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Brief Report

Atrial septal defect repair by inversion of a juxtaposed left atrial appendage

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Abstract A 7-year-old child was noted to have dextrojuxtaposition of the left atrial appendage at the time of surgical atrial septal defect repair. Given the favourable anatomic location and size of the atrial appendage, it was inverted and used to close the atrial defect. This is the first report of atrial septal defect repair using a juxtaposed atrial appendage. The cardiac anatomy and theoretical benefits of this repair are discussed.

Keywords: Congenital cardiac disease; cardiac surgery; anatomy

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Juxtaposition of the atrial appendages is a rare congenital cardiac defect in which both auricles are located on either the right or left side of the great arteries. Dextrojuxtaposition of the left atrial appendage to the right of the great vessels is exceedingly less common than levojuxtaposition of the right atrial appendage, and has been associated with other severe cardiac anomalies in all but one reported case.[1] Here we describe the incidental finding of juxtaposition of the left atrial appendage during an atrial septal defect repair in a patient with no associated anomalies aside from the atrial septal defect, and the first report of using an *in situ* juxtaposed atrial appendage as an atrial septal defect patch.

Case report

The child is a former 29-week premature infant without other significant medical history or apparent sequelae of prematurity. At 7 years of age, she presented with complaints of midsternal chest discomfort with activity. On examination, she was noted to have a right ventricular heave and a grade two-of-six murmur at the left sternal border. Electrocardiogram demonstrated right atrial enlargement and right ventricular hypertrophy. Transthoracic echocardiography demonstrated a 2.0-centimetre secundum-type atrial septal defect with left-to-right shunting and mild-to-moderate tricuspid regurgitation. The right atrium and ventricle were severely dilated with mildly reduced systolic function. Notably, the atrial septal defect was superiorly located with minimal tissue remaining in the superior limbic band. Owing to the size of the defect and proximity to the aortic root, it was not amenable to percutaneous transcatheter device closure.

The patient underwent partial sternotomy for elective atrial septal defect repair. A previously undiagnosed dextrojuxtaposition of the left atrial appendage was noted, with the appendage arising from the right side of the left atrium between the superior caval vein and aortic root (Fig 1). A careful search did not reveal any abnormal venous connections or other apparent malformations. After the patient was placed on cardiopulmonary bypass and the heart arrested, the enlarged right atrium was opened. A 2.0×1.5-centimetre atrial septal defect was noted with a very small superior limbic band (Fig 2a). The left atrial appendage was inverted to inspect for thrombus, and noted to be approximately 2 centimetres in length. Given the superior

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position of the atrial septal defect, the superior limbic band was the reflection point when inverting the left atrial appendage. This allowed the inverted appendage to be naturally positioned in the atrial septal defect (Fig 2b).

The inverted left atrial appendage was then secured circumferentially as a double-layer atrial septal defect patch with a running 5-0 polypropylene suture (Fig 2c). Sutures were not required at the superior margin, as the base of the inverted left atrial appendage comprised this portion of the repair. At the conclusion of the repair, intraoperative transoesophageal echocardiography demonstrated a stable patch with no residual defect. There was no obstruction or distortion of systemic or pulmonary venous flow. The child was extubated and transferred to the paediatric cardiac intensive care unit where she had an uneventful recovery. A transthoracic echocardiogram performed on post-operative day 4 demonstrated a stable patch repair without residual left-to-right shunting. Repeat transthoracic echocardiogram at 1 year demonstrated a stable repair with appropriate regression of right ventricular enlargement.

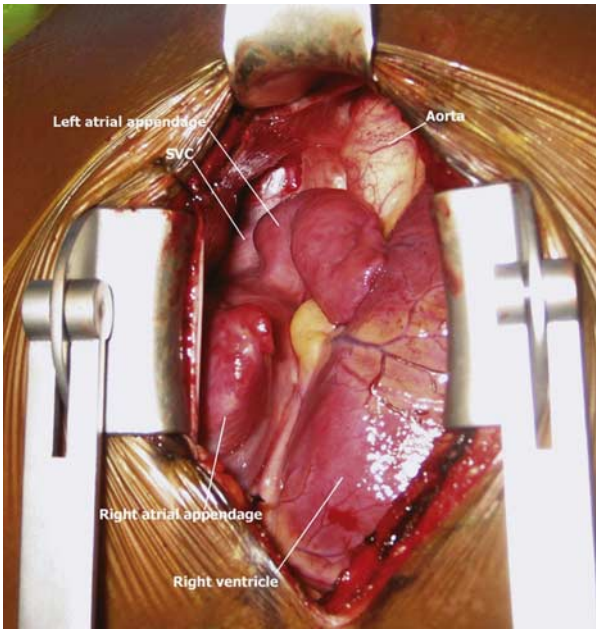

Figure 1.
Intraoperative photograph via partial lower median sternotomy demonstrating juxtaposition of the left atrial appendage to the right of the aorta.

Discussion

Juxtaposition of the left atrial appendage is a rare congenital cardiac defect, with less than 65 cases described in the literature.[2,3] Juxtaposition of the left atrial appendage occurs six to eight times less frequently than juxtaposition of the right atrial appendage,[3,4] and was found in 0.09% – 7 out of 7996 – of patients referred for echocardiography in one series[3] and 0.1% – 2 out of 1842 – of hearts with congenital lesions in another.[5] Juxtaposition of the left atrial appendage is an ominous finding, associated with complex atrioventricular defects in half of all cases,[2–4] and other severe cardiac anomalies in all but one reported case.[1] The patient described here represents the second only reported case of juxtaposition of the left atrial appendage without other major intracardiac anomalies and normal segmental anatomy.

Atrial septal defects are common congenital cardiac lesions that are frequently closed in the cardiac catheterisation laboratory.[6] Our patient had a large atrial septal defect with a small superior rim of tissue that would have made percutaneous closure difficult. Surgical closure of atrial septal defects is

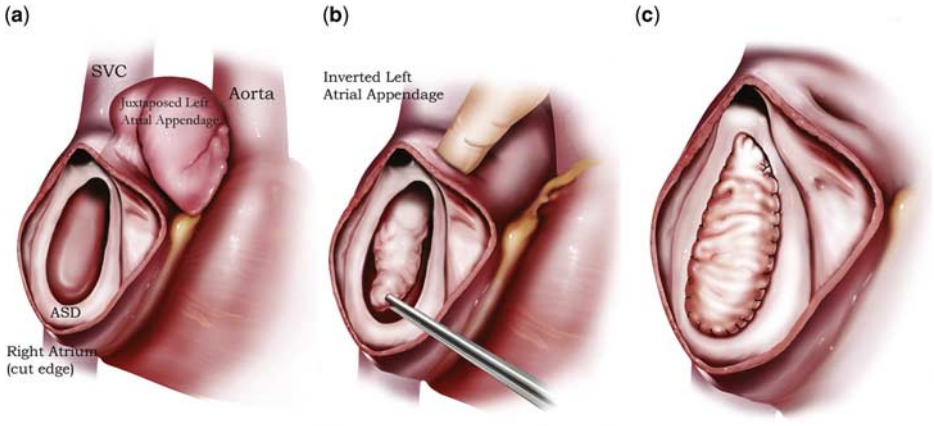

Figure 2.
*Artist's rendition with the free wall of the right atrium cut away demonstrating (**a**) the atrial septal defect and juxtaposed left atrial appendage, (**b**) inversion of the juxtaposed left atrial appendage, and (**c**) completed patch repair of the atrial septal defect.*

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usually performed with pericardial or synthetic patches, although repair with free right atrial wall has recently been reported, with normal action potentials demonstrated within the patches.[7] The unexpected finding of a juxtaposed left atrial appendage in our patient allowed a novel repair technique. The inverted left atrial appendage has the benefit of being covered with native endocardium on both atrial surfaces to minimise the risk of foreign body reaction, thromboembolism, and patch endocarditis associated with intracardiac prosthetic material.[8,9] Furthermore, this viable, muscular patch will continue to grow with the patient. As there is no interruption of the myocardium continuous with the left atrial appendage by an incision or suture line, the electromechanical properties of the patch are believed to be undisturbed.

In summary, this report describes the second case of juxtaposition of the left atrial appendage without other associated major cardiac anomalies and the novel use of the juxtaposed atrial appendage for atrial septal defect repair.

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