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Percutaneous occlusion of sinus venosus atrial septal defect using a covered stent: the importance of imaging methods for printing three-dimensional models and better clinical decision-making

Oclusão percutânea da comunicação interatrial do tipo seio venoso com stent coberto: a importância dos métodos de imagem para impressão de modelos tridimensionais e a melhor tomada de decisão clínica

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ABSTRACT – A superior sinus venosus atrial septal defect is usually accompanied by an anomalous venous connection to a right superior or middle pulmonary vein or veins, draining directly into the superior vena cava or even into the cavoatrial junction. This is a case report of a 62-year-old female patient, diagnosed with a sinus venosus atrial septal defect, with overload of the right chambers, for whom a percutaneous occlusion procedure was planned, using a previous cardiovascular tomography and, sequentially, a three-dimensional anatomical study, with the publicly available software 3D Slicer. In addition, a resin model was printed for inspection and simulation of a stent implantation. The patient was treated percutaneously with a 60-mm covered Chetham-Platinum stent, with total occlusion of the defect, absence of residual shunts, and draining flow from the right superior pulmonary vein to the left atrium, through a posterior communication between the atria, a *sine qua non* prerequisite to perform this type of procedure. Planning of the percutaneous occlusion procedure of the sinus venosus atrial septal defect involves careful evaluation of imaging tests. The printing of virtual or physical models, derived from computed tomography angiography of the heart, is essential for a detailed study of the defect and associated anatomical structures, minimizing the occurrence of complications.

Keywords: Heart septal defects, atrial; Vena cava, superior; Pulmonary veins; Therapeutic occlusion

RESUMO – A comunicação interatrial do tipo seio venoso superior geralmente acompanha-se de uma conexão venosa anômala de veia ou veias pulmonares superiores ou médias direitas, que drenam diretamente na veia cava superior ou, ainda, na junção cavoatrial. Relatamos o caso de uma paciente do sexo feminino, de 62 anos, com diagnóstico de comunicação interatrial do tipo seio venoso, com sobrecarga das câmaras direitas, para quem foi planejado o procedimento de oclusão percutânea do defeito por meio da realização prévia de tomografia cardiovascular e, sequencialmente, estudo anatômico tridimensional, com o software de acesso público 3D Slicer. Além disso, foi realizada a impressão do modelo em resina para inspeção e simulação de implante de um stent. A paciente foi tratada de maneira percutânea com um stent Chetham-Platinum coberto de 60mm de extensão, com oclusão total do defeito, ausência de shunts residuais e direcionamento do fluxo da veia pulmonar superior direita para o átrio esquerdo por comunicação posterior entre os átrios, condição essencial para a realização desse tipo de procedimento. O planejamento do procedimento de oclusão percutânea da comunicação interatrial do tipo seio venoso passa por avaliação criteriosa dos exames de imagem. A impressão de modelos virtuais ou físicos, derivados da angiotomografia cardíaca, é fundamental para estudo detalhado do defeito e das estruturas anatômicas associadas, minimizando a ocorrência de complicações.

Descritores: Comunicação interatrial; Veia cava superior; Veias pulmonares; Oclusão terapêutica

INTRODUCTION

The superior form of sinus venosus atrial septal defect is usually accompanied by an anomalous venous connection to a right superior or middle pulmonary vein or veins, draining directly into the superior vena cava or even into the cavoatrial junction. Deficiencies in the posterior wall separating the superior vena cava from the superior pulmonary veins allow communication between the right and left atria, causing a left-to-right shunt and volume overload of the right chambers, in addition to pulmonary hypertension.¹

The objective of this case report is to highlight the planning phases of the procedure by analysis of three-dimensional models derived from cardiac tomography, with a step-by-step description of the percutaneous closure of the sinus venosus atrial septal defect with the implantation of a covered stent.

Santa Casa de Misericórdia de Limeira evaluated and approved this study, under protocol 6.419.717, CAAE 74147323.6.0000.5608.

CASE REPORT

A 62-year-old female patient, retired teacher, with a known diagnosis of superior sinus venosus atrial septal defect. She was referred for evaluation at our service, regarding the possibility of occluding the defect using the percutaneous technique. The patient complained of fatigue and dyspnea after moderate exertion and had always refused surgical treatment.

The echocardiogram showed integrity of the interatrial septum, moderate overload of the right chambers, and indirect signs of pulmonary hypertension. There was an atrial septal defect above the superior cavoatrial junction. Computed tomography angiography revealed partial anomalous draining of the right superior pulmonary vein into the superior vena cava. The orifice was estimated at 25mm in the largest diameter (caudal) and 17mm in the smallest axis, with posterior communication between the right superior pulmonary vein and the left atrium (Figure 1, Video 1).

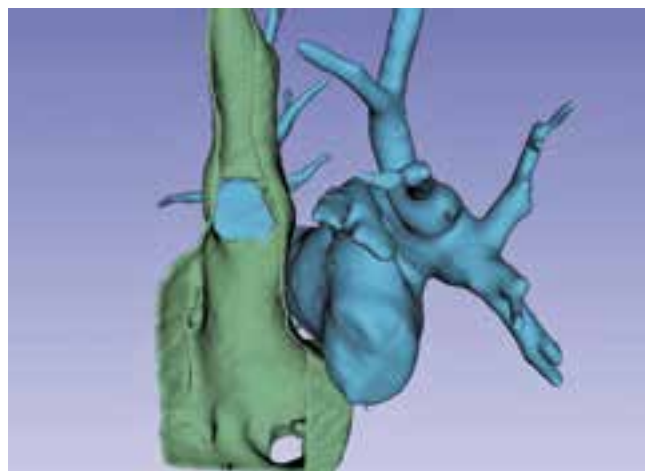
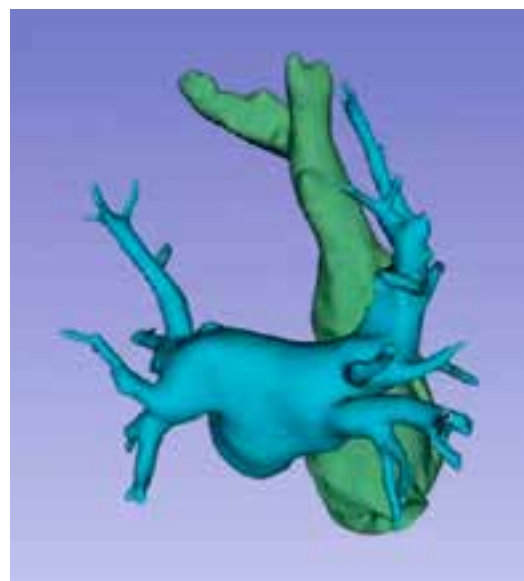


Figure 1. Three-dimensional reconstruction derived from tomography showing superior sinus venosus atrial septal defect, draining from the right superior pulmonary vein into the cavoatrial junction (central blue hole). The anterior wall of the superior vena cava was removed for a better appreciation of the defect.



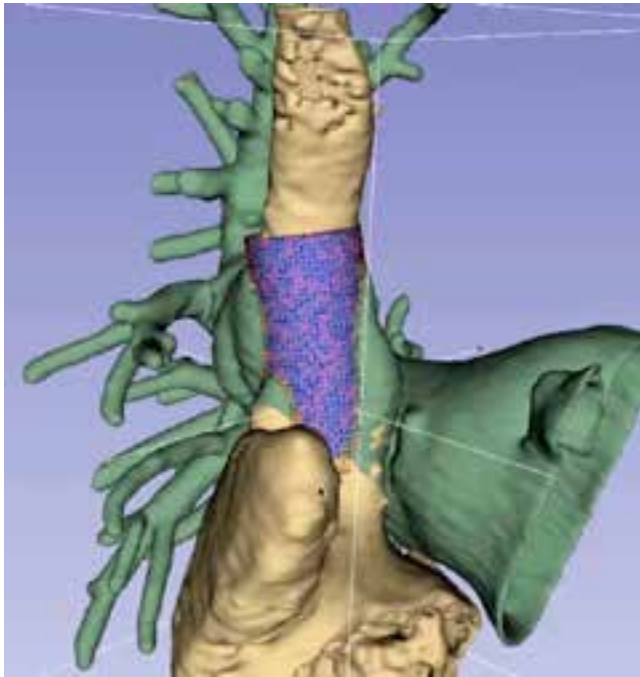
Video 1. Three-dimensional reconstruction derived from tomography with 3D Slicer software, showing superior sinus venosus atrial septal defect.

A three-dimensional anatomical study was performed using the publicly available software 3D Slicer (www.slicer.org), simulating the implantation of a 22-mm diameter and 65-mm long stent, which demonstrated the occlusion of the communication (Video 2). A resin model (Figure 2) was printed for inspection and simulation of an uncovered Cheatham-Platinum 45-mm long stent implantation (NuMed, Hopkinton, New York, United States). As the stent was expanded, a craniocaudal shortening of the stent and an inadequate coverage of the ostium of the anomalous draining of the right superior pulmonary vein into the superior vena cava were observed.

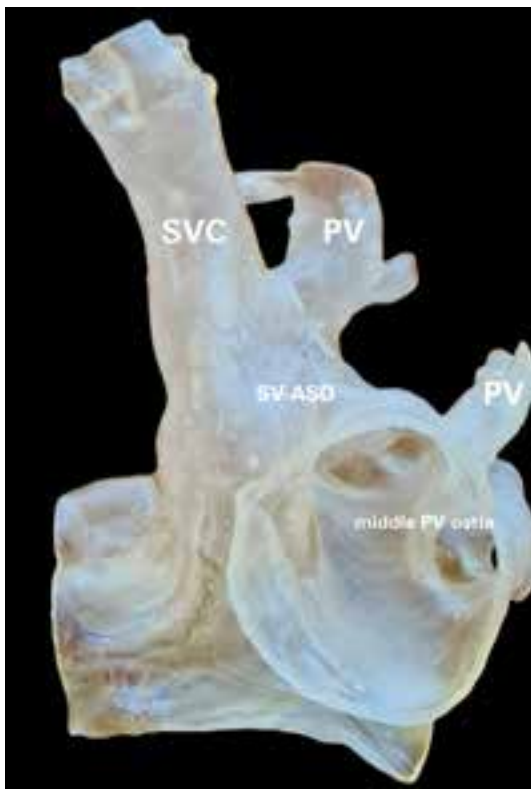
The procedure was performed under general anesthesia, with orotracheal intubation, fluoroscopic monitoring and three-dimensional transesophageal echocardiography. Intravenous prophylactic antibiotic therapy was performed with cefazolin 2.0g. We used a long hydrophilic 20F 65-cm long DrySeal sheath (W.L. Gore and Associates, Flagstaff, Arizona, United States) positioned in the inferior vena cava. The right common jugular vein was also punctured with a 7F sheath for angiographic control.

A 0.035"x260cm Amplatz Super Stiff™ guidewire was positioned in the left subclavian vein, and then a 5F MPA2 catheter was positioned in the right superior pulmonary vein, through a left common femoral vein puncture. The occlusion test of the superior vena cava at the level of the anomalous draining was performed with a 25mmx50mm Cristal balloon (Balt Extrusion, Montmorency, France) with injection of contrast in the right superior pulmonary vein, to rule out its obstruction. The angiography showed no signs of right upper pulmonary vein obstruction.

A 5F pigtail catheter was positioned in the right pulmonary artery to mark the upper border of the anomalous draining into the superior vena cava, and to help in the correct positioning of the stent, since the transesophageal echocardiogram has poor ability to map this region.



Video 2. Virtual simulation of implantation of a 22-mm diameter and 65-mm long tubular stent, revealing the occlusion of the defect and preservation of venous draining of the right pulmonary veins into the left atrium.



SVC: superior vena cava; PV: pulmonary vein; SV-ASD: sinus venosus atrial septal defect; middle PV ostia: ostia of middle pulmonary veins.

Figure 2. Printing of a three-dimensional resin model for inspection and subsequent simulation of a 45-mm Cheatham-Platinum stent implantation.

Next, we crimped a covered 60-mm long Cheatham-Platinum stent onto a balloon-in-balloon (BIB, NuMed) catheter measuring 26mm in the external diameter and 60mm in length, positioning it in the superior vena cava. The cranial position of the defect was obtained by injection of contrast in the right jugular vein, and the inferior position of the defect was obtained by transesophageal echocardiography. After verifying the correct positioning of the device, we performed the implantation with the inflation of the internal balloon, and, after adjusting its proper positioning, we inflated the external balloon. The control images showed poor apposition of the stent in its caudal portion, and an additional dilation was performed with a 30mmx50mm Cristal balloon, obtaining complete apposition of the stent with no residual flow (Figure 3).

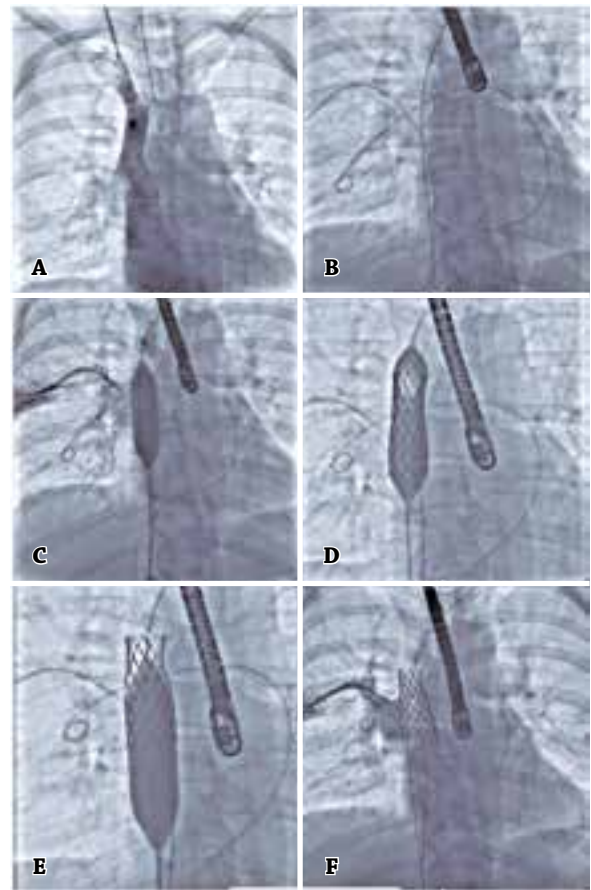


Figure 3. Procedure illustration. Implantation sequence. (A) Injection into the right jugular vein to enhance the cranial implant region. (B) MPA catheter positioned in the right upper pulmonary vein and pigtail catheter positioned in the right pulmonary artery. Guidewire positioned in the innominate vein. The defect was located where the three structures met. (C) A 25-mm balloon inflated at the level of the defect, and simultaneous injection into the right superior pulmonary vein, with free draining of the contrast agent into the left atrium. (D) Implantation of the covered Cheatham-Platinum stent at the defect level. (E) Post-dilation of the stent in its caudal portion with a 30-mm diameter balloon. (F) Result: occlusion of the defect and venous draining from the right superior pulmonary vein into the left atrium.

The patient was discharged after 24 hours. There was a subsequent intercurrent event with emergence of a small pseudoaneurysm in the left common femoral artery, which presented thrombosis after ultrasound-guided compression. A three-dimensional reconstruction model of control computed tomography angiography printed 6 months after the procedure showed the stent in an adequate position in the superior vena cava, at the right atrial outflow tract (Figure 4, Video 3).

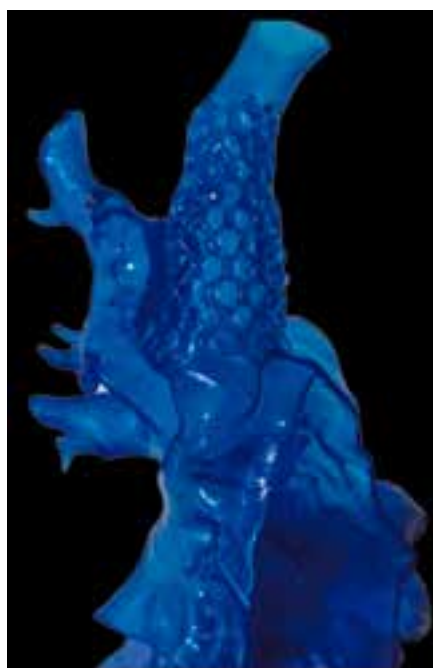


Figure 4. Control CT-derived three-dimensional model printed in resin 6 months after stent implantation.



Video 3. Final appearance of the anatomical model printed in resin.

DISCUSSION

In 2014, Abdullah et al. introduced the technique of percutaneous occlusion of a superior sinus venosus atrial septal defect,² later reproduced by Garg et al.³ and by other groups, in sequence.^{4,5} In 2020, in Brazil, Chamié et al. published a series of two patients treated with implantation of 45-mm Cheatham-Platinum covered stents. Out of four patients initially selected, two received a stent implantation with adequate occlusion of the defect. In two patients, however, after testing the balloon occlusion of the defect, there was no adequate draining flow from the right pulmonary veins into the left atrium, and the procedures were interrupted. The authors commented that longer stents were more suitable for treatment of these patients.⁶ The central idea of the procedure is to implant a PTFE-covered stent, to seal the posterior wall of the superior vena cava (which is deficient in this pathological condition) and redirect the anomalous pulmonary vein flow to the left atrium.

The printing of three-dimensional models helps in the decision-making process on the possibility of effective closure of the defect, ensuring there is no obstruction in the return of the pulmonary veins involved.⁷ Therefore, the interventional cardiologist, with the aid of a cardiac imaging specialist, can conduct a virtual evaluation of the patient's anatomy (observing the digital reconstruction of the anatomical specimen), and simulate the implantation of different caliber and length devices, or even create a print of the defect to test the implantation of stents in the model itself. In the case described in this article, it was evident that, by occluding the defect of the posterior wall of the superior vena cava, there would be a posterior defect allowing access of the right superior pulmonary vein and its branches to the left atrium, ensuring the safe execution of the procedure for the intervention team.

The planning of a percutaneous occlusion of a sinus venosus atrial septal defect involves a careful evaluation of imaging tests. Virtual or physical printed models derived from cardiac computed tomography angiography are essential for a detailed study of the defect, associated anatomical structures, and the possibility of successfully performing the procedure, minimizing complications.

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None.

CONFLICTS OF INTEREST

The authors declare there are no conflicts of interest.

CONTRIBUTION OF AUTHORS

Conception and design of the study: PTT, ABP and MOL; data collection: PTT, ABP, WPGN, JFBT, LAG and MOL; data interpretation: PTT, ABP and MOL; text writing: PTT and ABP; approval of the final version to be published: PTT, ABP, MOL, WPGN, JFBT and LAG.

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