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Ascending Aortic Stiffness And Dilatation In Repaired Tetralogy of Fallot: A Systematic Review

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ADMINISTRATIVE INFORMATION

Support - None.

Review Stage at time of this submission - Data analysis.

Conflicts of interest - None declared.

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Amendments - This protocol was registered with the International Platform of Registered Systematic Review and Meta-Analysis Protocols (INPLASY) on 11 April 2024 and was last updated on 11 April 2024.

INTRODUCTION

Review question / Objective Is ascending aortic arterial stiffness associated with ascending aortic dilatation in patients with repaired Tetralogy of Fallot?

Rationale The prevalence of AscAo dilatation in adults late after repaired TOF (rTOF) has been estimated to range between 15-50%. Aortic aneurysm formation in TOF might lead to potentially debilitating complications such as acute aortic syndrome or aortic regurgitation, which is also associated with late aortic dilatation.
Histological analysis of dilated aortas in rTOF has highlighted degeneration of the tunica media of the AscAo, including fibrosis, (cystic) medial necrosis, fragmentation and loss of elastic fibers, and loss of smooth muscle cells, resembling the findings in degenerative aortic aneurysms. Considering the presentation of rTOF related aortic aneurysms at an early age as compared to degenerative aortic aneurysms, the importance of aortic stiffness in the pathophysiology of this disease is unclear.

Condition being studied Tetralogy of Fallot (TOF) is the most prevalent cyanotic congenital heart disease. It is characterized by pulmonary valve stenosis, ventricular septal defect, overriding aorta and right ventricular hypertrophy. TOF patients may suffer from difficulty of breathing, low oxygen levels, poor weight gain, and fatigue, yet may even be asymptomatic. In the first decades of congenital cardiac surgery, palliative surgery using the Blalock-Taussig shunt was preferred before a complete repair because of the conditional instability in prematurely born infants. For the last four decades, a single-stage complete repair to treat TOF in infancy has been preferred over palliation or late repair. Complete repair has excellent outcomes if performed within the first year of life.

METHODS

Search strategy Two independent authors identified performed a PubMed/MEDLINE database search, updated as recently as April 11th, 2024 (end-search date). The literature search was performed using the following search query:

"("Aortic Diseases"[Majr]) OR "Aorta"[Majr] OR "Aortic Aneurysm"[Majr]) AND ("Vascular Resistance"[Mesh:NoExp] OR "Vascular Stiffness"[Mesh:NoExp] OR "Elasticity"[Mesh:NoExp]) AND "Tetralogy of Fallot"[Majr] ". Filters and limiters were not applied. Search in COCHRANE and EMBASE databases did not yield any articles.

Participant or population Repaired Tetralogy of Fallot patients.

Intervention Techniques measuring ascending aortic stiffness and ascending aortic dimension.

Comparator Patients without cyanotic congenital heart disease or healthy individuals.

Study designs to be included Observational studies.

Eligibility criteria Exclusion criteria were studies investigating aortic dilatation and arterial stiffness prior to surgical TOF repair, and studies that included pregnant participants.

Information sources Two independent authors identified performed a PubMed/MEDLINE database search. Search in COCHRANE and EMBASE databases did not yield any articles.

Main outcome(s) One of the main outcomes was ascending aortic dimension, which could be measured using any technique. Furthermore, any method of estimating arterial stiffness was accepted, including distensibility, pulse wave velocity (PWV), arterial impedance, aortic relative area change (RAC), compliance and aortic strain.

Quality assessment / Risk of bias analysis Risk of bias analysis was performed using Cochrane's Risk Of Bias In Non-randomized studies - of Interventions (ROBINS-I).

Strategy of data synthesis The focus of this systematic review was on aortic stiffness and aortic dilatation in rTOF patients. Two researchers screened titles and abstracts independently from each other for eligibility. Original prospective human clinical studies, published in English, reporting on patients with rTOF (without specifically including for subtypes as pulmonary atresia, double outlet right ventricle, (subaortic) ventricular septal defect etc.), examination of dimension as well as arterial stiffness in the AscAo were included. Any method of estimating arterial stiffness was accepted, including distensibility, pulse wave velocity (PWV), arterial impedance, aortic relative area change (RAC), compliance and aortic strain. Exclusion criteria were studies investigating aortic dilatation and arterial stiffness prior to surgical TOF repair, and studies that included pregnant participants. The selected articles were then analyzed. Risk of bias analysis was performed using Cochrane's Risk Of Bias In Non-randomized studies - of Interventions (ROBINS-I).
Various indices were used to determine aortic stiffness. The greatest dimensions of the ascending aorta reported in the included articles were retrieved. The ascending aorta was defined as the aortic annulus extending to the first branching. A pooled analysis was not performed due to heterogeneity of the studies.

Subgroup analysis No subgroup analysis was performed.

Sensitivity analysis The selected articles were then analyzed. Risk of bias analysis was performed using Cochrane's Risk Of Bias In Non-randomized studies - of Interventions (ROBINS-I). A pooled analysis was not performed due to heterogeneity of the studies.

Country(ies) involved Netherlands.

Keywords Tetralogy of Fallot, arterial stiffness, ascending aortic aneurysm, ascending aortic dilatation, congenital heart disease.

Dissemination plans After completion of data analysis and writing, this systematic review will be submitter to a professional scientific journal.

Contributions of each author

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