¹ Hospital SOS Córdio, Florianópolis, SC, Brazil.

² Universidade Federal de Santa Catarina, Florianópolis, SC, Brazil.

Occasional diagnosis of ovarian cyst during percutaneous occlusion procedure of atrial septal defect

Diagnóstico ocasional de cisto ovariano durante procedimento de oclusão percutânea de comunicação interatrial

Luiz Carlos Giuliano^{1iD}, Luis Sérgio Carvalho Luciano^{1iD}, Sthéfani Spricigo Portilho^{2iD}, Leandro Waldrich^{1iD}, José Fernando Arruda^{1iD}, Luiz Eduardo Koenig São Thiago^{1iD}

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ABSTRACT – We present a case report in which a conventional intervention for the correction of atrial septal defect resulted in an exploratory videolaparoscopy and exeresis of an ovarian cyst. Atrial septal defect is one of the primary congenital cardiopathies diagnosed in adults; ovarian cysts present a high prevalence and are frequently asymptomatic. The possible relation between atrial septal defect and ovarian cyst is debated. The femoral venous puncture techniques, guided by anatomic landmarks and by ultrasonography, are analyzed from the perspective of current scientific evidence.

Keywords: Ovarian cysts/diagnosis; Heart septal defects, atrial; Therapeutic occlusion; Videolaparoscopy

RESUMO – Apresentamos um relato de caso no qual uma intervenção convencional para correção de comunicação interatrial resultou em videolaparoscopia exploradora e exérese de cisto ovariano. A comunicação interatrial é uma das principais cardiopatias congênicas diagnosticadas em adultos; cistos ovarianos apresentam alta prevalência e são frequentemente assintomáticos. Discute-se a possível relação entre comunicação interatrial e cisto ovariano. As técnicas de punção venosa femoral guiada por reparos anatômicos e por ultrassonografia são analisadas sob a óptica das evidências científicas atuais.

Descritores: Cistos ovarianos/diagnóstico; Comunicação interatrial; Oclusão terapêutica; Videolaparoscopia

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Corresponding author:

Luis Sérgio Carvalho Luciano
Hospital SOS Córdio
Rodovia SC-401, 121 – Itacorubi
Zip code: 88030-000 – Florianópolis, SC, Brasil
E-mail: luiscl@msn.com

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INTRODUCTION

Atrial septal defect (ASD) is one of the primary congenital cardiopathies diagnosed in adults. Its late diagnosis occurs because it rarely causes significant hemodynamic repercussions in the first years of life.¹ The probability of survival until adult age is 97%. Atrial septal defect has an estimated incidence of 1.6 per thousand newborns. Additionally, women are affected twice as many times as men.²

Another frequent condition in women of child-bearing age is ovarian cyst (OC). Functional cysts account for 24% of OC, benign cysts for 70%, and malignant cysts reach 6%.³

As one of the congenital cardiopathies with greatest incidence among us, ASD can be associated with other malformations. In this report, we describe an incidental diagnosis of OC during a percutaneous occlusion procedure for ASD.

This study was evaluated by the Research Ethics Committee of the *Instituto de Cardiologia de Santa Catarina* (opinion 4.159.975). It was approved as CAAE 34842820.0.0000.0113.

CASE REPORT

A 37-year-old female patient, previously presenting with migraine, and asymptomatic from the cardiovascular point of view. Upon a routine visit to the cardiologist, she presented as New York Heart Association (NYHA) functional class I. The 12-lead

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electrocardiogram at rest showed sinus rhythm with adequate heart rate, with right bundle branch conduction disorder, and normal ventricular repolarization. An outpatient transthoracic echocardiography was requested, and an *ostium secundum* (OS) ASD was noted in addition to slight enlargement of the right ventricle. The patient was referred by the assisting cardiologist to the reference service for percutaneous occlusion of the atrial septal defect.

She was admitted electively for performance of the procedure in November 2019. The patient was submitted to general anesthesia in the cath lab, with a simultaneous transesophageal echocardiography to guide the procedure. The transesophageal echocardiography showed an atrial septum with interruption of the *fossa ovalis* membrane, a single defect measuring 10x9mm, located on the antero-superior rim of the interatrial septum, with flow directed exclusively from the left to the right. Next, the experienced operator performed a puncture directed towards the right femoral vein using the Seldinger technique, guided by anatomical landmarks. The 6F femoral introducer was positioned, and unsuccessful attempts were made to advance the 0.035" teflonized and hydrophilic guidewires towards the inferior vena cava and the right atrium. A dark liquid was aspirated through the hemostatic valve of the introducer, in an attempt to confirm its intravascular position. After a new attempt to insert the guidewires, an injection of contrast medium was made through the hemostatic valve. Contrast opacification of a large volume circumferential cavity was noted, that did not allow washing of the contrast towards the inferior vena cava (Figure 1).

Figure 1. Enhancement of the ovarian cyst in the cath lab.

The case was discussed with the general surgery team, and after the family's consent, the choice was made for exploratory videolaparoscopy within the same anesthetic procedure. The OC was identified to the right, with approximately 15cm³ of volume, with the introducer transfixing the anterior wall of the cyst (Figure 2). Right videolaparoscopic oophorectomy was performed with no complications. The patient showed good progress and was discharged from hospital on the second postoperative day.

The patient was readmitted in January 2020, for percutaneous occlusion of the OS ASD. Again, under general anesthesia, a puncture was made in the right femoral vein and unfractionated heparin was administered intravenously, with control of the activated clotting time. New measurements of ASD were made, along with the periprocedural transesophageal echocardiography and evaluation of its stretched out diameter equal to 12mm (Figure 3). The

Figure 2. Videolaparoscopic image of the introducer transfixing the anterior wall of the ovarian cyst.

Figure 3. View of the *ostium secundum* atrial septal defect on the three-dimensional transesophageal echocardiography and measured in its diameter stretched out in the two-dimensional method.

CeraFlex™ ASD Occluder 14 mm (LifeTech Scientific, China) was implanted. An echocardiography at the end of the procedure showed a well-positioned prosthesis in the interatrial septum, and absence of residual flow and of interference in the adjacent structures (Figure 4).

Figure 4. Three-dimensional transesophageal echocardiography showing a well-positioned prosthesis in the interatrial septum.

The patient was discharged from hospital on the first postoperative day, after a control transthoracic echocardiogram that showed a well-positioned prosthesis and absence of flow through it.

The transthoracic echocardiography performed 30 days after the procedure revealed findings similar to those of the previous echocardiogram. The patient continues in outpatient follow-up with a gynecologist and a cardiologist, showing no complications during this period.

DISCUSSION

The OS ASD accounts to approximately 10% to 12% of all congenital heart diseases. The diagnosis is usually suggested by the presence of a systolic murmur in a pulmonary area, and fixed splitting of the second heart sound. On the chest radiograph, a slightly enlarged cardiac area was noted, besides pulmonary hyperflow, and bulging of the pulmonary artery trunk. The electrocardiogram showed right ventricle overload.¹

Ovarian cyst can manifest as pelvic pain, dysmenorrhea, dyspareunia, menstrual disorders, infertility, or even as pressure effect on adjacent organs. It can, however, be

asymptomatic for a long period.⁴ These cysts can eventually be diagnosed inside the uterus by means of obstetric ultrasonography, like other abdominal cysts.⁵

Atrial septal defect can be related to other conditions, such as stroke, in addition to pulmonary hyperflow and hemodynamic repercussion in the right heart chambers.² Few studies have shown any relation between OC and ASD. Rizzo et al. followed up fetuses diagnosed with OC by ultrasonography and reported a case with associated congenital heart disease that evolved to death during the postnatal period.⁶

The Jarcho-Levin syndrome and Mayer-Rokitansky-Kuster-Hauser syndrome, both rare, can present with OC e ASD, albeit with other diverse findings.⁷ Additionally, besides these syndromes, the triplication of the 22q11.2 region results in cardiac malformation associated with urogenital malformations,⁸ but the data still are scarce.

Endocrine disorders of an ovarian origin are related to cardiovascular diseases.^{9,10} Furthermore, OS ASD and polycystic ovaries can be present in the severe insulin resistance syndrome.¹¹

Some authors declared there is no connection for the coexistence of OC and ASD,¹² but the prevalence of both makes simultaneous occurrence possible. In addition, there are syndromes described in which its concomitance becomes even more probably.

The femoral venous puncture guided by anatomical landmarks was amply compared to the femoral venous puncture guided by ultrasonography. The latter showed evidence of a significant increase in success rate of the procedure^{13,14} and reduced the number of attempts and the number of complications related to the puncture.¹³ Additionally, the average cost of the ultrasonography-guided procedure was lower than that of guided by anatomical landmarks.¹⁴ Both experienced operators and residents showed better results with the ultrasonography-guided method, although skilled operators experienced a more modest benefit when compared to physicians under training.¹⁵

Puncture of the femoral vein guided by ultrasonography diminishes the cost and the chance of complications, while it increases the safety and success rate of the procedure. The possibility of having OC should be taken into consideration in the differential diagnosis of complications related to femoral venous punctures.

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None.

DECLARATION OF CONFLICTS OF INTEREST

The authors declare there are no conflicts of interest.

CONTRIBUTION OF AUTHORS

Conception and design of the study: LCG and LSCL; data collection: LSCL, SSP and JFA; data interpretation: LCG and LSCL; text writing: LCG, LSCL and SSP; approval of the final version to be published: LCG, LSCL, SSP, LW, JFA and LEKST.

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