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Case report

Tricuspid-regurgitation-mediated flow-driven right-to-left cardiac shunting caused systemic hypoxemia in a patient with patent foramen ovale without elevated right atrial pressure

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ABSTRACT

The prevalence of patent foramen ovale (PFO) is 20–25% among adults. The role of right-to-left shunting through the PFO in systemic hypoxemia remains poorly understood. Right-to-left shunting through the PFO can occur either due to elevated right atrial pressure (pressure-driven) or directed venous flow toward the PFO (flow-driven). Herein, we report a rare case of flow-driven right-to-left shunting via the PFO in a patient with traumatic tricuspid regurgitation. A 45-year-old Chinese woman was admitted due to progressive dyspnea for 3 years, presenting with cyanosis and digital clubbing. She was hypoxic, with an oxygen saturation of 83% on room air, and arterial blood gas showed an oxygen tension of 53 mmHg. Echocardiography showed severe tricuspid regurgitation with ruptured chordae tendinea, causing regurgitant jet flow directed toward the interatrial septum, leading to intermittent right-to-left shunting between the septa primum and secundum. Swan–Ganz catheterization revealed normal-high right atrial pressure and excluded pulmonary hypertension. The patient underwent tricuspid valve repair and PFO closure. Her oxygen saturation returned to 95% and her symptoms resolved. Right-to-left shunting through the PFO could cause systemic hypoxemia via a flow-driven mechanism, occasionally manifesting as cyanosis and clubbing digits. PFO closure and treatment of underlying disease are effective in improving hypoxemia.

1. Introduction

A patent foramen ovale (PFO) occurs as an embryological remnant when the foramen ovale fails to close. The foramen remains patent or, more precisely, is able to open in 20–25% of the adult population [1]. Right-to-left shunting through a PFO can occur either due to elevated right atrial pressure (pressure-driven) or directed venous flow toward the PFO (flow-driven). Although generally asymptomatic, PFO can be associated with various medical disorders, including cerebrovascular accidents, transient ischemic attacks, migraines, decompression syndrome, and systemic desaturation or hypoxemia. Recent observational studies and clinical trials have

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described the efficacy and safety of percutaneous PFO occlusion for the secondary prevention of cryptogenic stroke [2,3] and possible treatment of migraine with aura [4]. Although the connection between PFO and neurological conditions has been established, the role of PFO in systemic hypoxemia has not been adequately studied. PFO-associated hypoxemia occurs when deoxygenated venous blood from the right atrium passes across the PFO and mixes with the oxygenated blood in the left atrium. However, such conditions rely on elevated right atrial pressure (pressure-driven mechanism), which is commonly caused by pulmonary pathologies, such as chronic obstructive pulmonary disease, obstructive sleep apnea, and pulmonary hypertension. Systemic hypoxemia caused by flow-driven right-to-left shunting through the PFO has seldom been reported. Herein, we report a rare case of systemic hypoxemia caused by flow-driven right-to-left shunting through the PFO in a patient with traumatic tricuspid regurgitation (TR).

2. Case presentation

A 45-year-old Chinese woman was admitted with progressive shortness of breath for 3 years, with symptom exacerbation for 1 month (Table 1). She also presented with cyanosis and digital clubbing and was found to be hypoxic with an oxygen saturation of 83% on room air without improvement when her body position changed. The arterial blood gas showed an oxygen tension of 53 mmHg. Routine blood tests showed an elevated hemoglobin level of 179 g/L and an increased red blood cell count of 6.06×10^{12}/L. The patient reported a history of high-fall injury from the third floor, which caused blunt chest trauma and rib fracture 23 years prior. After conservative treatment with pectoral girdle fixation at a local hospital for approximately 1 week, the patient was discharged without any symptoms. No further examination or follow-up was performed, and the patient remained asymptomatic until 3 years prior to this admission.

Transthoracic echocardiography (TTE) revealed severe TR due to posterior leaflet prolapse and a dilated right heart (right atrium: supero-inferior diameter of 70 mm, septo-lateral diameter of 60 mm, right ventricle: middle right ventricular diameter of 40 mm, tricuspid annulus: anteroposterior diameter of 41 mm, and septo-lateral diameter of 42 mm). Right ventricular systolic function slightly decreased as the tricuspid annular plane systolic excursion was 18 mm. Furthermore, the presence of a PFO was suspected as a 3.5 mm-width transseptal flow was detected. Swan–Ganz catheterization showed a pulmonary artery pressure of 33/10 mmHg, pulmonary capillary wedge pressure of 11 mmHg, right atrial pressure of 9 mmHg, and right ventricle pressure of 32/6 mmHg.

The patient was scheduled for a tricuspid repair and PFO closure. Intraoperative transesophageal echocardiography (TEE) confirmed the diagnosis of severe TR with posterior leaflet prolapse (Fig. 1C), which was thought to be primarily due to her history of blunt chest trauma. The tricuspid regurgitant jet flow was directed toward the interatrial septum (Fig. 1C), leading to intermittent right-to-left shunting through the separation of the septum primum and septum secundum (Fig. 1A–B). After median sternotomy, a cardiopulmonary pass was established by the ascending aorta and bicaval cannulation. Cardioplegia was achieved, and the right atrium was opened after bicaval occlusion. Intraoperatively, the PFO was 5–7 mm in size and was closed by running suture with 4–0 Prolene. The tricuspid annulus was severely dilated and the posterior leaflet prolapsed because of the corresponding chordae tendinea rupture. One artificial chorda was implanted into the posterior leaflet, and a 30# Medtronic Contour 3D tricuspid ring was implanted for tricuspid annuloplasty. Postoperative TEE revealed trivial TR and no transseptal flow. The postoperative course was uneventful, and the patient was discharged on the seventh postoperative day with a normal oxygen saturation of 95% on room air. After 4-year follow-up, the patient presented with normal oxygen saturation and without cyanosis. The TTE showed trivial TR and no transseptal flow.

Table 1
The patients' disease course and timeline.

23 years prior	● Fall from the third floor resulted in blunt chest trauma due to fracture of the right ribs. After conservative treatment, the patient was discharged without any symptoms.
3 years prior	● Presented with progressive shortness of breath and cyanosis.
1 month prior	● Presented with severe symptoms of shortness of breath.
Day 1	● Physical examination showed cyanosis and clubbing digits. ● Transthoracic echocardiogram showed severe tricuspid regurgitation, right ventricular dilation, and transseptal flow, which led to the consideration of the presence of a patent foramen ovale (PFO). ● Arterial blood gas showed marked hypoxemia (oxygen tension of 53 mmHg).
Day 3	● Right heart catheterization showed nearly normal right atrial pressure (9 mmHg) and slightly elevated pulmonary arterial pressure (33/10 mmHg).
Day 4	● Patient underwent tricuspid repair and PFO closure. ● Intraoperative transesophageal echocardiography revealed direct tricuspid regurgitant jet flow toward the interatrial septum and intermittent right-to-left shunting through the separation of septa primum and secundum.
Postoperative day 1	● Arterial blood gas showed normal oxygen tension (92 mmHg) and SaO2 (98%)
Postoperative day 6	● Patient was discharged home with an oxygen saturation of 95 on room air.
Postoperative year 3	● Patient remained asymptomatic and transthoracic echocardiography showed mild tricuspid regurgitation, normal size of the right ventricle, and no blood shunt across the atrial septum.

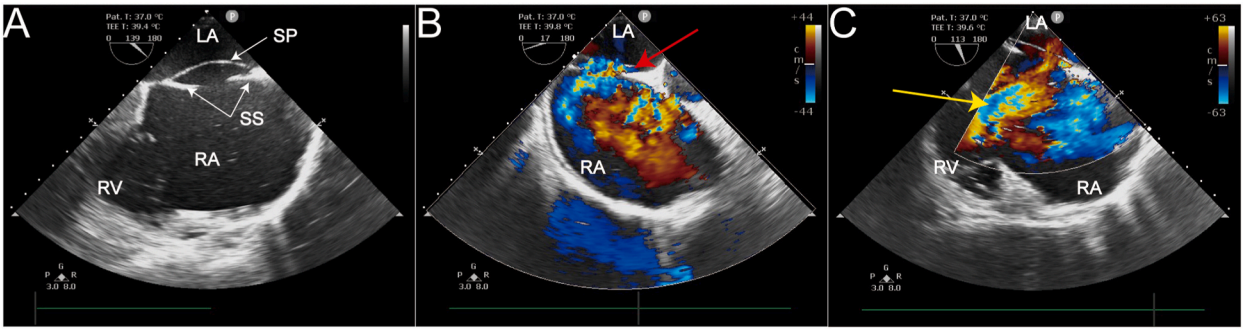

Fig. 1. Tricuspid regurgitant jet flow caused right-to-left shunting. (A) Intraoperative transesophageal echocardiography (TEE) shows large separation of the septum primum (SP) and septum secundum (SS), suggesting patent foramen ovale (PFO); (B) Doppler TEE shows right-to-left transseptal flow (red arrow) through PFO; and (C) Doppler TEE confirming tricuspid regurgitant jet flow (yellow arrow) directed toward the interatrial septum, leading to intermittent right-to-left shunting through the separation of SP and SS·
PFO, patent foramen ovale; LA, left atrium; RA, right atrium; RV, right ventricle; SP, septum primum; SS, septum secundum; TEE, transesophageal echocardiography. (For interpretation of the references to colour in this figure legend, the reader is referred to the Web version of this article.)

3. Discussion

Our study reported a rare case of PFO-associated hypoxemia presenting with cyanosis and digital clubbing induced by direct tricuspid regurgitant jet flow toward the PFO. Generally, the right-to-left shunting through PFO is considered insignificant to cause systemic circulatory changes. However, this case highlights an important point: right-to-left shunting through a PFO could cause severe systemic hypoxemia, cyanosis, and digital clubbing without elevated right atrial pressure.

PFO is a remnant of fetal circulation through which unidirectional blood flows from the right to the left atrium. Hemodynamic or anatomic changes can cause right-to-left shunting through the PFO via pressure-driven or flow-driven mechanisms. PFO-associated hypoxemia occurs when deoxygenated venous blood from the right atrium passes across the PFO and mixes with oxygenated blood in the left atrium. This phenomenon is commonly associated with pressure-driven right-to-left shunting due to pulmonary pathologies such as chronic obstructive pulmonary disease, obstructive sleep apnea, and pulmonary hypertension. Platypnea-orthodeoxia syndrome (POS), the most common PFO-associated hypoxemia without elevated pulmonary pressure, is characterized by dyspnea and objective evidence of hypoxemia in the upright position that resolves in the supine position [5]. One of the proposed mechanisms for POS is interatrial septal distortion and a change in the septal relationship to the inferior vena cava (IVC), streaming IVC blood toward the PFO or septal defect [6,7]. Moreover, the restricted motion of the anterior leaflet and an anteriorly-directed jet across the PFO has been reported as a mechanism for flow-driven right-to-left shunting in cases of carcinoid tricuspid disorder [8].

In the present case, an injury associated with a fall from a height 23 years ago was considered the primary cause of tricuspid chorda tendinea rupture and traumatic TR. The tricuspid regurgitant jet flow was toward the fossa ovalis, which led to the intermittent right-to-left shunting through the separation of septum primum and septum secundum. Hence, after a more than 20-year period of hypoxemia, the patient developed symptoms of systemic hypoxemia, including cyanosis and digital clubbing. To the best of our knowledge, such a shunting mechanism has never been reported in patients with severe systemic hypoxemia.

Percutaneous PFO closure has been proven effective for cryptogenic stroke [3], POS [5] and shunt-induced cyanosis [9]. Based on this case study, we recommend surgical PFO closure in patients with surgical indications for underlying heart disease.

In conclusion, although rare, flow-driven right-to-left shunting through a PFO can cause severe sequelae, such as systemic hypoxemia. Understanding the mechanism of intracardiac shunting is vital for its accurate diagnosis and treatment. PFO closure and treatment of the underlying disease are effective in improving hypoxemia.

Declarations

Author contribution statement

SJW and GLL: conceptualization. PT and HGZ: writing, review, and surgery. SY: echocardiographic consultation.

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Data availability statement

Data included in article/supplementary material/referenced in article.

Institutional review board statement

Ethical approval for the study was waived due to the retrospective nature of this study.

Informed consent statement

Written informed consent was obtained from the participant for the publication of this case report. Written informed consent was obtained from the individuals for the publication of any potentially identifiable images or data included in this article.

Declaration of competing interest

The authors declare that they have no known competing financial interests or personal relationships that could have appeared to influence the work reported in this paper.

Appendix A. Supplementary data

Supplementary data to this article can be found online at https://doi.org/10.1016/j.heliyon.2023.e13556.

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