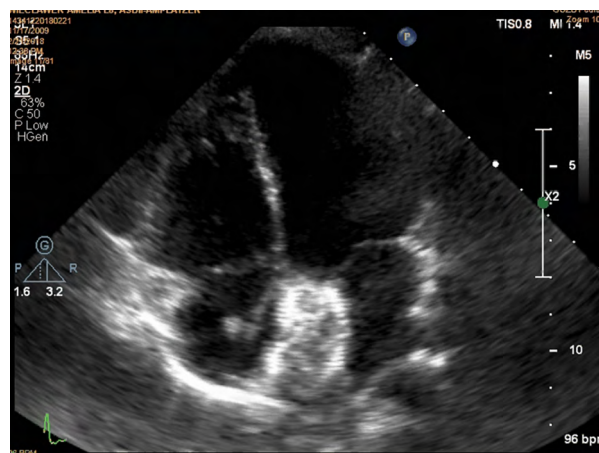


Figure 2. Transthoracic echocardiography 4-chamber view. Correctly positioned implant, No septal leakage. Significant reduction in the size of the chambers of the right heart

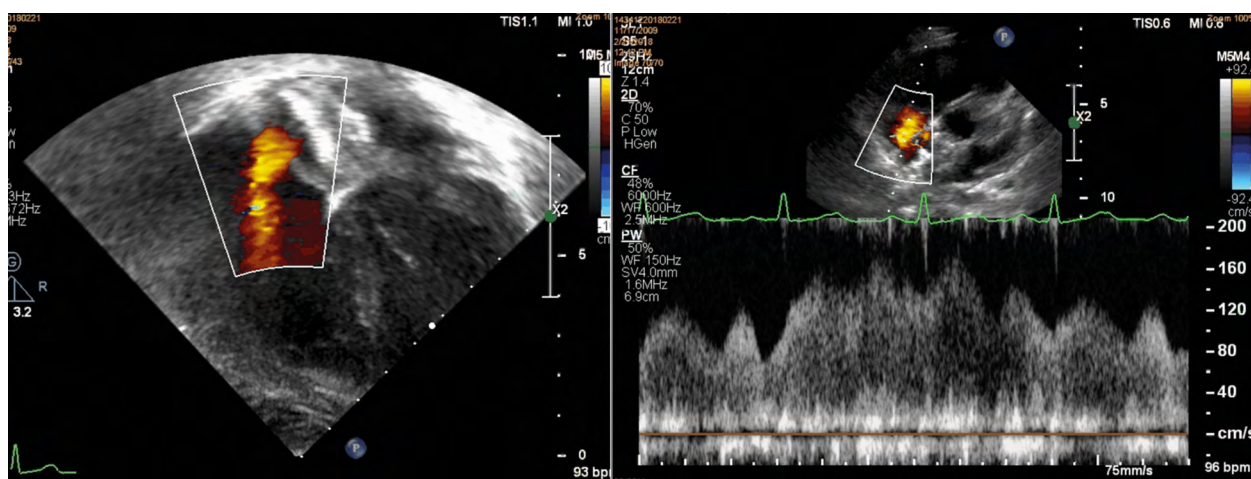


Figure 3. There is a left-to-right volume shunt around the Amplatzer occlude

over 6 months after the procedure [5, 7]. Complications after endovascular intervention are relatively rare and are estimated at 2.2–8.6% [5].

Case report

A 6-year-old girl was admitted to the pediatric cardiology department to close ASD2 diagnosed at 1 year of age. Physical examination revealed a systolic murmur at the left parasternal border and a stiffly split second heart sound. The electrocardiogram showed the right axis deviation and features of a partial right branch bundle block. Transthoracic echocardiography confirmed an ASD2, 17–20 mm in diameter with Qp:Qs = 1.9:1. The patient underwent a transvascular closure with a 19 mm Amplatzer device

(Figure 1). The procedure and the early postoperative period were uneventful, the implant correct position and no residual flow was observed. An increasing leakage around the implant was found after 6 months in control TTE, along with hyperechogenic adjacent structures. After another 18 months, she was referred to the department due to fatigue, increased sweating and lack of appetite. Performed ultrasound imaging examinations (TTE, TEE) and magnetic resonance imaging exposed a correctly positioned implant and an abnormal linear structure parallel to the right atrial disc, as well as significant flow towards the right atrium (Figure 2). In addition, a fistula approximately 2 mm in diameter was visualized between the aorta and the left atrium (Ao-LA) (Figure 3). The girl underwent a cardiosurgical consult and qualified for surgical treatment (operation No. 1).

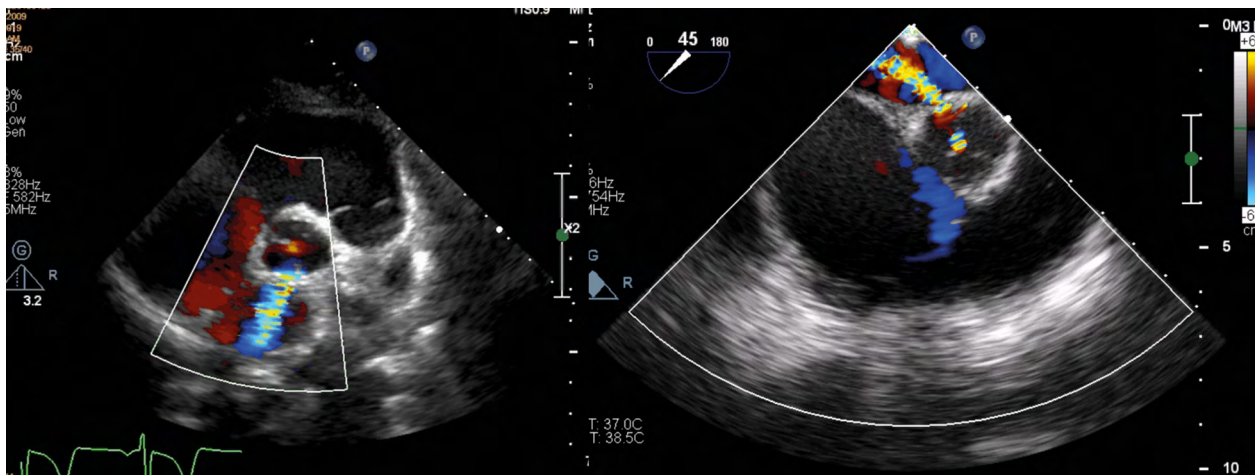


Figure 4. The fistula between the aorta and the left atrium. On the left, transthoracic echocardiography. On the right, the same image in transoesophageal echocardiography

The implant was removed, along with the adjacent cyst and fibrous tissue. The ASD was closed with an autopericardial patch and the Ao-LA shunt was ligated. After 8 days, the fistula recanalized and another operation was required to re-close the fistula (operation No. 2). The patient was discharged after several days and readmitted a month later due to fever, decreased appetite, malaise, increased fatigue and chest pain. Laboratory tests showed an increase in inflammatory markers with sterile blood cultures. Imaging revealed an Ao-LA fistula of approximately 2.5 mm. A wait-and-see attitude was adopted, with the introduction of antibiotic therapy, angiotensin-converting enzyme inhibitors, β -blockers and dehydrating drugs treatment. After 2 years, the fistula was still present, hence a decision for another reoperation was undertaken. Interventional treatment was abandoned due to the close distance of the fistula to the coronary artery. Moreover, a hypothesis of contact allergy to nickel was taken under consideration, resulting in such complications. The suspicion was later confirmed by a positive nickel allergy patch test ([+++] after 96 hours). Another operation was performed (operation no. 3) and the fistula was successfully closed. In a 3-year follow-up, no recanalization of the fistula (Figure 4) is observed. The patient remains asymptomatic under outpatient care, avoiding contact with nickel compounds in any form and state.

Discussion

The presented atypical course of ASD2 closure with the Amplatzer device indicates that allergy to nickel compounds contained in the implant may prevent effective intervention treatment. The known post-interventional complications include embolization, erosion, headaches, thrombus formation or vegetation, tachyarrhythmias, access site

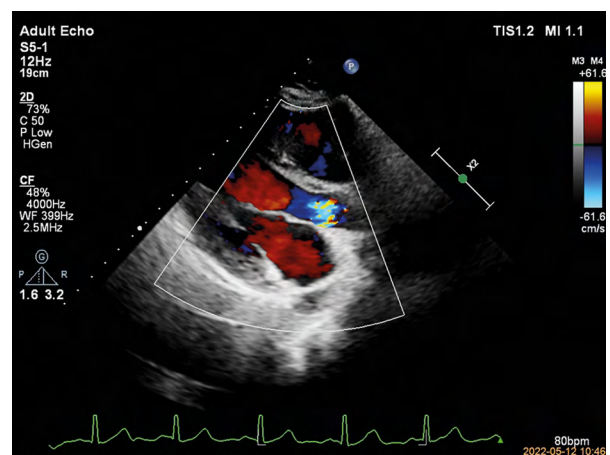


Figure 5. Transthoracic echocardiography, left ventricular long axis. Follow-up observation; no aorta and the left atrium shunt

complications, bleeding, etc. [5, 8, 9], while nickel allergy is listed last as a possible complication and the frequency of this reaction is not known [5]. The implant is covered with nitinol, which may release traces of nickel into the blood and cause an allergic reaction. In general, symptoms appear within the first few days or months and include headache, rash, fever, palpitations and shortness of breath, sometimes with pericardial effusion. If pharmacological treatment is ineffective, it may be necessary to remove the occluder [8, 9]. It is indicated that in patients with a positive family history, a nickel allergy skin test should be performed before the procedure [10]. This patient has not provided any information indicating her allergy; therefore the test was not performed. To the authors' knowledge and review of the literature, the child has not developed a reaction around the clasp with the simultaneous formation of

a self-reproducing fistula between the Ao-LA, requiring as many as three operations to close.

Conclusions

Each interventional treatment of an interatrial defect carries the risk of rare and unexpected complications, including an allergic reaction to nickel with the formation of a fistula. Surgical treatment of a fistula carries a high risk of low effectiveness.

Conflict of interest

The authors do not report any financial or personal connections with other persons or organizations that could adversely affect the content of the publication and claim the right to this publication

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Streszczenie

Sześciolatniej dziewczynce skutecznie zamknięto duży ASD2 za pomocą zapinki Amplatzer (ASO). Po 2 latach wymagane było chirurgiczne usunięcie implantu z powodu wytworzenia miejscowego odczynu i powstania przecieku wokół zapinki, zamknięcie ASD łątką z własnego osierdzia oraz usunięcie nowo powstałej przetoki pomiędzy aortą a lewym przedsionkiem (Ao-LA). W ciągu kolejnych 2 lat jeszcze dwukrotnie obserwowano samoodtwarzanie się przetoki wymagające leczenia chirurgicznego. W tym czasie pojawiały się stany gorączkowe z dodatnimi wskaźnikami zapalnymi, jednak posiewy z krwi były jałowe. Nie zdecydowano się na leczenie interwencyjne przetoki ze względu na bliską lokalizację z naczyniem wieńcowym, jak również z powodu podejrzenia reakcji uczuleniowej na związki niklu zawarte w ASO, które mogły odpowiadać za komplikacje i bardzo nietypowy przebieg leczenia. Wykonano uczuleniowy test płatkowy, który był dodatni (+++) po 96 godzinach. Aktualnie, po 3 latach obserwacji, nie stwierdza się przecieku na przegrodzie międzyprzedsionkowej ani ponownego wytworzenia przetoki Ao-LA. Dziewczynka pozostaje pod okresową kontrolą kardiologiczną i nie wykazuje żadnych niepokojących objawów, unika kontaktu ze związkami niklu, które mogą być obecne w produktach spożywczych oraz licznych przedmiotach codziennego użytku.

Słowa kluczowe: ubytek w przegrodzie międzyprzedsionkowej, zapinka Amplatzer, alergia na nikiel, przetoka

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