Late detection of communication between juxtaposed atrial appendages

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Abstract

A 40-year-old woman underwent an atrial septal defect closure with juxtaposition of the atrial appendages without communication between both appendages 4 years before presentation. She recently experienced desaturation on exercise, and right-to-left shunt coming from the residual communication between both appendages was found. The residual communication was closed from the right to the left atrium. Herein, we report a rare case of juxtaposition of the atrial appendages with residual communication between them after an atrial septal defect closure.

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Abstract:

A 40-year-old woman underwent an atrial septal defect closure with juxtaposition of the atrial appendages without communication between both appendages 4 years before presentation. She recently experienced desaturation on exercise, and right-to-left shunt coming from the residual communication between both appendages was found. The residual communication was closed from the right to the left atrium. Herein, we report a rare case of juxtaposition of the atrial appendages with residual communication between them after an atrial septal defect closure.

Introduction:

Juxtaposition of the atrial appendages (JAA) is a rare congenital cardiac malformation with both atrial appendages located on the same side of the great arteries. Majority of reports described this disease to be complicated with other malformations; however, communication between the appendages is rarely reported, and thereby, its surgical procedure is not yet established. We present the case of a 40-year-old woman with left JAA associated with residual communication between them after an atrial septal defect closure.

Case Report:

A 40-year-old woman presented with dyspnea and desaturation on exertion. She had a history of atrial septal defect (ASD) patch closure 4 years ago and chronic pulmonary artery hypertension [mean pulmonary artery pressure (mPAP): 31 mmHg], and JAA was found simultaneously. During that time, no obvious communication was found between the appendages before or after the operation. Enhanced computed tomography (eCT) was performed for detailed examination on this admission and showed slight communication between the appendages (Fig. 1). Additionally, right heart catheter angiography showed a right-to-left shunt through the tunnel. The catheter data showed mPAP of 30 mmHg, pulmonary vascular resistance of 232 dyn s/cm^5, and Qp/Qs of 1.1. Cardiology team decided to close this shunt due to a risk of paradoxical embolus.

Through a median resternotomy, cardiopulmonary bypass was established. After right atriotomy, the tunnel ostium was observed in the right atrium (Fig. 2A). Next, left atriotomy from the right side of the left atrium was performed, and the ostium at the left atrium was found by pouring saline from the ostium of the right atrium (Fig. 2B). Both ostia were closed with polypropylene running suture. The subsequent course was uneventful. Postoperative eCT showed no residual communication. Dyspnea on effort gradually improved, and desaturation on exertion was resolved; thus, further follow-up is required.

Discussion:

JAA is a rare congenital cardiac malformation named by Dixon in 1954.[1] It is defined as the presence of both appendages on the same side of the right or left great arteries.[1,2] Left JAA is more frequent than right JAA and is sometimes classified as complete or partial according to its location.[3] The present case is of left JAA, and a diminutive right auricle also exists at the right side of the heart. Therefore, this is also a partial form of JAA. JAA is a known complication of different congenital cardiac diseases with dextrocardia, great artery transposition, tricuspid valve atresia or stenosis, right ventricle hypoplasia, and ASD.[4] The communication between two appendages was a rare anomaly, shown in a few reports.[5] In this case, tunnel-like forms with communication between two appendages existed just behind the atria, which were not detected during ASD closure. Thus, in the current operation, both sides of the ostium at the right and left atria were closed. Injecting saline from the right atrial appendage is useful in determining the left-sided ostium.

Detecting the communication between appendages using the conventional transthoracic echocardiography or computed tomography (CT) is difficult in ASD because the major shunt flow is derived from ASD. Late occurrence of residual communication between the appendages with the juxtaposition of atrial appendages after ASD closure is a rare phenomenon. Therefore, the presence or absence of the communication should be confirmed with enhanced CT or angiography when JAA occurs.

Conclusion:

Late communication between the appendages could occur in JAA after ASD closure; hence, the possibility of communication between appendages in patients with JAA should be considered, and closing it from both sides is beneficial.

Author contributions:

S.I., M.M., T.F. and K.Y. designed and performed the experiments, analyzed data and interpreted it. S.I. and K.Y. Drafted article. S.I., M.M., F.T. and K.Y. revised it critically. S.I., M.M., F.T. and K.Y. approved of the article, collected data and supported technical and logistical.

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Figure 1: Enhanced computed tomography showing a left juxtaposition of the atrial appendages. (A) The right atrial appendage is not observed at the rightful site and a little right auricle exists on the right side of the heart (yellow arrow). Left atrial appendage is in normal position (red arrow). The tunnel exists between the appendages (red asterisk). (B) The sagittal plane shows that the hole of the actual communication between appendages is 10 mm (yellow arrow). LAA, left atrial appendage

Figure 2: Intraoperative images obtained from the surgeon's perspective. (A) The ostium of the tunnel (yellow arrow) was found in the right atrium. (B) The ostium in the left atrium (blue arrow) was found during the right lateral left atriotomy.