Review Article

Suicidal right ventricle in children and adults: Trends, triggers, and treatment: A systematic review of a rare but catastrophic event

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Abstract

Background: Suicidal right ventricle (SRV) is defined as a dynamic right ventricular outflow obstruction with a clinical hemodynamic significance. This is a rare and dangerous event in cardiology practice. **Objectives:** This systematic review is aimed at ascertaining different scenarios that can precipitate SRV in cardiology practice. **Methods:** Data for the systematic review was retrieved from Cochrane database, Google Scholar, PubMed review, Institute for Scientific Information "Web of Science," and Medline. The search engine yielded 164 items on SRV. 148 articles were excluded thereafter because they did not meet the criteria for SRV with 16 articles left. The remaining 16 articles fulfilled the inclusion criteria and were further assessed for eligibility and 9 were excluded further, because they did not strictly fulfill the criteria for SRV. Finally, 7 research articles were included in the systematic review and quantitative synthesis. **Results:** Valvar pulmonary stenosis is the most common diagnosis warranting balloon pulmonary valvotomy (BPV). There is no gender predilection for SRV; 7 males and 6 females. The age range is 5–28 years with a mean of 19.8 ± 6.6 at 95 CI. The most common etiology of SRV was caused by a prior supra systemic right ventricle. In one study, 108 patients had balloon pulmonary valvuloplasty, but only 0.92% (1/108) of the patients who was >5 years developed SRV. Furthermore, out of six patients in another study who had BPV, only one (16.6%) had SRV. The commonest clinical presentation of SRV is cardiovascular collapse, the SRV was relieved in some cases by removing the offending agent (catheter) while some were treated with volume expanders and Beta-blockers. **Conclusion:** This review has shown that SRV is a rare abnormality in children and adults. It normally occurs during balloon valvotomy or surgical valvotomy.

Keywords: Balloon valvotomy, suicidal right ventricle, surgical valvotomy, systematic review

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INTRODUCTION

Suicidal right ventricle (SRV) is a term used for a clinical situation where right ventricular (RV) infundibular spasm results in hemodynamic collapse following relief of pulmonary valvar stenosis either by balloon valvotomy or

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surgical valvotomy.[1] The prevalence of SRV is not known, this is because it is a rare and dangerous occurrence in cardiology practice. SRV is an uncommon anomaly in children and adults that can result in death. It normally occurs when there is a very severe RV outflow spasm or obstruction. For instance, in severe pulmonary valvar stenosis, after relief with balloon valvuloplasty, there could be a build-up of supra-systemic RV pressure and this can lead to low cardiac output.[1] This phenomenon can also occur during lung transplant for Eisenmenger syndrome.[2] SRV could occur spontaneously, especially iatrogenic causes, but it can be rare in children. It is very dangerous and very prominent when closing acyanotc heart lesions such as PDA, VSD, or ASD that has undergone Eisenmenger syndrome.[3,4]

There are very few studies on this topic with varying etiology and much is not known about how deleterious this episode can be. A careful search in the literature showed that this is the first time a systematic review on this rare event is been undertaken. The systematic review is therefore aimed at ascertaining different scenarios that can precipitate SRV in cardiology practice. We also highlighted possible complications, management, and outcome of this episodic event.

METHODS (SEARCH STRATEGY AND SELECTION CRITERIA)

The majority of the studies include case reports. Others are original articles. Data for the systematic review was retrieved from the Cochrane database, Google Scholar, PubMed review, Institute for Scientific Information "Web of Science," and Medline. The search items used were: SRV, balloon pulmonary valvotomy (BPV); surgical pulmonary valvotomy; children, adult. We also embarked on a manual search for any study that might highlight information on the topic by using references cited in original papers selected for review from March 2020 to July 2020. Criteria for selection, in this review, are any patient, whether children or adults with SRV diagnosed with angiography or echocardiography or had a surgical intervention for open-heart surgery. Articles without etiology of SRV or those that did not fulfill the criteria for the definition of SRV and incomplete data were excluded.

Search engine yielded 164 items on SRV. 148 articles were excluded thereafter because they did not meet the criteria for SRV with 16 articles left. The remaining 16 articles fulfilled the inclusion criteria and were further assessed for eligibility and 9 were excluded further, because they did not strictly fulfill the criteria for SRV.[1] Finally, 7 research articles were included in the systematic review and quantitative synthesis. PRISMA[5] flowchart summarizing the data collection process is presented in Figure 1. The highest number of studies were conducted in India.

RESULTS

Majority of the study was from 1987 to 2016 [Table 1]. Valvar pulmonary stenosis is the commonest diagnosis warranting the procedure. The most common etiology of SRV was a prior supra systemic right ventricle. In one study, 108 patients had balloon pulmonary valvuloplasty, but only 0.92% (1/108) of the patients who were above 5 years developed SRV. Also out of six patients in another study who had BPV, only one (16.6%) had SRV. There is no gender predilection for SRV; 7 males and 6 females. The age range is 5–28 years with a mean of 19.8 ± 6.6 at 95 CI. The most common clinical presentation of SRV is cardiovascular collapse, diaphoresis, restlessness, hypoxemia, and cyanosis. Other clinical features noted in the study of review are severe intravascular volume depletion, and "hypercyanotic spell." These features are sudden in onset and progressive.

The SRV was relieved in some cases by removing the offending agent (catheter) while some were treated with volume expanders and Beta-blockers. Two studies, which are original articles, documented only the mean pre- and post-procedure RV outflow tract (RVOT) pressure of all the subjects. This cannot be used since only one patient had SRV. The other two case reports showed a maximum pre-procedure RVOT pressure of 140 mmHg and a

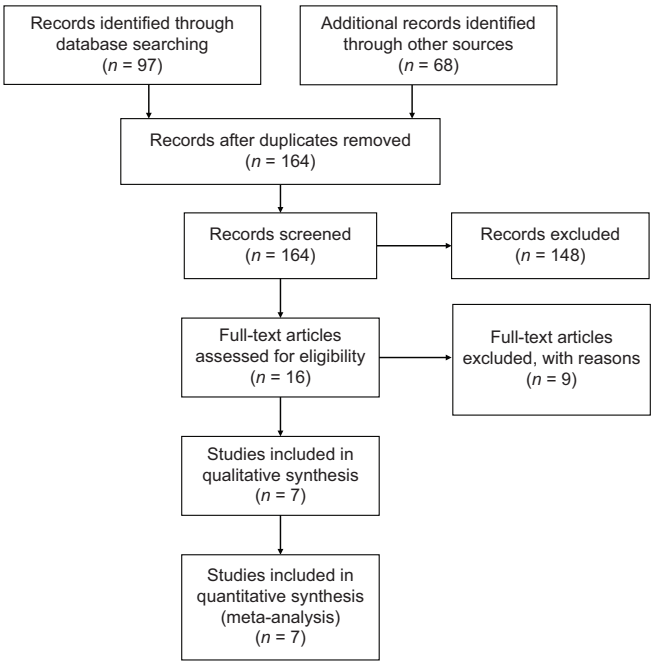

Figure 1: PRISMA[5] Flowchart summarizing data collection process

minimum postprocedure RVOT pressure of 30 mmHg. Mortality was noted in one case who was 5 years of age.

A careful search of SRV from Surgical valvotomy yielded no item except RVOT obstruction and infundibular spasm not stemming from SRV. These features alone did not fulfill the criteria for SRV. The time lag between procedure and event was not exactly documented, but some of the authors stated a sudden episodic event.

DISCUSSION

General overview

This systematic review showed a critical analysis on trends and patterns of SRV. The systematic review is the first that really examine the different scenarios and the influence of age and time on the spectrum of SRV.

The rarity of SRV, its causes, and complications appear to have a very wide variation. There is an increase in the incidence of SRV among children with balloon valvuloplasty (BPV). We could not find any case on surgical valvotomy after a thorough search.

Clinical implications of suicidal right ventricular

Series of RV infundibular spasm from SRV had resulted in hemodynamic collapse following relief of pulmonary valvar stenosis either by balloon valvotomy or surgical valvotomy.

SRV after BPV or surgical valvotomy, normally results in all the cases when the RV pressure is supra systemic before the procedure. Removal of the catheter has resulted in severe and life-threatening hypotension.[1-6] It is very pertinent to secure a good withdrawal pressure in the course of cardiac catheterization. It is important to note, in a study by Singhal *et al*.,[2] that many a times, efforts to manipulate the catheter in the RVOT when obtaining gradients or when positioning the catheter for an outflow angiogram can precipitate severe dynamic RVOT obstruction with attendant cessation of forward flow. This can cause a life-threatening hypotension.

At-risk population

From the systematic review, all ages were at risk of the suicidal ventricle; however, mortality may be more in younger ages. This is shown in our study where the mean age is mainly in older adolescents. For instance, Ezhumalai *et al*.,[7] performed BPV in 108 patients including 37 children and 71 adults. However, SRV was seen in only one patient whose is 5 years old. Unfortunately, the child expired. Other risk factors noted by Singhal *et al*.[2] are anemia and dehydration. They noted that after BPV and device closure of the VSD, a severe dynamic infundibular hypercontractility with markedly raised RV systolic pressures resulted in embolization of the device into the left ventricle.

Etiology

Several causes of SRV were noted in this review. All the authors noted pre-procedure supra-systemic RV pressure, while some authors encountered this scenario when they tried to manipulate a catheter across the RVOT during the performance of RV angiogram.[7] Furthermore, timing during BVP could also be considered as an important cause of SRV. For instance, Al Kasab *et al*.[8] had prolonged balloon inflation in one of his six patients who later developed SRV.

In addition, it has been noted that though dynamic infundibular obstruction otherwise call SRV is rare, however, it is not uncommon after BPV especially if the intravascular volume is very low.[9] However, its occurrence in catheter manipulation is very possible, especially during catheter manipulation across the RVOT. It is therefore advisable to be aware of such a possibility so that one can take timely corrective steps to avert fatal complications.[9]

Sudden RVOT obstruction, or "suicide right ventricle," was also seen in the subpopulation of patients undergoing lung transplants.[10,11] The patients had Eisenmenger's syndrome from VSD with RVOT obstruction as an initial indication for the lung transplant, both patients suffered progressive deterioration and hemodynamic instability from sudden spasm of RVOT.[10] This spasm arose from the use of probes during trans-oesophageal echocardiography. Myectomy was done to relieve this sudden spasm in the RVOT but only one patient survived. It is therefore very expedient to have a very high index of suspicion for this event during the intraoperative performance of trans-oesophageal echocardiography, as well as during direct gradient measurement.[9,10]

SRV had also been confirmed in young adult with eisenmenger's syndrome who had closure of a patent ductus arteriosus, closure of a mid-muscular VSD and peri-membranous ventricular septal defect with bilateral lung transplantation.[10] There was an episode of severe RVOT obstruction, i.e., SRV causing hemodynamic collapse The patient recovered after Extra-Corporeal Membrane Oxygenation and relief of RVOT obstruction with a RV outflow muscle resection and a patch at the outflow tract.[10,11]

Initial diagnosis

Majority of cases of the study had valvar pulmonary stenosis as the initial diagnosis. Only one subject presented

Table 1: Characeristics of patients with suicidal right ventricle

Name of author	Clinical implications of suicidal RV	At risk population	Aetiology	Geographic distribution	Initial diagnosis	Possible interventions available	The time lag between procedure and event
Ezhumalai *et al*. 2016	Sudden death from severe shock and cardiac arrest	Pre- and post- procedure RVOT pressure not documented for the index patient, only mean of the total cases was documented	There was one mortality in a child (5 years of age) because of development of suicide RV secondary to balloon dilatation of PV	India	Pulmonary valvar stenosis	Dead	Not exactly stated. "Sudden" was used to describe interval between procedure and event
Singhi *et al*. 2015	Severe hypotension Preprocedure RVOT pressure=140 mmHg Postprocedure RVOT pressure=20 mmHg	Pre-BVP supra-systemic RVP, anaemia and dehydration	Dynamic infundibular hypercontractility leading to embolization of the device into the left ventricle	India	VSD+PS	Device retrieval, volume expansion and beta blockade	Not exactly stated. "few hours" was used to describe interval between procedure and event
Singhal *et al*. 2015	Complete cessation of forward flow leading to life threatening hypotension, diaphoresis and poor respiratory effort Preprocedure RVOT pressure: 130–140 mmHg Postprocedure RVOT pressure=30–50mmHg	Supra-systemic RV pressures	Manipulation of the catheter across the RVOT	India	Severe valvar and sub- valvar PS	The catheter was instantaneously withdrawn	Not exactly stated. "Sudden" was used to describe interval between procedure and event
Hala *et al*. 2012	Worsening low cardiac output and small volume peripheral pulses	Supra-systemic right ventricle	Supra-systemic right ventricular pressure after successful valvuloplasty	Egypt	Severe pulmonary valvar stenosis	Hypotension was treated with inotropes	Not stated
Al Kasab *et al*. 1987	1 out of six patient had severe infundibular spasm, cyanosis or poor perfusion	Pre- and post- procedure RVOT pressure not documented. only mean of the total cases was documented	Prolonged balloon inflation	India	Pulmonary valvar stenosis	Propranolol 20 mgs 3 times a day for 3 days	Not stated
Kroshus *et al*. 1995	Progressive hypovolemia unresponsive to aggressive medical therapy and eventual demise of one of the two patients	Preprocedure supra-systemic RVP (100 mmHg)	Lung transplantation		Eisenmenger syndrome from VSD	Surgical vulvotomy and myectomy	Not stated
Shivanand *et al*. 2018	Severe shock		Lung transplantation		Eisenmenger syndrome from VSD and PDA	Central veno-arterial extra-corporeal membrane oxygenation. surgical vulvotomy and myectomy	

RVOT–Right ventricular outflow tract; RV–Right ventricular; PDA–Patent ductus arteriosus; VSD–Ventricular septal defect; RVP–Right ventricular pressure; BVP–Balloon Valvuloplasty, PS–Pulmonary stenosis

with a muscular VSD and pulmonary stenosis and two with Eisenmenger's syndrome following VSD and PDA.

Time lag between procedure and event

The time lag between procedure and event was not exactly

documented, but some of the authors stated that SVR was sudden and episodic. This is very important as it will help the clinician to rather identify people at risk, especially those with supra-systemic RV pressure and manage them properly before any procedure.

Intervention

Device retrieval, the use of inotropes as volume expander, and beta-blockade were used to relieve these events. Administration of oral beta-blockers can cause relief of infundibular spasm. Fluid administration combined with intravenous beta-blockers or calcium channel blockers may be helpful in treating this condition.

Singhal *et al*.[2] reported an 18-year-old female with severe valvar and sub-valvar PS with supra-systemic right ventricle. In an attempt to manipulate a catheter across the RVOT during an RV angiogram, he noted sudden hypotension in a diaphoretic patient with poor respiratory effort. It was noticed that catheter manipulation had triggered SRV. The catheter was immediately withdrawn with rapid return of systemic pressure to normal and clinical recovery.

CONCLUSION

This review has shown that SRV is a rare abnormality in children and adult. It normally occurs during BPV or surgical relief of valvar obstruction. This usually occurs in patient who had supra-systemic RV pressure.

Recommendation

A high index of suspicion is required when performing BPV or surgical relief of RV outflow obstruction, especially for patients with supra-systemic RV pressure. The use of beta-blocker could be helpful before such procedures. This could cause a relaxation of the RVOT spasm.

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Conflicts of interest

There are no conflicts of interest.

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