Received: 25 January 2022 | Accepted: 22 February 2022

DOI: 10.1002/ppul.25869

ORIGINAL ARTICLE

WILEY

Bronchoscopy in the management of children from developing countries undergoing congenital heart surgery

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Abstract

Objectives: To study the clinical characteristics and impact of bronchoscopy in children from developing countries, referred for cardiac surgery, through the "Save a Child's Heart" (SACH) organization.

Methods: We performed a retrospective hospital-chart review of SACH children (0–18 years old) referred between 2006 and 2021 who underwent fiberoptic bronchoscopy. We examined demographics, congenital-heart-disease (CHD) types, bronchoscopy's indications and findings, subsequent recommendations, number of ventilation, and intensive-care-unit days. The primary outcome was percent changes in management and diagnosis, following the bronchoscopy. We included a control group matched-for-age and CHD type, who did not undergo bronchoscopy.

Results: We performed 82 bronchoscopies in 68 children: 18 (26.5%) preoperatively; 46 (67.6%) postoperatively; and four (5.9%) both. The most prevalent CHDs were Tetralogy-of-Fallot (27.9%) and ventricular-septal-defect (19.1%). The main indications were persistent atelectasis (41%) and mechanical ventilation/weaning difficulties (27.9%). Bronchoscopic evaluations revealed at least one abnormality in 51/68 (75%) children. The most common findings were external airway compression (23.5%), bronchomalacia (19.1%), and mucus secretions (14.7%). Changes in management were made in 35 (51.4%) cases, with a major change made in 14/35 (40%) children. Compared to the control group, the children undergoing bronchoscopy were both ventilated longer (median 6 vs. 1.5 days, $p < 0.0001$) and stayed longer in the intensive care unit (median 1.5 vs. 18.5 days, $p < 0.0001$).

Conclusion: A bronchoscopy is an important tool in the diagnosis and management of the unique group of children from developing countries with CHD referred for cardiac surgery. The results of our study, reveal a more complicated clinical course in children requiring bronchoscopy compared to controls.

KEYWORDS

bronchoscopy, congenital malformations, critical care, International Health

Michael Schnapper and Ilan Dalal contributed equally to this study.

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1 | INTRODUCTION

Congenital heart diseases (CHD) are among the most common congenital anomalies, with approximately 8.2 cases per 1000 live births.[1] Many children with CHD will require surgical interventions during infancy and childhood.[2] Congenital airway anomalies are a common concomitant finding, and a number of studies have described an estimated prevalence of 4% of CHD with such anomalies.[3–5] At least in some cases, the cardiovascular lesion itself may be responsible for the airway problems.[4] This is most commonly due to forced external compression of the major airways by the aorta, enlarged pulmonary arteries, enlargement of the heart atria, massive cardiomegaly, and major aorto-pulmonary collaterals (MAPCAs). Alternatively, the intraluminal bronchial obstruction may occasionally be caused by bronchial or lymphatic vessels.[4] Thus, it is clear that a respiratory tract anomaly in the setting of CHD is a considerable contributor to complications and unfavorable outcomes, particularly in children undergoing surgical interventions.[6–9] Indeed, a recent publication reported a significantly elevated mortality risk in children with CHD and concomitant congenital airway anomalies compared to children with CHD alone. Hence, it has been suggested that clinicians should actively look for airway anomalies in such children.[5] In this context, bronchoscopy is considered an important, safe, and feasible tool that can help diagnose, guide management, and potentially treat children with CHD with suspected airway anomalies, especially those who have a complicated perioperative course due to respiratory morbidity.[10–12]

Save a Child's Heart (SACH) is a United Nations population award recipient, non-profit organization that was established in 1996 in partnership with our institution, "E. Wolfson Medical Center," which is located in the central district of Israel. The organization focuses on bringing children from the developing world and the Palestinian authority, to Israel for treatment of congenital or acquired heart malformations, as well as investing in the education and training of foreign physicians from developing countries.[13,14] To date, SACH has treated over 5000 children from more than 50 countries, many with complex CHD. Since many of these patients undergo surgical interventions later than the acceptable standard in the developed world, and have different environmental exposures and nutritional status, they represent a unique, and so far, unexplored population.

To our knowledge, no data exist regarding the advantages of bronchoscopy in this distinctive group of children, but this information might be relevant to other humanitarian organizations or cardiac centers in developing countries that occasionally do treat such patients. Hence, our study was specifically designed to examine the impact of add-on pre and/or postoperatively bronchoscopy in the diagnosis and management of this population of children. We also compared the clinical characteristics of this particular group to a matched control group who did not undergo bronchoscopy.

2 | METHODS

2.1 | Design

We performed a retrospective chart review of all hospital records of children (0–18 years) who arrived in Israel for corrective heart surgery through SACH, and who also underwent perioperative flexible bronchoscopy between September 2006 and May 2021. We performed most of the bronchoscopies at the bedside in the cediatric intensive are unit (PICU) following the decision of a joint multidisciplinary team, including pediatric intensivists, cardiologists, pulmonologists, and cardiac surgeons. Data recorded included demographics, CHD diagnosis, and associated genetic syndromes, as well as the length of mechanical ventilation and hospitalization in the PICU. In addition, we also collected the following information related to the bronchoscopy: (a) timing of the procedures, (b) indication for the procedures, (c) bronchoscopy findings, (d) whether bronchoscopy was performed through a tracheal tube or not, (e) bronchoalveolar lavage fluid (BALF) results, and (f) management changes made following the procedures.

A control group of children from SACH who did not undergo a bronchoscopy during their perioperative period was prepared by matching age and type of cardiac defect to children in the study population. A 2:1 control to case ratio was maintained where possible, although due to the rarity of some cardiac malformations, we were not able to identify matched controls for all cases. The institutional Helsinki committee approved the study. All data remained anonymous and according to the Helsinki Committee, there was no need for informed consent due to the retrospective chart review nature of the study. When we performed more than one bronchoscopy for a child, we reported the overall major findings and any changes in management, per unique case.

2.2 | Statistical analyses

Data analysis was carried out using SPSS 27 statistical analysis software (SPSS, Inc.). A two-tailed $p < 0.05$ was considered significant. Distributions of continuous variables were assessed using the Kolmogorov–Smirnov test (cut off at $p = 0.01$). Continuous variables with approximately normal distribution were reported as mean ± standard deviation and were compared by using two-tailed paired or independent sample *t*-tests. When variables were highly skewed, we used the Mann–Whitney non-parametric *U*-test. Categorical variables were compared by using X^2 test or Fisher's exact test as appropriate.

3 | RESULTS

3.1 | Study participants

A total of 2951 children from SACH were admitted for heart surgery during the study period. We performed bronchoscopies in 70 children. Two children were excluded due to incomplete data. Hence, a total of

TABLE 1 Congenital heart disease of the study population

Cyanotic (*N* = 44)	Number	Acyanotic (*N* = 24)	Number
Tetralogy of Fallot	20	Ventricular septal defect	13
Pulmonary atresia with MAPCA's	(5)		
Absent pulmonic valve	(4)		
Double outlet right ventricle	6	Coarctation of the aorta	3
TOF type	(3)		
TGA type	(3)		
Truncus arteriosus	5	Partial anomalous pulmonary return	3 (scimitar-2)
Transposition of the great arteries	4	Others: Right aortic arch with anomalies of the left subclavian artery with PDA from the LSCA to the MPA, pulmonary artery aneurysm and stenosis, patent ductus arteriosus, atrial septal defect, pacemaker replacement.	**5**
Complete atrioventricular canal	2		
Pulmonary atresia	2		
Others: Tricuspid atresia, DORV-AVC-TGA-PS/dextrocardia -TGA-VSD-PS/ Dextrocardia-TGA-VSD-PA-ASD/atrio-ventricular discordance	5		

Abbreviations: ASD, atrial septal defect; AVC, atrio-ventricular canal; LSCA, left subclavian artery; MAPCA, major aorto-pulmonary collateral arteries; MPA, main pulmonary artery; PA, pulmonary atresia, PDA, patent ductus arteriosus; PS, pulmonic stenosis; TGA, transposition of the great arteries; TOF; tetralogy of Fallot, VSD; ventricular septal defect.

68 (2.3% out of 2951) children were included in our study. The mean age was 28 ± 40 months, 63% were males, 57% originated from the Palestinian authority, 34% from African countries and 9% from other countries. Eight children (11.7%) were diagnosed with genetic abnormalities: Trisomy 21 (*n* = 5), DiGeorge (*n* = 2), and VACTREL association (*n* = 1). The most common congenital heart defects (Table 1) were tetralogy of Fallot (TOF; *n* = 19, 27.9%), ventricular septal defect (VSD; *n* = 13, 19.1%), and double outlet right ventricle (DORV; *n* = 6, 8.8%).

3.2 | Bronchoscopy indications and findings

A total of 82 bronchoscopies were performed in 68 children (10 children needed more than one bronchoscopy). We executed the bronchoscopy postoperatively in 46 (67.6%) children, preoperatively in 18 (26.5%), and both pre- and postoperatively in four children (5.9%). Thirty children (44%) were intubated during the procedure, due to their clinical condition and not specifically for the procedure. Surgery for two children was postponed due to the preoperative bronchoscopy findings together with the complexity of the cardiac anomaly, (severe airway anomaly with bilateral bronchiectasis and severe critical extensive tracheal stenosis). One child died before surgery.

The most common indication for bronchoscopies was persistent atelectasis, which was present in 28/82 (34.1%) cases, of which 5/28 (18%) procedures were preoperative and 23/28 (82%) postoperative. This was followed by hypoxia and hypercarbia despite adequate mechanical ventilation or difficulties in weaning from mechanical ventilation in 19/82 (27.9%), and preoperative assessment due to abnormal findings in chest X-ray or CT-scan in 11/82 (16.1%, Table 2).

TABLE 2 Indications for bronchoscopy

Indications	Number total (unique cases)
Preoperative	22
Atelectasis	5
Wheezing/stridor	5
Difficult intubation	3
Others[a]	9
Postoperative	60
Atelectasis	23 (20[b])
Failed extubation	12 (11[b])
Difficult ventilation	7 (3[b])
Persistent consolidation	6
Wheezing/stridor	5 (3[b])
Recurrent unexplained pneumothorax	3
Bronchial bleeding	3
Follow up	1

[a]For suspicion of bronchiectasis (3), tuberculosis (2), abnormal imaging (2), tracheoesophageal fistula (1), para bronchogenic cyst (1).
[b]Unique cases.

Bronchoscopy revealed at least one abnormality in 51/68 (75%) children. The most common findings were external compression of

the airway (23.5%), followed by bronchomalacia (19.1%), and bronchial obstruction due to excessive mucus secretion (14.7%, Table 3). We performed broncho-alveolar lavage in 20 (24.4%) bronchoscopies, which identified infective agents in nine (45%) children, and consequently led to specific antibiotic treatments. There were no significant adverse events or complications reported in any of the bronchoscopies.

TABLE 3 Abnormal findings at bronchoscopy

Bronchoscopy findings	Number	% Patients
Abnormal findings	51	75
Bronchial obstruction with excessive secretions	10	14.7
Plastic bronchitis	(1)	
Positive cultures	(9)	
Airway anomaly		
Airway compression	16	23.5
(pulsating)	(3)	
Bronchomalacia	13	19.1
Pig bronchus	3	4.4
Tracheal granulation	2	2.9
Bronchial stenosis	4	4.4
Complete tracheal ring	3 (1—fatal)	4.4
Tracheomalacia	3	4.4
Laryngomalacia	2	2.9
Others		
Tracheoesophageal fistula	1	1.4
Para bronchogenic cyst	1	1.4
Foreign body	1	1.4

Note: More than one finding per bronchoscopy was observed in some children. We calculated the percentage from the total number of unique cases.

3.3 | Changes in management as a consequence of bronchoscopy findings

Management changes were made in 35 (51.4%) cases as a result of bronchoscopy findings (Table 4). Relatively minor changes, made in 21/35 (60%) children included: (1) specific antibiotic treatment, (2) intensive airway clearness therapy (hypertonic saline and chest physiotherapy), and (3) additional workup including chest CT or a ventilation-perfusion scan. In contrast, the other 14 out of 35 (40%) children required a major change of management: (1) change in surgical plan, (2) surgery postponement due to extensive tracheal stenosis and severe bronchiectasis, and (3) other options.

3.4 | Clinical characteristics and outcomes of the bronchoscopy group versus controls

The rarity of some CHD, prevented us from assigning an age and cardiac diagnosis matched control for every case. However, we were able to match a 2:1 control to case ratio for the majority, 47/68 (69%), of children who underwent bronchoscopy.

The bronchoscopy and the control groups were similar in age (mean 28 ± 40 vs. mean 23 ± 38 SD, $p = 0.96$), gender (male 63% and 67%, $p = 0.89$), and country of origin (34% vs. 31.8%, African origin, $p = 0.58$), respectively. However, there was a significantly longer

TABLE 4 Changes in management following bronchoscopy

Major changes ($N = 14$)	N	Minor changes ($N = 21$)	N
Change in surgical plan ($N = 6$)		Additional workup including chest CT or ventilation-perfusion scan	11
Repair of newly diagnosed vascular ring	2		
Lobectomy	2		
Aortopexy	1		
Pulmonic valve replacement	1		
Surgery postponement[a]	2	Specific antibiotic treatment	8
Tracheostomy	2	Intensive airway clearness (hypertonic saline and chest physiotherapy)	7
Subglottic granulation tissue removal	2		
Insertion of biological bronchial stenting	1		
Removal of foreign body	1		

Note: More than one minor recommendation per case was observed in some children.

[a]Postponement due to extensive critical tracheal stenosis and severe airway anomaly with bilateral bronchiectasis.

period of invasive mechanical ventilation (median = 6 days, IQR: 3–15.5 vs. median = 1.5 days IQR: 1–3, $p < 0.0001$), and longer PICU stay (median 18.5 days, IQR: 7.25–39 vs. median 1.5 days, IQR: 1–3, $p < 0.0001$), in the bronchoscopy group compared to the control group, respectively.

4 | DISCUSSION

The results of our study demonstrate that perioperative bronchoscopy plays an important role in the management of children undergoing surgery for CHD who also presented with respiratory symptoms. Indeed, we demonstrated that for those children who required bronchoscopy, the bronchoscopic findings revealed abnormal findings in 75% of the members of our unique patient population. Furthermore, the new findings prompted significant changes in the clinical management of 53% of these children. Such changes include specific antibiotic treatment, intensive airway clearance, and even major decisions such as changes in the surgery plan or surgery postponement. Our study population comprises children from developing countries referred for cardiac surgery, and as such, is unique in terms of demographic characteristics. Significantly, these children, often seek treatment at a later stage than is considered standard, both in terms of timing of diagnosis and in timing of interventions. To the best of our knowledge, no other study has investigated this specific group of children and their perioperative course.

We detected a wide range of cardiac defects in our study population, although almost half the population suffered from TOF (27.9%) or VSD (19%). Previous literature pertaining to bronchoscopy findings in children with CHD is limited. Efrati et al.[10] reported a similar rate (43%) of these two cardiac abnormalities. In contrast, other studies reported lower rates of TOF or VSD in children who underwent bronchoscopy, and specifically, 21/59 (35%) children, 22/72 (30.5%), or as low as 12/58 (20.6%).[10,15,16,17]

Atelectasis and failed extubation were the most common indications in our study for performing bronchoscopy, with vascular compressions, bronchomalacia, and mucus secretions being the most common findings. Two other studies[10,16] performed bronchoscopies for similar indications but their most common finding was vascular pressure on the bronchi and fewer cases of tracheobronchomalacia. There have been previous reports that certain airway anomalies may be associated with specific cardiac defects, such as TOF with pulmonary atresia and Major Aortopulmonary Collaterals (TOF/PA/MAPCA).[18] Interestingly, a recent study reported that when 11% of the children with TOF/PA/MAPCA referred for unifocalization surgery, underwent perioperative bronchoscopy, an extra diagnosis was made in 81% of cases. This was predominantly bronchomalacia and airway obstruction due to secretions.[19] Moreover, as in our study, this subgroup spent longer in the ICU and on invasive mechanical ventilation (IMV).

Similarly, the bronchoscopy group in our cohort spent significantly longer on IMV and in the PICU than the control group. This might be expected, since this group of children tends to have more complex abnormalities and hence require a prolonged course of recovery.

Extensive congenital tracheal stenosis was found preoperatively in three of our cases. Consequently, this led to a complicated postoperative course, which involved several surgical interventions such as tracheostomy and tracheoplasty in one patient, postponement of surgery in a second patient, and death before surgery in the third patient. In this context, a recent study by Riggs et al.[20] reported that the risk of morbidity and mortality of children undergoing CHD surgery was higher if they also had tracheal abnormalities. In their database registry of over 198,674 operations, 0.2% of children were also subjected to tracheal surgery. Despite our limited number of cases with CHD and tracheal stenosis, our results highlight the need to identify such cases preoperatively, to prepare the proper surgery plan.

Our study has several limitations. Although Lee et al.[5] reported congenital airway abnormalities in approximately 4% of children from developed countries who undergo congenital cardiac surgery, we performed bronchoscopy in only 2.4% of the children with CHD referred for surgery from developing counties. Obviously, due to the retrospective nature of our study and ethical considerations, bronchoscopy was not performed in all cases but only in specific cases as dictated by clinical questions and considerations. Specifically, a bronchoscopy was performed when the clinical findings suggested the presence of an abnormality that might affect subsequent treatment. Thus, it is possible that performing a bronchoscopy in all cases might have identified more children (possibly approaching 4%) with airway anomalies. However, we believe that these theoretically missed airways anomalies were not of critical importance to the early perioperative management of these children. In addition, this is a retrospective study with the inherent drawbacks, such as missing data. Finally, we performed 44% of the bronchoscopies in patients who were intubated and ventilated, which limits the ability to appreciate dynamic airway malformations, such as tracheomalacia and or bronchomalacia. Significantly, other studies have also reported high percentages of bronchoscopy via an endotracheal tube in complicated children, with numbers reaching 36.5%, 88.9%, and even as high as 97.5% in a cohort of children hospitalized in the PICU.[15,17,22]

5 | CONCLUSION

Our results indicate that bronchoscopy may be an important tool in the diagnosis and management of selected children from developing countries with congenital heart disease referred for cardiac surgery. Abnormal findings were revealed in 75% of our cases and caused a change in the management in 53% of the children. Children requiring bronchoscopy experienced a more complicated clinical course than matched controls.

ACKNOWLEDGMENTS

The authors gratefully acknowledge “Save A Child's Heart Organization,” and the medical staff and caregivers who are part of it.

AUTHOR CONTRIBUTIONS

Michael Schnapper: Conceptualization (lead); Data curation (lead); Formal analysis (lead); Investigation (lead); Methodology (lead); Project administration (lead); Validation (lead); Visualization (lead); Writing–original draft (lead). **Ilan Dalal**: Data curation (equal); Investigation (equal); Methodology (equal); Project administration (equal); Resources (lead); Supervision (equal); Validation (equal); Visualization (equal); Writing–review & editing (equal). **Avigdor Mandelberg**: Conceptualization (equal); Data curation (equal); Formal analysis (equal); Investigation (equal); Methodology (equal); Project administration (equal), Validation (equal); Writing–review & editing (equal). **Alona Raucher Sternfeld**: Formal analysis (equal); Investigation (equal), Methodology (equal); Writing–review & editing (equal). **Lior Sasson**: Formal analysis (equal); Investigation (equal); Methodology (equal); Writing–review & editing (equal). **Keren Armoni Domany**: Conceptualization (lead); Data curation (lead); Formal analysis (lead); Investigation (lead); Methodology (lead); Project administration (lead); Software (lead); Supervision (lead); Validation (lead); Visualization (lead); Writing–review & editing (lead).

DATA AVAILABILITY STATEMENT

The data that support the findings of this study are available from the corresponding author upon reasonable request.

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How to cite this article: Schnapper M, Dalal I, Mandelberg A, Raucher Sternfeld A, Sasson L, Armoni Domany, K. Bronchoscopy in the management of children from developing countries undergoing congenital heart surgery. *Pediatric Pulmonology*. 2022;57:1196-1201. doi:10.1002/ppul.25869