

Right-sided juxtaposition of the left atrial appendage with orifice origin in unhabitual location: case report

Yuxtaposición a la derecha de la orejuela auricular izquierda con el orificio de origen en una localización poco habitual: reporte de caso

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Case presentation

A 4-year-old female patient, present with a history of esophageal atresia who was surgically treated in the neonatal period in another hospital, was referred to our institution at 5 months of age for a heart murmur. The diagnosis of atrial septal defect (ASD) ostium secundum was made, and medical management was started.

Her follow-up with transthoracic echocardiogram showed: an ASD and an hypoechoic circular image in close continuity with the ascending aorta on its right side (Fig. 1A). Due to the inability to identify the cause, a computed tomography (CT) angiography was requested. This revealed bronchial and abdominal situs solitus, inconclusive atrial situs with a circular and elongated image to the right of the ascending aorta (Figs. 1 B and C). The diagnostic possibility was wide and ranged from aneurysm of the Valsalva's sinus to coronary anomaly, among others. Cardiac catheterization was performed with the following findings: atrial situs solitus, right-sided juxtaposition of the left atrial appendage (LAA), (Fig. 2), normal venous and systemic returns, concordant atrioventricular and ventriculo-arterial connection, ASD ostium secundum, without good rims (by intracardiac echocardiogram), intact interventricular septum, unobstructed left aortic arch, and

closed duct. Six months later, definitive treatment was established by surgical closure of ASD, which was performed without eventualities. She was discharged 4 days later. At present, the patient is asymptomatic, hence a good long-term prognosis is expected.

The atrial appendages are structures that form part of atrial cavities. They originate from each primitive atrium during embryonic life. Normally, each atrial appendage lies to the respective side of the two great arteries¹. Juxtaposition of the atrial appendages is a rare congenital malformation in which both atrial appendages are at the same side, rather than being located to their respective side of the arterial pedicle, thus; the atrial appendages are both located to the right or left side of the great arteries. Since its first description by Birmingham in 1893, and subsequently reintroduced by Dixon in 1954², several reports have shown an association with severe congenital heart disease, particularly when the juxtaposition is left-sided, and less frequently when is right-sided³. The prevalence of this entity in clinical and anatomopathological series is variable (0.28-3.9%)⁴⁻¹⁰.

Van Praagh et al.⁴ reviewed the juxtaposition of the atrial appendages in a series of autopsies and proposed a classification focusing on the morphology of the malpositioned atrial appendages.

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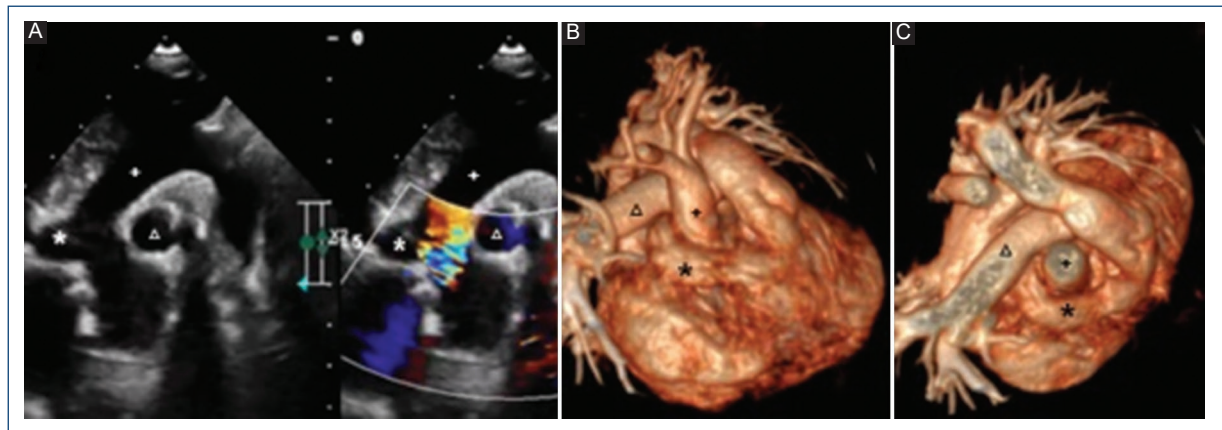


Figure 1. **A:** transthoracic echocardiogram: circular image adjacent to ascending aorta. **B** and **C:** computed tomography angiography: circular and elongated structure in an anterior position and at the right side of the ascending aorta. (*) Left atrial appendage, (+) ascending aorta, (Δ) right pulmonary artery.

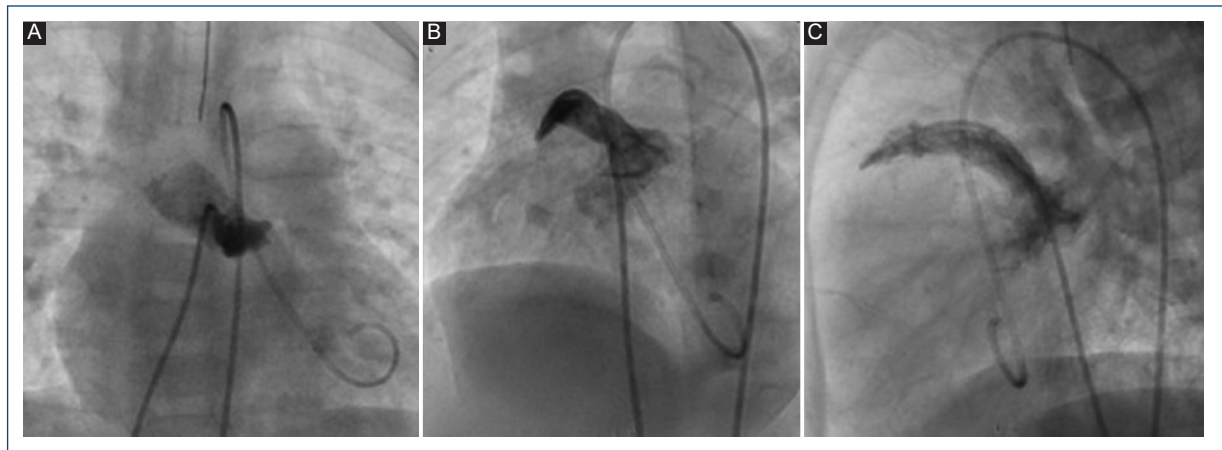


Figure 2. Angiographic imaging: right-sided juxtaposition of the left atrial appendage (we put a pigtail catheter into the left ventricle through the descending aorta with the aim to determine the position of the ascending aorta). **A:** posterior-anterior projection. **B:** 4-chamber projection and **C:** left lateral projection.

The morphology of the atrial appendages and their position are very characteristic when there is a lateralized situs; but there is the possibility of anomalies or variants in the position of the auricular appendages, a situation which is much rarer¹⁻⁴. Similarity and juxtaposition of both atrial appendages are frequently observed in hearts where the situs is isomeric¹⁻⁴.

The aim of this manuscript is to share the case of a girl with *situs solitus*, levocardia, and atrioventricular and ventriculoarterial concordance with right-side juxtaposition of the LAA with the origin of its orifice in the roof of the left atrium located posteriorly and associated to an ASD. This type of origin of the orifice has not been previously described in the literature. Moreover, this

anomaly could not be diagnosed until cardiac catheterization was performed; thus, providing us an avenue to reach the diagnosis of this rare case; however, cardiac catheterization is not available in most centers, representing a limitation for the diagnosis.

The left-side juxtaposition of the right appendage is more frequent, and the opposite juxtaposition being very uncommon³⁻¹⁰. In the present case, the atrial appendage anomaly could not be diagnosed until cardiac catheterization was performed. This difficulty in the diagnosis could be attributed to the fact that it is very uncommon (outside the cases with atrial isomerism), this is the first case of this type of atrial appendage anomaly in our hospital in the last 25 years. Our case

differs from the cases that have been previously reported, since the origin of its orifice is found in a location posterior in the roof of the left atrium; while in the literature, the habitual origin is anterior, a conception that may have led to the confusion in its diagnosis⁵⁻¹⁰.

In several reports, the diagnostic details have been made in anatomopathological studies^{1-4,6}. In 2016; Sarwark et al.⁶ described a heart with inferior and right-side juxtaposition of the LAA, a situation, which had not been previously reported, in this case, the LAA was on the floor of the left atrium, close to coronary sinus.

In a study performed in India⁵ on 129 cases of juxtaposition of atrial appendages, only 5 cases (3.9%) diagnosed with multidetector CT angiography were found to be right-sided of LAA.

Nowadays, these anomalies can be easily detected in CT angiography or magnetic resonance images^{9,10}. However, in some cases, pitfalls in the diagnosis are possible, as in the present case.

This is important in interventional procedures, such as balloon atrial septostomy or when the cutting devices are necessary, to clearly determine or diagnose the anomalies of atrial appendages, especially in the setting of left-sided juxtaposition of the right atrial appendage. This would aid the interventional personnel not to confuse the right appendage with the left atrium, since this situation is very dangerous. In the right-sided, juxtaposition of the LAA is particularly important to be aware of this anomaly in patients who need the closure of LAA, in the setting of platypnea-orthodeoxia syndrome³⁻¹⁰. This advice is also applicable in patients that need an atrial septal puncture^{3,5-10}.

The medical and surgical implications of the juxtaposed appendages have been discussed in many studies, particularly when the juxtaposed appendages are associated with a discordant ventriculoarterial connection, tricuspid atresia or both³⁻¹⁰. In the above studies, the authors reviewed hearts with juxtaposition of the atrial appendages with particular attention to the associated malformations and concentration on those characteristics that seemed to be of major surgical importance.

The right-sided juxtaposition of the LAA is a very rare anomaly. We share a case with this anomaly associated with an ASD, in which the orifice of the atrial appendage was not in its habitual location, instead, it was on the posterior part of the roof of the left atrium. This is a very rare or uncommon situation and may lead to diagnostic pitfalls in the non-invasive imaging.

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Conflicts of interest

The authors declare no conflicts of interest.

Ethical considerations

Protection of humans and animals. The authors declare that the procedures followed complied with the ethical standards of the responsible human experimentation committee and were in accordance with the World Medical Association and the Declaration of Helsinki. The procedures were authorized by the Institution's Ethics Committee.

Confidentiality, informed consent, and ethics approval. The study does not involve personal data of patients, nor does it require ethics approval. SAGER guidelines do not apply.

Declaration on the use of artificial intelligence. The authors declare that they did not use any type of generative artificial intelligence in the writing of this manuscript.

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