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Case Report

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Spontaneously resolved pseudoaneurysm after valve-sparing aortic root replacement in Marfan syndrome: a case report‡

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Abstract

Pseudoaneurysm after valve-sparing aortic root replacement is uncommon but potentially life-threatening. We report an unusual case of spontaneous resolution of an aortic root pseudoaneurysm that developed in a 14-year-old boy with Marfan syndrome following valve-sparing aortic root replacement. This case suggests that, in carefully selected situations, non-surgical management with intensive follow-up may be beneficial.

A 14-year-old male patient was referred due to aortic regurgitation and aortic root aneurysm (Figure 1a, b). He had been under follow-up for aortic root dilatation associated with Marfan syndrome. We performed valve-sparing aortic root replacement using a 28-mm-sized Gelweave Valsalva graft (Terumo Aortic, Sunrise, FL, USA). We placed five sub-annular sutures: three at each cusp's nadir and two at the commissures using 5-0 polypropylene, except at the right-noncoronary commissure to avoid conduction disturbance. Haemostatic running sutures were placed between the graft and aorta using 5-0 polypropylene. Both coronary buttons were reimplanted using continuous 6-0 polypropylene sutures, reinforced with interrupted stitches.

Postoperative CT showed no abnormal findings, but a persistent fever delayed discharge. On day 15, he developed chest pain and blurred vision; CT revealed new aortic root pseudoaneurysms (Figure 1c, d).

An urgent reoperation was performed to repair the pseudoaneurysms. During cardiopulmonary bypass and with an aortic cross-clamp in place, we identified two dehisced sites; both sites were in native aortic tissue, showing no evidence of suture line laceration or contact with any artificial materials. These sites were primarily closed (Supplementary Figure 1).

Twelve days after the second operation, repeat CT revealed a newly developed pseudoaneurysm which was anterior to the right coronary sinus, while previously identified pseudoaneurysms had been resolved (Figure 2a–d). Despite recommendations for reoperation, the patient's parents refused surgery, and he was discharged with antihypertensive drugs.

Five months later, CT angiography showed neither haemopericardium nor any contrast leakage suggestive of pseudoaneurysm (Figure 2e). The patient has remained stable for more than 2 years, with normal daily activities.

This study was approved by the Institutional Review Board of Seoul National University Hospital, and written informed consent for publication was obtained from the patient's legal guardian.

Discussion

This case reports spontaneous resolution of pseudoaneurysm that developed after aortic root surgery. Although rare, pseudoaneurysm following aortic surgery can be life-threatening, even in asymptomatic patients. Therefore, corrective surgery is generally recommended.[1] Since the late 20th century, open surgical repair has been the treatment of choice for pseudoaneurysms. However, with advancements in interventional medicine, alternative techniques such as coiling[2] and thoracic endovascular aortic repair[3] have been increasingly utilised in eligible patients.

Several risk factors for pseudoaneurysms following aortic surgery include aortic dissection, connective tissue disorders, infection, and the use of biological glues, causing tissue necrosis.[1] Our patient with Marfan syndrome underwent surgery using fibrin glue, not biological glue. Despite suspected infection, cultures were negative.

It is well established that the combination of beta-blockers and angiotensin receptor blockers can slow the rates of aortic enlargement and delay the need for aortic surgery in Marfan syndrome.[4] Given the patient's refusal of further intervention, he was under short-term follow-up with prescribed amlodipine 5 mg, atenolol 25 mg, and losartan 50 mg.

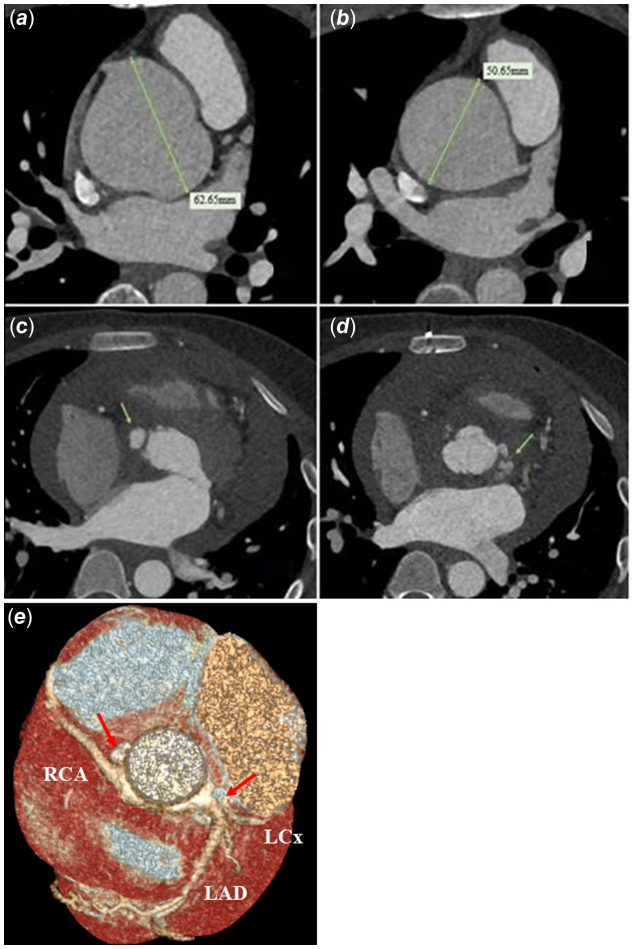


Figure 1. Repeat computed tomography (CT) findings showing two pseudoaneurysms around both coronary buttons. (***a***, ***b***) Preoperative CT. (***c***,***e***) CT on postoperative day 15 demonstrating newly developed pseudoaneurysms after the patient complained of chest pain.

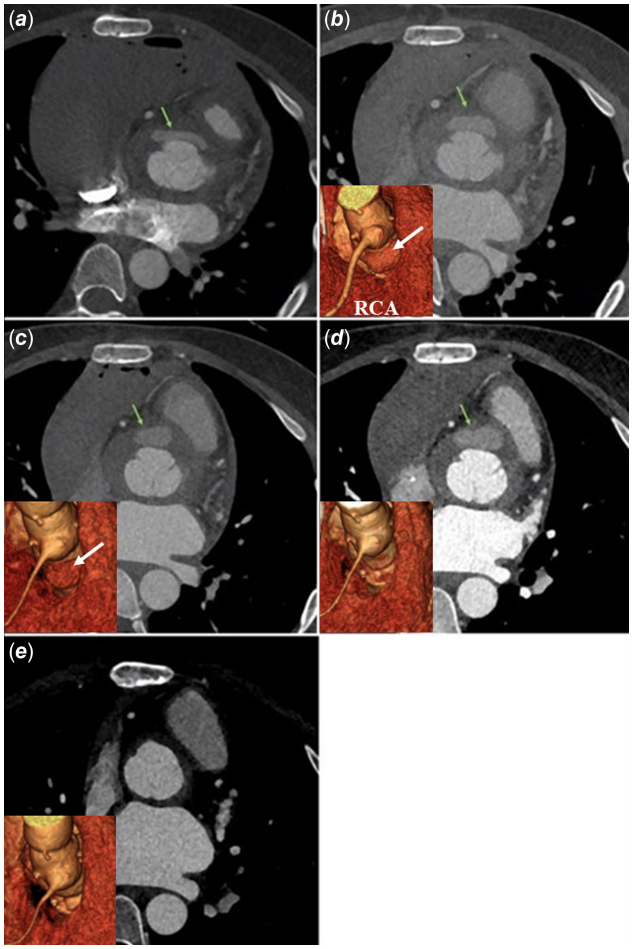


Figure 2. Serial postoperative computed tomography (CT) findings. (***a***) CT on postoperative day (POD) 31/12 (from first/second operation) showing a newly developed pseudoaneurysm around the right coronary cusp. (***b***,***d***) Follow-up CTs on POD 33/14, POD 42/23, and POD 51/32 showing gradual resolution. (***e***) CT at the last follow-up (19 weeks after discharge) showing complete disappearance of the pseudoaneurysm.

Although surgical repair remains the standard treatment for pseudoaneurysms, this case indicates that spontaneous resolution may occur under highly uncommon circumstances. Therefore, observation should be considered only in exceptional situations where reoperation poses a substantial risk, with close surveillance.

Supplementary material. The supplementary material for this article can be found at https://doi.org/10.1017/S1047951125110305.

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Competing interests. None to disclose.

Ethics standard. This case report was conducted in accordance with the Declaration of Helsinki and was approved by the Institutional Review Board of Seoul National University Hospital (IRB No. 2411-006-1581, approved on 2024-11-04).

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