

SoC

Surgery on Children

Volume: 2

Issue: 2

Year: 2025



EDITOR-IN-CHIEF

Prof. Atilla ŞENAYLI

Department of Pediatric Surgery, Faculty of Medicine, Kırklareli University, Kırklareli, Türkiye

ASSOCIATE EDITOR-IN-CHIEF

Assist. Prof. Sevgi ULUSOY TANGÜL

Department of Pediatric Surgery, Faculty of Medicine, Kırklareli University, Kırklareli, Türkiye

EDITORIAL BOARD

Prof. Ahmet DEMİR

Department of Pediatric Oncology, Memorial Healthcare Group, Ankara, Türkiye

Asst. Prof. Amir Hossein LADAN

Department of Pediatric Surgery, Faculty of Medicine, Zanzan University of Medical Sciences, Zanzan, Iran

Spec. Ayman ELHOSNY, MD

Department of Pediatric Surgery, Windhoek Central Hospital, Windhoek, Namibia

Prof. Bülent BAKAR

Department of Neurosurgery, Faculty of Medicine, Kırıkkale University, Kırıkkale, Türkiye

Spec. Daniel MANZANO, MD

Department of Pediatric Surgery, Hospital de Especialidades de Fuerzas Armadas, Pichincha, Ecuador

Spec. Daniela LICCARDO, MD

Department of Pediatric Gastroenterology, Hepatology and Nutrition, Transplantation, Ospedale Pediatrico Bambino Gesù, Roma, Italy

Deepak Kumar GARNAIK

General Practitioner, All India Institute of Medical Sciences, Rishikesh, Uttarakhand, India

Spec. Gökhan DEMİRTAŞ, MD

Department of Pediatric Urology, Erzurum City Hospital, Erzurum, Türkiye

Spec. Günay EKBERLİ, MD

Department of Pediatric Urology, Adana City Hospital, Adana, Türkiye

Spec. Haldun UMUDUM, MD

Department of Medical Pathology, Güven Health Group, Ankara, Türkiye

Prof. Halil İbrahim UÇAR

Department of Pediatric Cardiovascular Surgery, Ankara Etlik City Hospital, Ankara, Türkiye

Prof. Haluk EMİR

Department of Pediatric Urology, Faculty of Medicine, İstanbul University, İstanbul, Türkiye

Prof. Hesham ALKADY

Department of Cardiovascular Surgery, Cairo University, Cairo, Egypt

Assoc. Prof. Hülya İPEK

Department of Pediatric Surgery, Faculty of Medicine, Hitit University, Çorum, Türkiye

Asst. Prof. İlhami JAFARLI

MRCs, FACS, FEBPS Department of Pediatric Surgery, Chelsea and Westminster Hospital, London, Imperial College London, United Kingdom

Asst. Prof. İsmail Essam ELHALABY

Department of Pediatric Surgery, Faculty of Medicine, Tanta University, Tanta, Egypt

Asst. Prof. İsmail BÜYÜKCERAN

Department Orthopedics and Traumatology, Faculty of Medicine, Ondokuz Mayıs University, Samsun, Türkiye

Spec. Mahmoud MOUSSA, MD

Department of Pediatric Surgery, Demenhur Hospital, Demenhur, Egypt

Assoc. Prof. Mehmet CANIKLIOĞLU

Department of Urology, Faculty of Medicine, Yozgat Bozok University, Yozgat, Türkiye

Assoc. Prof. Mehmet ERDOĞAN

Department of Nuclear Medicine, Suleyman Demirel University, Isparta, Türkiye

Spec. Natalia SNIGIREVA, MD

Department of Obstetrics and Gynecology, GMC Clinics, Dubai, United Arab Emirates

Spec. Nilüfer BIÇAKCI, MD

Department of Nuclear Medicine, Samsun City Hospital, Samsun, Türkiye

Spec. Oğuzhan KATAR, MD

Department of Ear Nose Throat, Faculty of Medicine, Hacettepe University, Ankara, Türkiye

Prof. Resul YILMAZ

Division of Pediatric Intensive Care, Department of Pediatrics, Faculty of Medicine, Selçuk University, Konya, Türkiye

Rezzan SAYAR, MSc

Department of Pediatric Surgery, Faculty of Medicine, Yozgat Bozok University, Yozgat, Türkiye

Serdal SÖKMEN

Anesthesia Technician, Department of Anesthesiology and Reanimation, Gülhane Training and Research Hospital, University of Health Sciences, Ankara, Türkiye



Prof. Serdar ALAN

Division of Neonatology, Department of Pediatrics, Faculty of Medicine, Kırıkkale University, Kırıkkale, Türkiye

Spec. Simay ERŞAHİN, MD

Department of Plastic, Reconstructive and Aesthetic Surgery, Bitlis State Hospital, Bitlis, Türkiye

Spec. Sushil RIJAL, MD

Department of Pediatric Surgery, University of Colorado Anschutz Medical Campus, Colorado, USA

Spec. Theodossis PAPA VRAMIDIS

Department of Endocrine Surgery, Euromedica General Hospital, Rhodes, Greece

Prof. Tutku SOYER

Department of Pediatric Surgery, Faculty of Medicine, Hacettepe University, Ankara, Türkiye

Prof. Ünal BIÇAKCI

Division of Pediatric Urology, Department of Pediatric Surgery, Faculty of Medicine, Ondokuz Mayıs University, Samsun, Türkiye

Asst. Prof. Yakup ACET

Department of Ophthalmology, Faculty of Medicine, Mardin Artuklu University, Mardin, Türkiye

Assoc. Prof. Yeşim ŞENAYLI

Department of Anesthesiology and Reanimation, Faculty of Medicine, Kırklareli University, Kırklareli, Türkiye

Asst. Prof. Yunus Emre DURMUŞ

Department of Neurosurgery, Faculty of Medicine, Ondokuz Mayıs University, Samsun, Türkiye

ENGLISH LANGUAGE EDITOR

Ahmet Baki ŞENAYLI

International Affairs and Global Politics, METU, Ankara, Türkiye

STATISTICS EDITOR

Tuncer HAZNECİ

Biostatistics Specialist, Ankara, Türkiye

Prof. Ünal ERKORKMAZ

Department of Biostatistics, Faculty of Medicine, Sakarya University, Sakarya, Türkiye

LAYOUT EDITOR

Hatice AKYIL

Biologist, MediHealth Academy Publishing, Ankara, Türkiye

ORIGINAL ARTICLES

Management of thyroglossal duct cysts: a 14-year single-center experience 38-40

Sarıkaya M, Özcan Sıkı F, Sekmenli T, Gündüz M, Yağmurlu İ, Çiftçi İ.

Surgical management of childhood empyema: a clinical review..... 41-47

Külçe EC, Aldemir H, Çelik N, Küçük Külçe G.

Maternal awareness of home accidents in children aged 0-3: a cross-sectional study..... 48-53

Tural Y, Tural E.

Comparison of outcome of preformed silo with traditionally constructed silo among neonates
with gastroschisis 54-59

Mona NJ, Hasan S, Mitul AR.

REVIEWS

Respiratory functions after lung surgeries in children: review of the literature..... 60-65

Şenaylı A, Ulusoy Tanguil S.

Anesthetic management in pediatric cardiac surgery..... 66-71

Saraçoğlu AG, Salman N.

Chest wall deformities: diagnosis, classification, and treatment approaches..... 72-78

Uğurum Yüccemen A, Yenigün BM, Çetinkanat CG.

CASE REPORT

Evaluation of Yolk sac tumor in a pediatric patient with F-18 FDG PET/CT 79-81

Silov G, Bıçakcı N, Batı F, Kırtıloğlu B.

Management of thyroglossal duct cysts: a 14-year single-center experience

 Mehmet Sarıkaya,  Fatma Özcan Sıki,  Tamer Sekmenli,  Metin Gündüz,  İsmail Yağmurlu,  İlhan Çiftci

Department of Pediatric Surgery, Faculty of Medicine, Selçuk University, Konya, Türkiye

Received: 02/01/2025

Accepted: 05/02/2025

Published: 10/04/2025

Cite this article: Sarıkaya M, Özcan Sıki F, Sekmenli T, Gündüz M, Yağmurlu İ, Çiftci İ. Management of thyroglossal duct cysts: a 14-year single-center experience. *Surg Child.* 2025;2(2):38-40.

Corresponding Author: Mehmet Sarıkaya, drmehmetsarikaya@hotmail.com

ABSTRACT

Aims: The aim of this study is twofold: firstly, to present the clinical features of thyroglossal duct cysts (TGDC) and our surgical experience; and secondly, to emphasize the importance of wide hyoid bone resection and a more thorough dissection, especially in the treatment of recurrence cases.

Methods: The present study constitutes a retrospective analysis of data about patients who underwent surgical intervention for TGDC in our clinic between the years January 2010 and April 2023. A comprehensive data set was collated for all patients, encompassing demographic characteristics, presenting complaints, the presence of fistula, preoperative imaging methods, intraoperative complications, duration of hospitalization, pathology results, and postoperative recurrence rates.

Results: The present study included a total of 51 patients. Of these patients, 26 (50.9%) were male and 25 (49.1%) were female. The mean age was calculated to be 7.8 ± 4.43 years. The mean duration of hospitalization was 35 ± 11 hours. Postoperative complications included the development of an abscess at the incision site in three patients (5.8%) and subcutaneous emphysema in one (1.9%) patient. Three (5.8%) were primary cases operated in our center, while four patients (7.8%) developed recurrence after Sistrunk procedure performed in an external center and were referred to our center.

Conclusion: In conclusion, the management of recurrent cases in TGDC surgery requires careful planning. Extended surgical procedures such as the modified Sistrunk procedure are highly successful in terms of both low recurrence rates and appropriate management of recurrence cases.

Keywords: Thyroglossal duct cyst, thyroglossal duct, recurrence, Sistrunk, modified Sistrunk

INTRODUCTION

The thyroglossal duct extends from the cecal foramen as an embryonic structure connecting the anterior two-thirds and posterior third of the tongue to the anatomical resting place of the thyroid gland. It normally undergoes involution after the 5th embryonic week; however, failure of this process may lead to the formation of a thyroglossal duct remnant or cyst (TGDC).¹ It represents the most prevalent form of congenital neck mass in children, with an estimated prevalence of approximately 7% within the general population.²

TGDC is not only an aesthetic problem, but also associated with serious complications, including severe infection, pain, swelling, local inflammation, malignancy, and fistula formation.^{3,4} The potential sites of cyst formation encompass all regions traversed by the thyroglossal duct, including the lingual, suprahyoid, thyrohyoid, and suprasternal areas. However, the most prevalent location is the midline region beneath the hyoid bone.^{5,6} The differential diagnosis of such

a cyst includes pathologies such as dermoid cyst, branchial cyst, hemangioma, and lymph node swelling.

The surgical procedure described by Walter Ellis Sistrunk in 1920 is currently accepted as the standard approach in the treatment of TGDC. The Sistrunk procedure involves the excision of the cyst, in addition to the central part of the hyoid bone and the tract extending to the foramen cecum.⁷ When performed according to the highest standards of surgical technique, the recurrence rate is less than 3% and recurrences are usually seen within the first year after surgery.⁸ Nevertheless, the probability of recurrence is known to increase in the event of inadequate bone resection or incomplete removal of the tractus.

The aim of this study is twofold: firstly, to present the clinical features of TDGC and our surgical experience; and secondly, to emphasize the importance of wide hyoid bone resection

and a more thorough dissection, especially in the treatment of recurrence cases. A further objective is to provide a discussion of our findings on the impact of this surgical approach on recurrence rates, with reference to the extant literature.

METHODS

Ethics

The study was approved by the Selçuk University Faculty of Medicine Ethics Committee (Date: 10.10.2023, Decision No: 2023/474) and was conducted by the Declaration of Helsinki. The present study constitutes a retrospective analysis of data about patients who underwent surgical intervention for TGDC in our clinic between the years January 2010 and April 2023. The study also included four patients who underwent the Sistrunk procedure in other centers but subsequently experienced recurrence and were referred to our center. The final analysis encompassed data from a total of 51 patients.

Patient Selection and Data Collection

A comprehensive data set was collated for all patients, encompassing demographic characteristics, presenting complaints, the presence of fistula, preoperative imaging methods, intraoperative complications, duration of hospitalization, pathology results, and postoperative recurrence rates. All patients underwent a modified Sistrunk procedure. Revision surgery, including extensive hyoid bone resection and more extensive dissection of the surrounding tissues, was performed in patients who had been operated on in outside centres and who developed recurrence.

Surgical Protocol

All surgical procedures were performed under general anesthesia in the supine position. The subplatysmal skin flap was excised through a transverse incision over the most prominent part of the mass. The cyst and surrounding tissues were then meticulously dissected down to the hyoid bone, and the infrahyoid muscles (mostly sternohyoid and thyrohyoid muscles) were also dissected. The central part of the hyoid bone was then resected, along with the intact tract. The suprahyoid muscles (geniohyoid and mylohyoid) were then separated from the hyoid bone, and the surrounding tissues were dissected up to the foramen cecum (**Figure**). In cases of recurrence, the surrounding tissues were meticulously removed to ensure complete eradication of the disease, due to the possibility of multiple tracts leading to recurrence or fistula formation.



Figure. Thyroglossal duct cyst surgery (Modified Sistrunk Procedure)

Statistical Analysis

Ultrasonographic imaging was performed in all patients. Magnetic resonance imaging and scintigraphy were also utilized when necessary. Complications, pathology results, hospitalization times, and recurrence rates were meticulously documented in all surgical procedures. The study population included a total of seven recurrence cases, of which three occurred among patients who underwent primary surgical procedures at our center and four among patients referred from external centers. The latter group underwent more extensive surgical procedures, and no recurrences were observed during the long-term follow-up period.

This study utilized descriptive statistical methods. Continuous variables were expressed as mean±standard deviation, while categorical variables were presented as numbers and percentages. All data were analyzed using Microsoft Excel.

RESULTS

The present study included a total of 51 patients. Demographic and clinical data of the patients are given in **Table**. Of these patients, 26 (50.9%) were male and 25 (49.1%) were female. The mean age was calculated to be 7.8±4.43 years. All patients underwent diagnostic ultrasonography; one patient (1.9%) additionally underwent magnetic resonance imaging, and one patient (1.9%) underwent scintigraphy. At the presentation, a cutaneous fistula was present in five patients (9.8%).

Table. Demographic and clinical data of patients

Parameter	Value
Number of patients	51
Gender (male/female)	26 (50.9%)/25 (49.1%)
Mean age (years)	7.8±4.43
Cutaneous fistula at presentation	5 (9.8%)
Ultrasonography performed	51 (100%)
Additional imaging (MRI/scintigraphy)	1 (1.9%)/1 (1.9%)
Our recurrent cases	3 (6.3%)
Mean follow-up period (months)	18
Mean duration of hospitalization (hours)	35±11
Postoperative complications (abscess/emphysema)	3 (5.8%)/1 (1.9%)
Malignancy/ectopic thyroid in pathology	None
MRI: Magnetic resonance imaging	

Seven patients (13.7%) underwent surgical procedures due to recurrence. Of these, three (6.3%) were primary cases operated in our center, while four patients (7.8%) developed recurrence after Sistrunk procedure performed in an external center and were referred to our center. The seven cases of recurrence underwent extensive hyoid bone resection and more extensive dissection. No recurrence was observed during a mean follow-up period of 18 months.

The mean duration of hospitalization was 35±11 hours. Postoperative complications included the development of an abscess at the incision site in three patients (5.8%) and subcutaneous emphysema in one (1.9%) patient. The patients who were operated in our clinic and reoperated due to recurrence were these three patients who developed abscesses after surgery. Both complications were treated with conservative



methods, yielding successful outcomes. Subsequent pathology reports revealed no malignancy or ectopic thyroid tissue in any patient.

DISCUSSION

This study presents a retrospective analysis of 51 patients who underwent surgery for TGDC. We evaluated the effectiveness of wide surgical dissection in the management of recurrent cases. The findings of this study indicate that the modified Sistrunk procedure has effectively reduced the incidence of recurrence. However, inadequate surgical interventions were identified as a contributing factor to recurrence, underscoring the necessity for a more comprehensive array of surgical approaches in the management of these patients.

The recurrence rate of patients who underwent primary surgery at our center was 5.8%, which is consistent with the recurrence rates reported in the literature. The recurrence rates reported in the literature vary between 3% and 10%, and this difference is generally attributed to the adequacy of surgical techniques and the extent of tissues removed during the operation.^{8,9} Inadequate bone resection or incomplete removal of the tract are considered the primary causes of recurrence, along with the presence of multiple tracts or residual cysts.^{9,10}

In the treatment of recurrence cases, a wide hyoid bone resection and more extensive dissection of the surrounding tissues were employed, yielding successful outcomes in all recurrence cases. The present study corroborates the efficacy of this method in preventing recurrence, thereby supporting the conclusions of analogous studies documented in the extant literature. For instance, Perkins et al.¹¹ reported that extended surgery led to a significant reduction in recurrence rates and yielded superior long-term outcomes in recurrent cases. It is also noteworthy that the risk of recurrence is markedly elevated in cases involving minimal resection when surgical margins are deemed inadequate.¹⁰

Strategies recommended to reduce the risk of recurrence include bone resection beyond the central part of the hyoid bone and extensive removal of surrounding tissues.⁹ In the present study, the implementation of surgical techniques by these principles resulted in no recurrence during the follow-up period. Furthermore, early revision surgery in recurrent cases plays a critical role in minimizing the risk of recurrence.

The present study employs a retrospective design, and the data are based on a single center. Conducting a prospective evaluation of recurrence cases in a larger population could offer more robust evidence regarding the efficacy of extended surgery. Standardization of pathology reports would facilitate more detailed analysis of tissues removed during surgery.

A surgical approach that included extensive hyoid bone resection and removal of surrounding tissues proved to be effective in the management of recurrent cases in this study. This finding underscores the necessity of expanding the scope of the Sistrunk procedure to encompass both primary and recurrent cases. Future studies should investigate the feasibility of this method in other centers and in larger populations and evaluate long-term outcomes.

CONCLUSION

In conclusion, the management of recurrent cases in thyroglossal duct cyst surgery requires careful planning. The employment of extended surgical approaches has emerged as a highly effective strategy to reduce the incidence of recurrence and enhance long-term outcomes.

ETHICAL DECLARATIONS

Ethics Committee Approval

The study was approved by the Selçuk University Faculty of Medicine Ethics Committee (Date: 10.10.2023, Decision No: 2023/474).

Informed Consent

Because the study was designed retrospectively, no written informed consent form was obtained from patients.

Referee Evaluation Process

Externally peer-reviewed.

Conflict of Interest Statement

The authors have no conflicts of interest to declare.

Financial Disclosure

The authors declared that this study has received no financial support.

Author Contributions

All of the authors declare that they have all participated in the design, execution, and analysis of the paper, and that they have approved the final version.

REFERENCES

1. Thompson LD. Thyroglossal duct cyst. *Ear Nose Throat J.* 2017;96(2):54-55. doi:10.1177/014556131709600204
2. Coelho A, Sousa C, Marinho AS, et al. Five years experience with outpatient thyroglossal duct cyst surgery. *Int J Pediatr Otorhinolaryngol.* 2017;100(96):65-67. doi:10.1016/j.ijporl.2017.03.002
3. Shah R, Gow K, Sobol SE. Outcome of thyroglossal duct cyst excision is independent of presenting age or symptomatology. *Int J Pediatr Otorhinolaryngol.* 2007;71(11):1731-1735. doi:10.1016/j.ijporl.2007.07.010
4. Taha A, Enodien B, Frey DM, et al. Thyroglossal duct cyst, a case report and literature review. *Diseases.* 2022;10(1):7. doi:10.3390/diseases10010007
5. Horisawa M, Niinomi N, Ito T. Anatomical reconstruction of the thyroglossal duct. *J Pediatr Surg.* 1991;26(7):766-769. doi:10.1016/0022-3468(91)90134-f
6. Mondin V, Ferlito A, Muzzi E, et al. Thyroglossal duct cyst: personal experience and literature review. *Auris Nasus Larynx.* 2008;35(1):11-25. doi:10.1016/j.anl.2007.06.001
7. Sistrunk WE. The surgical treatment of cysts of the thyroglossal tract. *Ann Surg.* 1920;71(2):121-122. doi:10.1097/0000658-192002000-00002
8. Ibrahim FF, Alnoury MK, Varma N, Daniel SJ. Surgical management outcomes of recurrent thyroglossal duct cyst in children--a systematic review. *Int J Pediatr Otorhinolaryngol.* 2015;79(6):863-867. doi:10.1016/j.ijporl.2015.03.019
9. Rohof D, Honings J, Theunisse HJ, et al. Recurrences after thyroglossal duct cyst surgery: results in 207 consecutive cases and review of the literature. *Head Neck.* 2015;37(12):1699-1704. doi:10.1002/hed.23817
10. Batazzi A, Leng S, Ghionzoli M, et al. Thyroglossal duct cyst: factors affecting cosmetic outcome and recurrence. *Pediatr Int.* 2019;61(10):1020-1024. doi:10.1111/ped.13955
11. Perkins JA, Inglis AF, Sie KC, Manning SC. Recurrent thyroglossal duct cysts: a 23-year experience and a new method for management. *Ann Otol Rhinol Laryngol.* 2006;115(11):850-856. doi:10.1177/000348940611501110

Surgical management of childhood empyema: a clinical review

 Erol Can Külçe¹,  Hakan Aldemir¹,  Neval Çelik¹,  Güney Küçük Külçe²

¹Department of Pediatric Surgery, Antalya Training and Research Hospital, Antalya, Türkiye

²Department of Child Health and Diseases, Antalya Training and Research Hospital, Antalya, Türkiye

Received: 29/01/2025

Accepted: 12/02/2025

Published: 10/04/2025

Cite this article: Külçe EC, Aldemir H, Çelik N, Küçük Külçe G. Surgical management of childhood empyema: a clinical review. *Surg Child.* 2025;2(2):41-47.

Corresponding Author: Erol Can Külçe, erolcankulce@gmail.com

ABSTRACT

Aims: In the treatment of childhood empyema, various therapeutic options are available, including tube thoracostomy followed by fibrinolytic therapy and thoracic surgical interventions. The use of fibrinolytics and video-assisted thoracoscopic surgery (VATS) has emerged as a significant innovation in managing these complications. While VATS is less invasive than thoracotomy and is considered the gold standard surgical method by some studies, there is no definitive consensus on its superiority. This study aimed to evaluate how treatment strategies evolve based on disease progression and to assess the impact of new therapeutic approaches in patients experiencing progressive stages of empyema.

Methods: This retrospective, single-center study evaluated patients under 18 years of age who underwent surgical interventions for empyema at an educational and research hospital between January 1, 2019, and January 1, 2023.

Results: A total of 20 patients with empyema in stage 2 (n=12) or stage 3 (n=8) were included in the study. These patients underwent fibrinolytic therapy and/or thoracic surgery. Patients were divided into three groups: group 1 included those treated with fibrinolytic therapy alone (n=6); group 2 included those receiving combined therapy (n=6); and group 3 consisted of those directly treated with VATS (n=8). Advanced statistical comparisons of hospital stay durations revealed significant differences between groups 1 and 2 and between groups 2 and 3 (p<0.05).

Conclusion: There is no definitive evidence to suggest the superiority of one treatment method over another for managing pleural empyema. Given the absence of standardized treatment protocols, a multidisciplinary approach involving timely and meticulous evaluation of each patient is crucial. Such an approach can improve prognosis and reduce hospitalization durations. Establishing a standardized protocol for empyema treatment could enhance response rates, shorten hospital stays, mitigate thoracic surgical risks, and facilitate earlier surgical decision-making for patients requiring thoracic intervention.

Keywords: Pleural empyema, pediatric, fibrinolytic, thoracic surgery

INTRODUCTION

Most community-acquired pneumonias resolve with medical treatment; however, approximately 0.6–2% of cases in the pediatric age group progress to empyema, with an incidence of 3.3 per 100,000 children.^{1,2} Empyema, which develops due to an imbalance in the hydrostatic and oncotic pressure in the pleural space, progresses through three stages. These stages are: stage 1 exudative, stage 2 fibrinopurulent, and stage 3 organized.^{1,3,5,6} Various treatment options are available for the management of empyema, including fibrinolytic therapy and thoracic surgical procedures following tube thoracostomy.⁷⁻¹⁰ The use of fibrinolytics and video-assisted thoracoscopic surgery (VATS) is considered an innovation in the treatment of these complications during childhood.² In stage 1 empyema, tube thoracostomy and antibiotic therapy are the first treatment options. In stage 2 empyema, favorable outcomes are achieved with fibrinolytic therapy following

tube thoracostomy. Some centers administer fibrinolytic therapy through the chest tube, while others place thinner catheters into the empyema cavity under imaging guidance. For patients who do not respond to this treatment, additional options include thoracotomy or less invasive procedures such as debridement and decortication performed with VATS. Although some studies suggest that VATS, being less invasive than thoracotomy, is the gold standard surgical method, there is no established consensus.^{7,11,12} In some patients, despite the sequential application of appropriate treatments in all stages, the empyema process persists, and progression through the stages is observed. A study was conducted on patients who experienced stage progression during empyema treatment to evaluate the parameters influencing the changes in treatment strategies according to this progression and to assess the impact of new treatment strategies.



METHODS

The study was conducted with the permission of Antalya Training and Research Hospital Clinical Researches Ethics Committee (Date: 25.05.2023, Decision No: 7/14). All procedures were carried out in accordance with the ethical rules and the principles of the Declaration of Helsinki.

In our retrospective single-center study, conducted with the approval of the ethics committee under protocol number 2023-127, patients under the age of 18 who underwent surgical intervention for empyema at Antalya Training and Research Hospital between January 1, 2019, and January 1, 2023, were evaluated. The patients' medical histories were reviewed, with a focus on past illnesses and particularly pulmonary pathologies. The presence of immunological deficiencies was also assessed. Additionally, it was determined whether the patients had received any treatment related to their existing complaints before reaching this stage. The days on which patients presented for treatment were recorded. The initial hospitalization laboratory tests were reviewed to determine whether there were any significant findings in blood biochemistry. Radiodiagnostic evaluations conducted prior to tube thoracostomy were assessed. Tube thoracostomy performed for fibrinolytic therapy was carried out routinely in accordance with the literature. The tube's localization was confirmed two hours post-procedure using a control X-ray. The characteristics of the collected fluid were analyzed, laboratory tests were performed, and the results were evaluated. On the second day, thoracic ultrasonography (USG) was used for further assessment. Patients whose treatment effectiveness remained uncertain were evaluated using high-resolution computed tomography (HRCT) of the thorax.

In the second stage, additional fibrinolytic therapy and/or thoracic surgery were planned for patients whose empyema did not resolve with tube thoracostomy. At our center, tissue plasminogen activator (t-PA) was used for fibrinolytic therapy. As per standard protocol, t-PA was administered via the previously placed chest tube at a dose of 0.1 mg/kg (maximum 6 mg/day) for three consecutive days, following guidelines in the literature.

Patients were clinically monitored, and their response to treatment was assessed using imaging methods. Fibrinolytic therapy was discontinued for all patients who showed significant improvement on X-rays and thoracic computed tomography (CT) scans, had fluid output from the chest tube of less than 5 ml/24 hours, and no longer exhibited pleural effusion.

To evaluate the impact of fibrinolytic therapy on laboratory results, hemogram and C-reactive protein (CRP) levels were recorded one day prior to the procedure, two days after the procedure, and one day before discharge.

In the third stage, thoracic surgery was directly planned for patients in whom parenchymal damage and empyema did not regress after tube thoracostomy or for those with parenchymal damage so extensive that they would not benefit from fibrinolytic therapy. This decision was made collaboratively by pediatric infectious disease and pediatric surgery clinicians, concluding that fibrinolytic therapy would not be effective. The procedure was performed under general anesthesia with the patient in the lateral decubitus position.

Using 5–10 mm trocars appropriate for the patient's age and body weight, a total of three trocars (one for the camera and two for surgical instruments) were inserted. Before concluding the procedure, chest tubes were replaced in a sterile manner through the same intercostal access (ICA), and surgical excision specimens were sent for pathological analysis. Body fluid samples were sent to microbiology, biochemistry, and pathology laboratories for further examination. For patients undergoing thoracic surgery, hemogram and CRP levels were recorded one day before the procedure, on the fifth day after the procedure, and one day before discharge. The patients' gender, age, presenting complaints, type of oxygen support, the side of the lung affected by empyema, the number of days after admission when the chest tube was removed, the presence of a bronchopulmonary fistula, the need for repeat thoracic surgery, the type and duration of antibiotic use, the presence of comorbid conditions, and the total duration of treatment were retrospectively reviewed using the hospital information system. Oxygen support was classified as follows: simple oxygen support if provided via a simple mask, nasal cannula, or reservoir mask; and advanced oxygen support if provided via high-flow oxygen or mechanical ventilation. Differences among these parameters during clinical follow-up were compared and discussed in light of the literature.

Statistical Analysis

In a 3x2 mixed design with an independent group factor (3 levels) and a repeated measurement factor (2 levels), the group*time interaction effect was set to $f = 0.90$. Using the F-test family and selecting ANOVA: repeated measures, within-between interaction, it was determined that a total of 18 patients would be required for a type I error rate (α) of 0.05 and a type II error rate (β) of 0.20. To statistically demonstrate the differences in procedures to be applied to the subjects for the research topic, account for potential data loss during the follow-up period, and achieve 80% study power, the sample size was determined to be 18 participants. The sample size was calculated using the G*power (v.3.9.1.7) software with the "as in SPSS" option activated in the options menu.

In the study, the normality of continuous variables was assessed both graphically and using the Shapiro-Wilk test. Descriptive statistics for the variables were presented as mean $\pm$ SD (standard deviation) and median (minimum-maximum) values. Cross-tables were created to compare categorical variables based on advanced treatment, with frequencies (n), percentages (%), and Chi-square test statistics reported. The Kruskal-Wallis non-parametric variance analysis was used to compare treatment methods with age, gender, and preoperative absolute neutrophil values. One-way ANOVA was employed to compare postoperative absolute neutrophil counts and length of hospital stay among treatment groups. Bonferroni correction was applied for pairwise comparisons, and the results were reported. To compare CRP levels between the group treated with fibrinolytics alone and the group treated with combined therapy, an Independent Samples t-test was used. Similarly, an independent Samples t-test was employed to compare CRP levels between the combined therapy group and the VATS-only group. To evaluate whether CRP and absolute neutrophil values showed differences at different measurement times (preoperative and postoperative), a paired samples t-test was applied for normally distributed parameters,



and the Wilcoxon signed rank test was used for non-normally distributed parameters. Statistical analyses and calculations were performed using IBM SPSS statistics 21.0 (IBM Corp. Released 2012. IBM SPSS Statistics for Windows, Version 21.0. Armonk, NY: IBM Corp.) and MS Excel 2007. A p-value of <0.05 was considered statistically significant.

RESULTS

The demographic and clinical data of the patients are presented in **Table 1**. A total of 20 patients [stage 2 (n=12) or stage 3 (n=8)] with empyema were included in the study, for whom fibrinolytic therapy and/or thoracic surgery was planned. The patients were divided into three groups: group 1 (n=6), treated with fibrinolytic therapy alone; group 2 (n=6), treated with combined therapy; and group 3 (n=8), treated directly with

VATS. During and after thoracic surgery, none of the patients developed a bronchopulmonary fistula. One patient, despite resolution of the empyema following surgery, succumbed to sepsis. None of the patients required repeat thoracic surgery.

Cough was reported in 70% of the patients (n=14), difficulty in breathing in 65% (n=13), abdominal pain in 15% (n=3), and altered consciousness in 5% (n=1).

No statistically significant differences were found between the type of treatment applied and age, gender, the side of the lung affected by empyema, or the presence of comorbidities (p>0.05).

All patients who received fibrinolytic therapy alone (n=6) required only simple oxygen support. Among patients who underwent VATS at any stage of treatment, 42.9% (n=6) required simple oxygen support, while 57.1% (n=8) required

Table 1. Comparison of demographic and clinical characteristics based on advanced treatment methods

	Advanced treatment administered				Test statistical	P
	Fibrinolytic (n=6)	Fibrinolytic+VATS (n=6)	VATS (n=8)			
	n (%)	n (%)	n (%)			
Age						
Mean±SD	5.00±3.63	6.50±3.51	8.88±5.84	*χ²=2.022	0.364	
Median (min-max)	3.0 (3-12)	5.5 (3-12)	9.5 (1-15)			
Gender						
Male	3 (50.0)	3 (50.0)	5 (62.5)	χ²=0.479	0.999	
Female	3 (50.0)	3 (50.0)	3 (37.5)			
Location						
Right	3 (50.0)	3 (50.0)	1 (12.5)	χ²=4.040	0.325	
Left	3 (50.0)	3 (50.0)	6 (75.0)			
Bilateral	0 (0.0)	0 (0.0)	1 (12.5)			
Comorbidity						
None	5 (83.3)	4 (66.6)	2 (25.0)	χ²=8.767	0.099	
CP	0 (0.0)	1 (16.7)	0 (0.0)			
Multipl comorbidity	0 (0.0)	1 (16.7)	4 (50.09)			
Other	1 (16.7)	0 (0.0)	2 (25.0)			
Type of oxygen support						
Mask	4 (66.7)	1 (16.7)	1 (12.5)	χ²=13.323	0.028	
Nasal cannula	2 (33.3)	0 (0.0)	0 (0.0)			
Reservoir mask	0 (0.0)	3 (50.0)	1 (12.5)			
High flow oxygen	0 (0.0)	1 (16.7)	3 (37.5)			
MV	0 (0.0)	1 (16.7)	3 (3.75)			
Result						
Discharge	6 (100.0)	6 (100.0)	7 (87.5)	χ²=1.584	0.999	
Exitus	0 (0.0)	0 (0.0)	1 (12.5)			
Control examination						
Normal	6 (100.0)	4 (66.7)	7 (87.5)	χ²=4.936	0.158	
Expansion defect	0 (0.0)	2 (33.3)	0 (0.0)			
Exitus	0 (0.0)	0 (0.0)	1 (12.5)			
Echocardiography						
None	4 (66.7)	3 (50.0)	3 (37.5)	χ²=3.976	0.999	
Normal	2 (33.3)	3 (50.0)	3 (37.5)			
TD	0 (0.0)	0 (0.0)	1 (12.5)			
AD+TD	0 (0.0)	0 (0.0)	1 (12.5)			

* χ^2 : Kruskal-Wallis test, χ^2 : Ki kare test. VATS: Video-assisted thoroscopic surgery, SD: Standard deviation, Min: Minimum, Max: Maximum

advanced oxygen support. A statistically significant difference was found in the type of oxygen support required based on the advanced treatment applied ($p=0.024$). However, in a multivariate logistic regression model investigating the effect of “type of oxygen support” on patients who underwent VATS, no statistically significant association was found for the need for advanced oxygen support ($p>0.05$).

No statistically significant difference was found between group 1 and group 2 in terms of pre-fibrinolytic and post-fibrinolytic CRP values ($p>0.05$). However, in group 1, a statistically significant difference was observed between the two time-dependent CRP measurements (pre-fibrinolytic and post-fibrinolytic) ($p=0.013$) (Table 2). Similarly, in group 2, a statistically significant difference was found between the time-dependent CRP measurements (pre-fibrinolytic and post-fibrinolytic) ($p=0.021$) (Figure 1).

No statistically significant difference was found between group 2 and group 3 in terms of preoperative and postoperative CRP values ($p>0.05$). However, in group 2, a statistically significant difference was observed between the two time-dependent CRP measurements (preoperative and postoperative) ($p=0.027$). Similarly, in group 3, a statistically significant difference was found between the time-dependent CRP measurements (preoperative and postoperative) ($p=0.015$) (Table 3, Figure 2).

No statistically significant difference was found in pre-procedure and post-procedure absolute neutrophil values among patients based on the type of treatment applied ($p>0.05$). However, in group 1, a statistically significant difference was observed between the two time-dependent absolute neutrophil measurements (pre-procedure and post-procedure) ($p=0.028$). Similarly, in group 2, a statistically significant difference was found between preoperative and postoperative absolute neutrophil values ($p=0.001$). In group 3, a statistically significant difference was also detected between preoperative and postoperative absolute neutrophil values ($p=0.003$) (Table 4, Figure 3).

The mean length of hospital stay was 21.50 ± 5.82 days for group 1, 37.83 ± 11.37 days for group 2, and 22.00 ± 12.06 days for group 3. A statistically significant difference was observed in the length of hospital stay among the treatment groups ($p=0.046$). Pairwise comparisons of the treatment groups revealed statistically significant differences in the length of hospital stay between group 1 and group 2, and between group 2 and group 3 ($p<0.05$).

DISCUSSION

In childhood pneumonia, the presence of comorbidities is considered one of the criteria for hospitalization, as it can negatively affect treatment success.¹³ In a study conducted by

Table 2. Time-dependent changes in CRP levels after fibrinolytic therapy between group 1 and group 2

	Group-1 (n=6)		Group-2 (n=6)		p (group)	
	Mean $\pm$ SD	Median (min-max)	Mean $\pm$ SD	Median (min-max)		
CRP						
Before fibrinolytic	216.5 $\pm$ 102.35	201.5 (117-333)	204.0 $\pm$ 101.64	172.0 (122-398)	t=0.212	0.836
End of the fibrinolytic	113.83 $\pm$ 54.49	126.0 (30-181)	101.67 $\pm$ 53.11	83.5 (48-181)	t=0.392	0.704
p (time)	t*=3.752; p=0.013		t*=3.313; p=0.021			

CRP: C-reactive protein, SD: Standard deviation, Min: Minimum, Max: Maximum

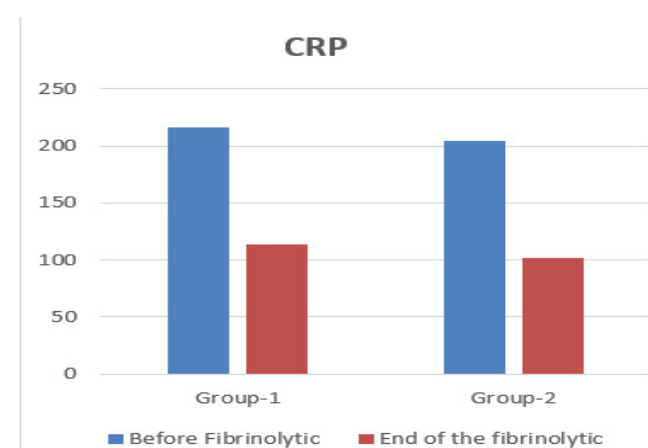


Figure 1. Distribution of CRP levels in group 1 and group 2

CRP: C-reactive protein

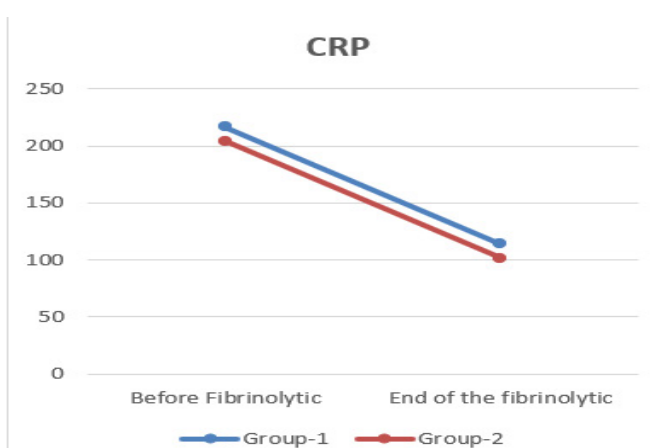


Table 3. Time-dependent changes in CRP levels after fibrinolytic application in group-2 and group-3 patient

	Group-2 (n=6)		Group-3 (n=8)		p (group)	
	Mean $\pm$ SD	Median (min-max)	Mean $\pm$ SD	Median (min-max)		
CRP						
Preop	66.00 $\pm$ 37.07	53.0 (21-126)	112.11 $\pm$ 85.38	87.5 (9.9-245)	t=1.366	0.202
Postop	48.50 $\pm$ 40.19	37.5 (12-110)	41.51 $\pm$ 37.60	30.0 (4.1-104)	t=0.334	0.744
p (time)	t*=3.085; p=0.027		t*=3.195; p=0.015			

CRP: C-reactive protein, SD: Standard deviation, Min: Minimum, Max: Maximum

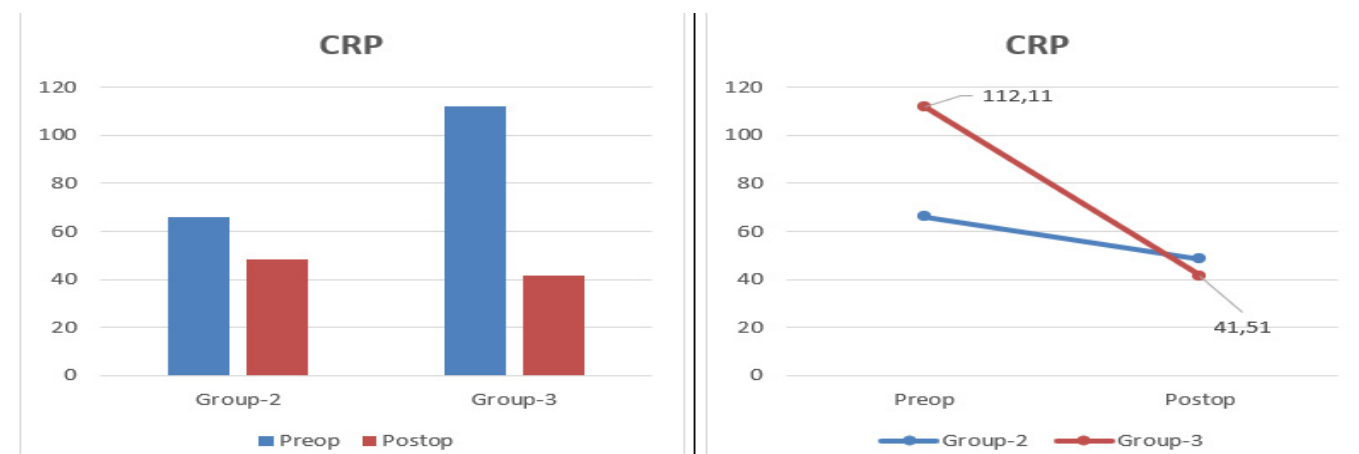


Figure 2. Group-2 and group-3 CRP distribution of values
CRP: C-reactive protein

Table 4. Time-dependent changes in absolute neutrophil values after treatment among groups

	Group-1 (n=6)	Group-2 (n=6)	Group-3 (n=8)		
	Mean±SD median (min-max)	Mean±SD median (min-max)	Mean±SD median (min-max)	p (day)	
NEU (%)					
Preop	71.83±15.85 76.5 (41-85)	68.67±20.17 77.0 (34-86)	74.94±10.82 78.5 (53-87.5)	$\chi^2=0.096$	0.953
Postop	46.67±18.82 49.5 (16-64)	48.50±23.68 53.5 (8-72)	53.00±15.27 46.0 (37-76)	F=0.207	0.815
p (time)	p=0.028	p=0.001	p=0.003		

χ^2 : Kruskal-Wallis test, F: One-Way ANOVA test, SD: Standard deviation, Min: Minimum, Max: Maximum

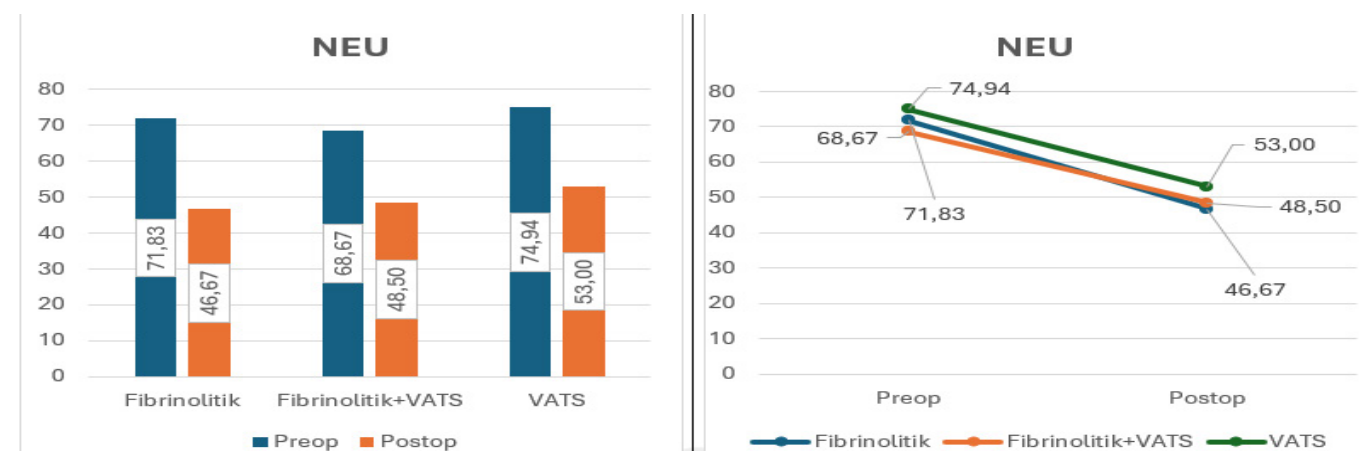


Figure 3. Distribution of absolute neutrophil values in treatment groups
VATS: Video-assisted thoracoscopic surgery

Çiftçi et al.⁷, it was observed that comorbidities in patients with empyema did not have an impact on the length of hospital stay. For patients with empyema who also have comorbidities, timely and meticulous treatment application may reduce the frequency of thoracic surgery. Conversely, in patients with empyema without comorbidities, delays in initial treatment or incorrect antibiotic choices may increase the need for thoracic surgery. Therefore, the treatment of such patients should be managed through a multidisciplinary approach.

De Benedictis et al.¹ highlighted that the World Health Organization (WHO) recommends initiating oxygen support when oxygen saturation falls below 90%, but noted existing disagreements. They argued that these disagreements stem from variations in the definition of hypoxia based on altitude. They emphasized the importance of advanced oxygen support to reduce the frequency of intubation in patients with acute respiratory distress.¹

More than half of the patients who underwent VATS at any stage of treatment required advanced oxygen support preoperatively. In contrast, patients treated with fibrinolytic therapy alone were able to maintain their oxygen saturation with simple oxygen support before the procedure, and a statistically significant relationship was found between these parameters. We believe that the completion of treatment in fibrinolytic therapy patients with simple oxygen support may indicate less lung damage compared to the other patient group, likely due to the correct treatment strategy.

There may be several reasons why patients require advanced oxygen support. One reason could be the lack of a standardized treatment protocol for childhood empyema, leading to delays or errors in treatment planning.^{7,11,12} Another possibility is the unexpected rapid clinical deterioration of the patient despite timely and appropriate treatment. We did not find any studies in the literature that specifically address these parameters.



Evaluating these parameters in studies with larger patient populations may provide more accurate results.

There is currently no established gold standard treatment strategy, either medical or surgical, for childhood empyema. In a comprehensive review, Buonsenso et al.¹⁴ noted the absence of a universally accepted treatment protocol for empyema and emphasized the importance of evaluating various surgical interventions in conjunction with appropriate antibiotic therapies. Elevated CRP levels in pneumonia patients despite treatment may indicate progression to pleural empyema.¹⁵ Studies have suggested that sequential measurements of acute phase reactants, as indicators of the inflammatory process, can be useful in assessing treatment response.^{1,12,16} Medeiros et al.¹⁷, in their study on sequential CRP measurements after surgical treatment of pleural empyema, evaluated CRP levels on the 2nd and 7th postoperative days. They observed a significant decrease in CRP by the 2nd postoperative day. While no significant difference in CRP reduction was found between patients treated with tube thoracostomy and VATS, they emphasized the utility of CRP changes in assessing hospital stay duration and treatment response.¹⁷

Ratta et al.², in their evaluation of treatment methods, found no superiority among methods in reducing laboratory parameters but highlighted that pleural debridement via VATS was significantly more effective. To evaluate surgical treatment response, we analyzed laboratory values one day before the procedure, on the 2nd postoperative day, and before discharge. In all cases, CRP and absolute neutrophil values showed significant decreases. By assessing the reduction in CRP and absolute neutrophil percentages at discharge and on the 14th day post-discharge, we concluded that all three treatment methods effectively reduced these parameters with no significant superiority among them.

Griffith et al.¹⁰ compared fibrinolytic therapy, VATS, and thoracotomy and found that VATS reduced both antibiotic use and hospital stay duration. Barglik et al.³, in their study comparing hospital stays of patients treated with fibrinolytic therapy versus direct VATS, concluded that patients who underwent direct VATS had shorter hospital stays. Studies on patients treated with direct VATS have emphasized shorter hospital stays, better empyema drainage, and a higher likelihood of improved lung volume.¹² In a review by Gates et al.¹⁸, it was noted that the shortest hospital stays were associated with thoracotomy, though VATS also resulted in very short stays. Considering the average duration of pleural empyema treatment to be 21 days, we observed that treatment durations in groups 1 and 3 were consistent with the literature, while the treatment duration in group 2 was significantly longer.¹⁹ In this group, the prolonged hospital stay was primarily due to the initial evaluation of the response to fibrinolytic therapy and the subsequent decision to proceed with VATS when no response was observed. We believe that this process is the main factor contributing to the extended hospital stay. Given the lack of significant differences in hospital stay duration, effects on laboratory parameters, complete recovery, and follow-up outcomes between groups 1 and 3, as well as the absence of a standard treatment protocol for pleural empyema, we recommend a multidisciplinary approach for careful evaluation of each patient. This approach can ensure dynamic contributions to the treatment process. If a standard treatment

protocol for the surgical management of empyema can be established, we believe that it would lead to faster treatment responses, shorter hospital stays, reduced thoracic surgery risks, and earlier decisions for patients requiring thoracic surgery.

Limitations

This study's retrospective, single-center design limits its generalizability. Multicenter studies with larger populations are necessary to obtain more accurate results.

CONCLUSION

There is no definitive evidence to suggest the superiority of one treatment method over another for managing pleural empyema. Given the absence of standardized treatment protocols, a multidisciplinary approach involving timely and meticulous evaluation of each patient is crucial. Such an approach can improve prognosis and reduce hospitalization durations. Establishing a standardized protocol for empyema treatment could enhance response rates, shorten hospital stays, mitigate thoracic surgical risks, and facilitate earlier surgical decision-making for patients requiring thoracic intervention.

ETHICAL DECLARATIONS

Ethics Committee Approval

The study was conducted with the permission of Antalya Training and Research Hospital Clinical Researches Ethics Committee (Date: 25.05.2023, Decision No: 7/14).

Informed Consent

Because the study was designed retrospectively, no written informed consent form was obtained from patients.

Referee Evaluation Process

Externally peer-reviewed.

Conflict of Interest Statement

The authors have no conflicts of interest to declare.

Financial Disclosure

The authors declared that this study has received no financial support.

Author Contributions

All of the authors declare that they have all participated in the design, execution, and analysis of the paper, and that they have approved the final version.

REFERENCES

1. de Benedictis FM, Kerem E, Chang AB, Colin AA, Zar HJ, Bush A. Complicated pneumonia in children. *Lancet*. 2020;396(10253):786-798. doi:10.1016/S0140-6736(20)31550-6
2. Ratta A, Nascimben F, Angotti R, et al. Pleural drainage vs video-assisted thoracoscopic debridement in children affected by pleural empyema. *Pediatr Surg Int*. 2023;39(1):287. doi:10.1007/s00383-023-05566-z
3. Barglik R, Grabowski A, Korlacki W, Pasierbek M, Modrzyk A. Pleural empyema in children-benefits of primary thoracoscopic treatment. *Wideochir Inne Tech Maloinwazyjne*. 2021;16(1):264-272. doi:10.5114/wiitm.2020.97443
4. Masters IB, Isles AF, Grimwood K. Necrotizing pneumonia: an emerging problem in children? *Pneumonia (Nathan)*. 2017;9(1):11. doi:10.1186/s41479-017-0035-0



5. Kuru M, Altinok T. Empyema in children. *Turk Gogus Kalp Damar Cerrahisi Derg.* 2024;32(Suppl1):S29-S36. doi:10.5606/tgkdc.dergisi.2024.25759
6. Klopp M, Pfannschmidt J, Dienemann H. Behandlung des pleuraempyems [treatment of pleural empyema]. *Chirurg.* 2008;79(1):83-96. doi:10.1007/s00104-007-1429-y
7. Turan Ciftci T, Akinci D, Unal E, Tanır G, Artas H, Akhan O. Percutaneous management of complicated parapneumonic effusion and empyema after surgical tube thoracostomy failure in children: a retrospective study. *Diagn Interv Radiol.* 2021;27(3):401-407. doi:10.5152/dir.2021.20331
8. Subotic D, Lardinois D, Hojski A. Minimally invasive thoracic surgery for empyema. *Breathe (Sheff).* 2018;14(4):302-310. doi:10.1183/20734735.025718
9. Ljuhar D, Rayner J, Hyland E, King S. Management of thoracic empyema in children: a survey of the Australia and New Zealand Association of Paediatric Surgeons (ANZAPS). *Pediatr Surg Int.* 2021;37(7):897-902. doi:10.1007/s00383-021-04887-1
10. Griffith D, Boal M, Rogers T. Evolution of practice in the management of parapneumonic effusion and empyema in children. *J Pediatr Surg.* 2018; 53(4):644-646. doi:10.1016/j.jpedsurg.2017.07.017
11. Do Manh H, Nguyen Thanh Q, Nguyen Thanh L, Nguyen Van L. Single trocar thoroscopic surgery for pleural empyema in children. *J Laparoendosc Adv Surg Tech A.* 2023;33(7):707-712. doi:10.1089/lap.2019.0637
12. Pogorelić Z, Bjelanović D, Gudelj R, Jukić M, Petrić J, Furlan D. Video-assisted thoracic surgery in early stage of pediatric pleural empyema improves outcome. *Thorac Cardiovasc Surg.* 2021;69(5):475-480. doi:10.1055/s-0040-1708475
13. Grief SN, Loza JK. Guidelines for the evaluation and treatment of pneumonia. *Prim Care.* 2018;45(3):485-503. doi:10.1016/j.pop.2018.04.001
14. Buonsenso D, Cusenza F, Passadore L, et al. Parapneumonic empyema in children: a scoping review of the literature. *Ital J Pediatr.* 2024;50(1):136. doi:10.1186/s13052-024-01701-1
15. Hsieh YC, Hsueh PR, Lu CY, Lee PI, Lee CY, Huang LM. Clinical manifestations and molecular epidemiology of necrotizing pneumonia and empyema caused by *Streptococcus pneumoniae* in children in Taiwan. *Clin Infect Dis.* 2004;38(6):830-835. doi:10.1086/381974
16. Coelho L, Póvoa P, Almeida E, et al. Usefulness of C-reactive protein in monitoring the severe community-acquired pneumonia clinical course. *Crit Care.* 2007;11(4):R92. doi:10.1186/cc6105
17. Medeiros IL, Terra RM, Choi EM, Pego-Fernandes PM, Jatene FB. Evaluation of serial C-reactive protein measurements after surgical treatment of pleural empyema. *Clinics (Sao Paulo).* 2012;67(3):243-247. doi:10.6061/clinics/2012(03)07
18. Gates RL, Caniano DA, Hayes JR, Arca MJ. Does VATS provide optimal treatment of empyema in children? A systematic review. *J Pediatr Surg.* 2004;39(3):381-386. doi:10.1016/j.jpedsurg.2003.11.045
19. Sabbula BR, Rammohan G, Sharma S, Akella J. Lung Abscess. In: StatPearls. Treasure Island (FL): StatPearls Publishing; June 8, 2024.

Maternal awareness of home accidents in children aged 0-3: a cross-sectional study

 Yeşim Tural¹,  Egemen Tural²

¹Department of Pediatrics, Koşuyolu İstanbul Medipol Hospital, İstanbul, Türkiye

²Department of Family Medicine, Çifteler Family Health Center, Eskişehir, Türkiye

Received: 19/01/2025

Accepted: 24/02/2025

Published: 10/04/2025

Cite this article: Tural Y, Tural E. Maternal awareness of home accidents in children aged 0-3: a cross-sectional study. *Surg Child*. 2025;2(2):48-53.

Corresponding Author: Egemen Tural, egementural1@gmail.com

ABSTRACT

Aims: The study sought to examine the awareness of home accidents (HAs) among mothers of children aged 0–3 and identify the factors that influence this awareness.

Methods: This prospective, cross-sectional study was conducted through face-to-face surveys with individuals visiting the Pediatric Health and Diseases Outpatient Clinics of a Private Hospital from October 1 to 31, 2024. A questionnaire was administered, developed based on a literature review, and including sociodemographic information such as age, education level, gender, income level, and occupation. Furthermore, the HAs awareness scale for mothers (HAASM) was used to assess awareness of HAs.

Results: The mean age of participants was 32.52 ± 4.49 years, and the mean age of the children was 13.38 ± 11.55 months. No statistically significant differences were found in HAASM scores based on demographic characteristics or other variables. A weak, negative, and statistically significant correlation was observed between child age and HAASM scores ($r = -0.208$; $p = 0.001$). A statistically significant difference in child age was found between participants whose children had experienced a home accident and those whose children had not ($p = 0.002$).

Conclusion: HAs are a leading cause of childhood morbidity and mortality. Mothers' awareness of HAs decreased as their children's age increased. Considering the growing risk with age, raising awareness, particularly for children approaching 3 years, is crucial. Community education programs and national accident prevention policies are recommended. Additionally, further studies in various settings could contribute to creating safer environments for children.

Keywords: Accidents, home, child health, awareness, pediatrics

INTRODUCTION

Children and the elderly are at higher risk of accidents due to their physical characteristics or health conditions, and in many cases of home accidents (HAs), individuals in the early or late stages of life are the most vulnerable.¹ Children are at a high risk of HAs because they are unable to recognize dangers, are more exposed to environmental risks, and possess a naturally curious disposition.²

From a public health perspective, child injuries resulting from HAs represent a significant concern. Globally, HAs are linked to the deaths of approximately one million children and injuries to 10 million children annually.³ Of these injuries, 95% occur in underdeveloped and developing countries. In developed countries, injuries are responsible for 40% of all deaths among children. Injuries such as burns, falls, poisoning, cuts, and drowning often carry a social dimension and are more commonly observed in economically disadvantaged

communities. For instance, the rate of emergency department visits due to accidental injuries among children under the age of five is 38%, with this rate being significantly higher in impoverished populations compared to wealthier ones.⁴

Although numerical data on HAs in Türkiye are not definitive, a study reported that domestic accidents account for approximately 18–25% of all accidents. Additionally, it was noted that 50–80% of all fatal falls among children aged 0–6 occur at home.⁵

In Turkish society, the responsibility of child-rearing typically falls on mothers.⁶ Therefore, raising mothers' awareness about HAs and improving their knowledge of behaviors that may lead to such accidents are crucial for reducing their occurrence.^{5,7,8} HAs are often overlooked, and there is a scarcity of scientific research examining parents' knowledge about appropriate

behaviors and attitudes to avoid during such incidents involving children.⁹

Particularly in the 1–3 age group, HAs occur more frequently due to factors such as weak motor skills, limited experience, and narrow visual fields.¹⁰ Nevertheless, there is a limited number of studies focusing on the awareness of mothers with children aged 0–3 regarding HAs.

In this study, we aimed to examine the awareness of HAs among mothers of children aged 0–3 and to identify the factors influencing this awareness.

METHODS

Our prospective, cross-sectional study was conducted as a face-to-face survey with individuals visiting the Pediatric Health and Diseases Outpatient Clinics of a Private Hospital. Ethical approval for the study was granted by the İstanbul Medipol University Non-interventional Clinical Researches Ethics Committee (Date: 12.09.2024, Decision No: 876). The study was conducted in accordance with the Declaration of Helsinki.

Participants were informed about the study, and written informed consent was obtained from those who voluntarily chose to participate. Subsequently, a questionnaire was administered, which was developed based on a literature review and included sociodemographic information such as age, education level, gender, income level, and occupation. Additionally, the home accidents awareness scale for mothers (HAASM) was applied to assess awareness of HAs.

Mothers aged 18 and above with children aged 0–3 who agreed to participate in the study were included. Participants under 18 years of age or those who did not complete the questionnaire and scales were excluded from the study.

An average of 590 patients visit the pediatric health and diseases outpatient clinics of the Private Hospital monthly. During the planned one-month study period (October 2024), the expected 590 individuals visiting the pediatric health and diseases outpatient clinics constituted the study population. Based on simple random sampling, the sample size was calculated at a significance level of $\alpha=0.05$ and 95% power, resulting in a minimum required sample size of 233 participants. A total of 241 individuals agreed to participate in the study; however, 1 individual was excluded for being under the age of 18, and 4 individuals were excluded due to incomplete questionnaires and scales. As a result, the study was conducted with 236 participants (Figure).

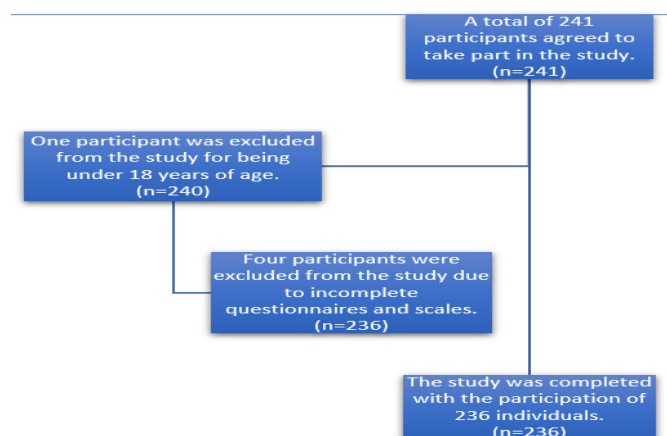


Figure. Flowchart of participant inclusion and exclusion process

HAASM was developed in 2023 by Gülbetekin et al.¹¹, and its validity and reliability were established. This scale consists of 55 items divided into four subdimensions: awareness of burns, falls, drowning-poisoning, and injuries caused by sharp or piercing objects. Higher scores on the scale indicate a greater level of maternal awareness regarding HAs.¹¹

Statistical Analysis

The distribution of demographic characteristics was presented using frequency (n) and percentage (%). The normality of continuous variables was assessed both graphically and using the Shapiro-Wilk test. Descriptive statistics were presented as mean±standard deviation or median (minimum–maximum) values.

For the comparison of HAASM scores across categorical variables with more than two groups, such as education level, occupation, income level, smoking status, and alcohol use, the Kruskal-Wallis non-parametric variance analysis was employed. The Mann-Whitney U test was used to compare HAASM scores between two-group variables.

The Spearman non-parametric correlation coefficient was calculated to analyze the relationship between HAASM scores and variables such as maternal age, child age, number of children, and household size.

IBM SPSS Statistics 21.0 (IBM Corp., 2012, Armonk, NY) and MS Excel 2007 were utilized for statistical analyses, with significance determined at $p<0.05$.

RESULTS

The mean age of the participants was 32.52 ± 4.49 years, and the mean age of the children was 13.38 ± 11.55 months. HAASM scores ranged from 186 to 275, with a mean score of 256.39 ± 17.85 . It was found that 71.6% (n=169) of the participants had one child, 22.5% (n=53) had two children, and 5.9% (n=14) had three children. Additionally, 0.8% (n=2) of the participants lived in households with two people, 66.9% (n=158) in households with three people, 25.1% (n=59) in households with four people, and 7.2% (n=17) in households with five people. The comparison of HAASM scores according to the demographic characteristics of the participants is presented in Table 1.

A weak, negative, and statistically significant correlation was found between child age and HAASM scores ($r=-0.208$; $p=0.001$). No statistically significant correlation was identified between maternal age, number of children, or household size and HAASM scores ($p>0.05$) (Table 2).

A statistically significant difference was observed in child age between participants whose children had experienced a home accident and those whose children had not ($p=0.002$), while no statistically significant difference was found in maternal age ($p=0.062$) (Table 3).

DISCUSSION

HAs in children are a significant concern, as they can lead to injuries and even fatalities.¹² Many young children are unintentionally injured in their own homes, places that are generally considered safe by everyone.¹³ However, compared to other diseases, childhood injuries receive much less attention,

**Table 1.** Comparison of the home accident awareness scale for mothers scores according to demographic characteristics of participants

		All participants	Home accidents awareness scale for mothers	Test statistic	
		n (%)	Median (min-max)	z; χ^2	p
Education level	Primary/secondary	4 (1.7)	240.5 (227-275)	$\chi^2=5.994$	0.112
	High school	21 (8.9)	270.0 (215-275)		
	College	18 (7.6)	262.5 (250-275)		
	University	193 (81.8)	259.5 (186-275)		
Marital status	Married	234 (99.2)	261.0 (186-275)	z=0.001	0.999
	Single	2 (0.8)	260.0 (256-264)		
	Not working/housewife	51 (21.6)	261.0 (206-275)		
Occupation	Official	83 (35.2)	262.0 (188-275)	$\chi^2=2.769$	0.429
	Worker	9 (3.8)	269.0 (239-275)		
	Self-employed	93 (39.4)	258.0 (186-275)		
	Below minimum wage	17 (7.2)	264.0 (206-275)		
Income level	Minimum wage	22 (9.3)	259.0 (186-275)	$\chi^2=3.436$	0.329
	Two times of minimum wage	76 (32.2)	259.0 (205-275)		
	Three times of minimum wage or more	121 (51.3)	261.0 (188-275)		
Chronic diseases	No	202 (85.6)	261.0 (186-275)	z=0.907	0.364
	Yes	34 (14.4)	263.0 (206-275)		
Regular medications	No	207 (87.7)	261.0 (186-275)	z=0.513	0.608
	Yes	29 (12.3)	260.0 (188-275)		
Smoking	No	164 (69.5)	262.0 (186-275)	$\chi^2=0.617$	0.734
	Yes	46 (19.5)	259.5 (224-275)		
	Quit	26 (11.0)	256.5 (206-275)		
Alcohol consumption	No	187 (79.3)	261.0 (186-275)	$\chi^2=0.214$	0.899
	Yes	39 (16.5)	261.0 (206-275)		
	Quit	10 (4.2)	258.0 (221-275)		
Does your child have their own room?	No	29 (12.3)	263.0 (186-275)	z=0.072	0.943
	Yes	207 (87.7)	261.0 (188-275)		
Child chronic diseases	No	227 (96.2)	261.0 (186-275)	z=1.535	0.125
	Yes	9 (3.8)	253.0 (227-272)		
Has your child had a home accident before?	No	206 (87.3)	261.0 (186-275)	z=1.178	0.239
	Yes	30 (12.7)	263.0 (225-275)		

z: Mann-Whitney U test, χ^2 :Kruskal-Wallis test. Min: Minimum, Max: Maximum**Table 2.** The relationship between the home accident awareness scale for mothers score and maternal age, child age, number of children, and number of household members

	Home accident awareness scale for mothers score	
	r	p
Maternal age (years)	-0.040	0.538
Child age (months)	-0.208	0.001
Number of children	-0.054	0.409
Number of household members	-0.046	0.481

r: Spearman correlation coefficient

despite their severity and preventability.¹⁴ In our study, which examined the awareness levels of mothers with children aged 0-3 years regarding HAs, no statistically significant differences were found in HAASM scores based on demographic characteristics and other variables. However, a negative correlation was identified between child age and HAASM scores.

Carlsson et al.¹⁵ conducted an intervention study with mothers of children under the age of 7, which showed that the interventions and informational sessions increased awareness regarding HAs. In the study, a three-question survey was used

Table 3. Comparison of maternal age and child age by home accident history

	No home accident history (n=206)	Home accident history (n=30)	Test statistic	
	Median (min-max)	Median (min-max)	z	p
Maternal age (years)	32.0 (21-45)	35.0 (25-45)	-1.865	0.062
Child age (months)	8.0 (0-36)	17.5 (1-36)	-3.084	0.002

z: Mann-Whitney U test, Min: Minimum, Max: Maximum



to assess awareness. Similarly, during the COVID-19 lockdown in Greece, a study was conducted with 140 parents of children aged 0-14 to investigate the practices implemented by families to prevent HAs. An online survey of 28 questions, created based on a literature review, was applied to the participants. The results indicated that the measures taken by the participants to prevent child HAs were insufficient and needed improvement.¹⁶ A 2024 study conducted in Saudi Arabia with 433 mothers of children aged 1-12 measured knowledge and awareness regarding the prevention of HAs. The study did not use a standardized scale but instead employed a survey created from literature research. In the survey, awareness levels were categorized as low for those scoring below 50%, moderate for those scoring between 50-75%, and high for those scoring 75% or above. The study found that 55% of participating mothers had a moderate level of awareness.¹⁷

Literature reviews indicate that studies on maternal awareness of HAs have used different age ranges and assessment scales. As far as can be determined, no studies have been found that use the HAASM. The discrepancies in study results may be attributed to the variations in the scope of questions included in the scales or surveys employed. It is recommended to increase the number of studies using scales that have undergone validity and reliability testing. Applying the same scale across different patient groups could lead to more comprehensive and reliable outcomes.

Alhadrami et al.¹⁷ conducted a study in Saudi Arabia involving mothers of children aged 1-12 years, finding that 54% of the participants had a child who had previously experienced a home accident. Another study conducted in Saudi Arabia, which investigated the prevalence of HAs in children under 5 years old, showed that, among 352 parents, 205 (58.2%) reported that their child had previously experienced a home accident, most of which were falls and burns.¹⁸ In a study conducted by Aydoğdu et al.¹ in Türkiye with 244 mothers of children aged 0-6 years, it was found that mothers whose children had not previously experienced a home accident were more likely to recognize safety measures for HAs than mothers whose children had experienced one. In the study, 42.6% of the participants reported that their child had previously experienced a home accident. In a study conducted by Üçüncü et al.¹⁹ in Türkiye, which explored the attitudes and knowledge of mothers with children aged 0-6 years regarding HAs and prevention strategies, it was found that 116 out of 217 participants (58%) had at least one child who had experienced a home accident. In a study by Gündüz and Aytekin² with mothers of children aged 1-3 years, 229 out of 400 participants (57.3%) reported that their children had previously experienced a home accident. In a study by Al Rumhi et al.²⁰, children who presented to the emergency department due to HAs were retrospectively examined. The study categorized the children's age groups as 12-18, 6-12, 3-6, 1-3, and 0-1 years. Among a total of 1333 emergency visits due to HAs, the smallest number and percentage were found in the 0-1 age group (111 children, 8.9%), while the largest number and percentage were in the 1-3 age group (370 children, 27.8%).²⁰

In our study, it was determined that 12.7% (30 participants) of the mothers had children with a history of HAs. Additionally, no statistically significant difference was found between mothers whose children had experienced a home accident and those whose children had not, in terms of their HAASM scores.

However, there was a statistically significant difference between those who had experienced HAs and those who had not, in terms of the children's age. One reason why our study has a lower prevalence of home accident history compared to other studies conducted in Türkiye could be that it was conducted in a private hospital. Due to the socioeconomic advantages of the participants, they may have been able to take more precautions against HAs, and it is possible that their homes are structurally safer. Another reason may be that, unlike other studies, our study specifically involved mothers of children aged 0-3 years, and the higher educational level of the participant mothers, compared to those in other studies, may have contributed to the findings.

Studies examining HAs in children frequently evaluate the education levels of families. Üçüncü et al.¹⁹ divided the mothers of children aged 0-6 years into two groups based on their education level in their study: those with education below high school and those with high school or higher education. The study found that the group with higher education had significantly higher awareness of HAs. The study also emphasized that mothers with higher education and those who had received training and information on preventing HAs would play an important role in protecting children from HAs.¹⁹ İnce et al.'s²¹ study, conducted in Türkiye, explored the attitudes of mothers toward accident and injury prevention. The results of the survey indicated that accident prevention scores improved with higher education levels.

On the other hand, in a study by Çiçekler et al.²², it was found that mothers with only elementary school education took more safety measures against HAs compared to mothers with university degrees. Santos et al.²³, in their study in Brazil with 256 participants caring for children under 6 years old, examined the relationship between the level of knowledge about precautions for HAs and education level. In the study, 170 participants (66.4%) had education levels higher than high school, but there was no statistically significant difference in the level of knowledge about HAs based on education level.²³ In our study, no statistically significant difference was found between the mothers' education level and HAASM scores. Although it is argued that mothers with higher education levels are likely to have more knowledge about child HAs, children's developmental characteristics, and risk factors for accidents², it should be noted that the level of knowledge about HAs may be influenced by access to information sources about home safety and cultural and social factors.¹⁷

When examining the factors affecting attitudes toward HAs, maternal age has been evaluated in several studies. In a study by Alhadrami et al.¹⁷, where more than 70% of the participants were over 30 years old, no relationship was found between maternal awareness of HAs and age. Similarly, in a study by Üçüncü et al.¹⁹, mothers were divided into two groups: under 35 and 35 or older, but no significant difference in awareness of HAs was found between the groups. In Aydoğdu et al.'s¹ study, the average age of participants was 30.8 years, and no relationship was found between age and the precautions taken regarding HAs. Consistent with the literature, our study found no statistically significant relationship between maternal age and HAASM scores.

Parents' attitudes and behaviors toward HAs may also vary depending on the child's age. In a study by İnce et al.²¹,



participants were grouped as parents of children aged 0-1, 1-4, and 5-9 years. The group with the highest attitude toward preventing accidents and injuries were parents of children aged 0-1 years. In a similar study by Eichelberger et al.²⁴, which included 404 parents of children under the age of 13, it was observed that parents of younger children were more inclined to engage in appropriate accident prevention behaviors. In our study, a statistically significant negative relationship was found between child age and HAASM scores. This may be due to the increased care dependency of children aged 0-1 years, making their parents more vigilant about HAs for this age group.

Papachristou et al.¹⁶ demonstrated in their study that participants with one or two children took more preventive measures against HAs compared to those with more children. It was suggested that larger families may have less time to dedicate to children and to organizing protective measures against accidents. Üçüncü et al.¹⁹ observed in their study that as the number of children and the number of people living in the household increased, knowledge of HAs and the adoption of safety measures decreased. A study conducted in Saudi Arabia in 2023 found that in families with more than seven members and families with more than four children, HAs occurred more frequently than in other families, with statistically significant results.¹⁸ However, in our study, no correlation was observed between the number of household members or the number of children and the HAASM scores. This outcome may be attributed to the relatively high and more homogeneous socioeconomic status of the participants in our study compared to other studies, or to the limited variability in HAASM scores due to the generally high levels of awareness among the participants, which may have hindered the detection of potential associations.

Limitations

Our study has several limitations. First, the cross-sectional design makes it challenging to establish a causal relationship between awareness of HAs and associated risk factors. Second, the use of the scale exclusively on mothers, without considering the perspectives of both parents, constitutes a limitation. Third, the fact that the study was conducted in a private hospital limits the ability to generalize the findings to a larger population. Finally, the location and structural features of the homes, the presence of any psychiatric pathologies in the mothers, whether the mothers received first aid training, and the type of HAs experienced were not examined in our study. These factors can also be considered as limitations of the study.

CONCLUSION

HAs are a major cause of morbidity and mortality in children globally. In our study, 12.7% of participants reported a history of HAs in their children. As children's age increased from 0 to 3 years, a decrease in mothers' awareness of HAs was observed. Considering that the risk of HAs increases with age, it is crucial to raise awareness, particularly regarding HAs for children nearing the age of 3. Community-based educational programs for mothers and families, as well as nationwide policies aimed at accident prevention, are recommended. Furthermore, conducting studies on HAs in different centers and among various patient groups could contribute to the prevention of accidents and provide a safer environment for children to grow up in.

ETHICAL DECLARATIONS

Ethics Committee Approval

Ethical approval for the study was granted by the İstanbul Medipol University Non-interventional Clinical Researches Ethics Committee (Date: 12.09.2024, Decision No: 876).

Informed Consent

All patients signed and free and informed consent form.

Referee Evaluation Process

Externally peer-reviewed.

Conflict of Interest Statement

The authors have no conflicts of interest to declare.

Financial Disclosure

The authors declared that this study has received no financial support.

Author Contributions

All of the authors declare that they have all participated in the design, execution, and analysis of the paper, and that they have approved the final version.

REFERENCES

1. Aydoğdu ZA, Ateş E, Set T. Assessment of mothers' measures against home accidents for 0-6-year-old children. *Türk Pediatri Ars.* 2019;54(3): 149-156. doi:10.14744/TurkPediatriArs.2019.64614
2. Gündüz G, Aytekin A. Annelerin çocuklarını ev kazalarından korumaya yönelik tutumları ve etkileyen faktörler. *İzmir Dr Behçet Uz Çocuk Hast Dergisi.* 2015;5(3):184-192. doi:10.5222/buchd.2015.184
3. Acar E, Dursun OB, Esin İS, Öğütlü H, Özcan H, Mutlu M. Unintentional injuries in preschool age children: is there a correlation with parenting style and parental attention deficit and hyperactivity symptoms. *Medicine (Baltimore).* 2015;94(32):e1378. doi:10.1097/MD.0000000000001378
4. Howe LD, Huttly SR, Abramsky T. Risk factors for injuries in young children in four developing countries: the young lives study. *Trop Med Int Health.* 2006;11(10):1557-1566. doi:10.1111/j.1365-3156.2006.01708.x
5. Turan T, Altundağ Dündar S, Yorgancı M, Yıldırım Z. 0-6 yaş grubu çocuklarda ev kazalarının önlenmesi. *Ulus Travma Acil Cerrahi Derg.* 2010;16(6):552-557.
6. Çalışkan K, Avcı Ö, Acar V, Dönmez CY. 0-6 yaş grubu çocuğu olan annelerin düşmelere ilişkin ilkyardım uygulamalarının incelenmesi. *Maltepe Üniversitesi Hemşirelik Bilim ve Sanatı Dergisi.* 2010;3(3):2-9.
7. Yiğit D, Şayık D, Açıkgöz A, Mumcu Ö. Diagnosis of mothers' safety measures for home accidents and determination of first aid self-efficiency during the pandemic. *STED.* 2023;31:451-460.
8. Uskun E, Alptekin F, Öztürk M, Kışioğlu AN. Ev hanımlarının ev kazalarını önlemeye yönelik tutum ve davranışları ile ev kazalarına yönelik ilkyardım bilgi düzeyleri [the attitudes and behaviors of housewives in the prevention of domestic accidents and their first aid knowledge levels]. *Ulus Travma Acil Cerrahi Derg.* 2008;14(1):46-52.
9. Coulibaly A, Sogo AE, Bara A, Wildhaber BE, Inglin S. Domestic accidents of children in the Orodara District of Burkina Faso: mothers' knowledge of first-aid practices. *Int J Environ Res Public Health.* 2024; 21(5):523. doi:10.3390/ijerph21050523
10. Arıkan D, Çelebioğlu A, Tüfekçi FG. Çocukluk dönemlerinde büyüme ve gelişme. Conk Z, Başbakkal Z, Yılmaz HB, Bolışık B (eds). *Pediatric Hemşireliği* Ankara. Akademisyen Tıp Kitapevi, 2013.
11. Gulbetekin E, Tufekci FG. A new scale for evaluating home accidents: home accidents awareness scale for mothers with 0-3-year-old children. *Med Bull Haseki.* 2023;61(2):88-95. doi:10.4274/haseki.galenos.2023.8868
12. Ablewhite J, Peel I, McDaid L, et al. Parental perceptions of barriers and facilitators to preventing child unintentional injuries within the home: a qualitative study. *BMC Public Health.* 2015;15(1):280. doi:10.1186/s12889-015-1547-2
13. Simpson J, Fougere G, McGee R. A wicked problem: early childhood safety in the dynamic, interactive environment of home. *Int J Environ Res Public Health.* 2013;10(5):1647-1664. doi:10.3390/ijerph10051647
14. Sleet DA. The Global challenge of child injury prevention. *Int J Environ Res Public Health.* 2018;15(9):1921. doi:10.3390/ijerph15091921



15. Carlsson A, Dykes AK, Jansson A, Bramhagen AC. Mothers' awareness towards child injuries and injury prevention at home: an intervention study. *BMC Res Notes*. 2016;18(9):223. doi:10.1186/s13104-016-2031-5
16. Papachristou E, Deftereos S, Asimakidou M, et al. Parental home safety practices for domestic accident prevention: how prepared were parents for COVID-19 confinement? A cross-sectional study. *Clin Pract*. 2023; 13(6):1449-1459. doi:10.3390/clinpract13060129
17. Alhadrami LM, Habib HS, Osman BK, Alqarni LM, Almutiri SF, Alhomoud SA. Maternal knowledge and awareness of preventive measures for domestic accidents among children in Jeddah, Saudi Arabia. *Cureus*. 2024;16(10):e71226. doi:10.7759/cureus.71226
18. Alamr F, Alzahrani HMA, Alghamdi AMA, et al. Prevalence and risk factors of home accidents among children under five years of age in Al-Baha, Saudi Arabia. *Cureus*. 2023;15(10):e46846. doi:10.7759/cureus.46846
19. Ucuncu M, Ucuncu M, Toprak D. The knowledge, attitude, and behavior of mothers with children aged 0-6 years on home accidents, and preventive measures. *J Ist Faculty Med*. 2019;82(4):219-228. doi:10.26650/IUITFD.2018.0011
20. Al Rumhi A, Al Awisi H, Al Buwaiqi M, Al Rabaani S. Home accidents among children: a retrospective study at a tertiary care center in Oman. *Oman Med J*. 2020;35(1):e85. doi:10.5001/omj.2020.03
21. İnce T, Yalçın S, Yurdakök K. Parents' attitudes and adherence to unintentional injury prevention measures in Ankara, Turkey. *Balkan Med J*. 2017;34(4):335-342. doi:10.4274/balkanmedj.2016.1776
22. Çiçekler CY, Er KR, Pirpir AD, Büyükbayraktar Ç. 0-6 yaş grubunda çocuğu olan annelerin ev kazalarına yönelik güvenlik önlemlerinin çeşitli değişkenlere göre incelenmesi. *Çukurova Üniversitesi Sosyal Bilimler Enstitüsü Dergisi*. 2012;21(3):157-174.
23. Santos RR, Machado MED, Gomes ALM, Aguiar RCB, Christoffel MM. Prevention of domestic accidents in childhood: knowledge of caregivers at a health care facility. *Rev Bras Enferm*. 2022;75(2):e20210006. doi:10.1590/0034-7167-2021-0006
24. Eichelberger MR, Gotschall CS, Feely HB, Harstad P, Bowman LM. Parental attitudes and knowledge of child safety. A national survey. *Am J Dis Child*. 1990;144(6):714-720. doi:10.1001/archpedi.1990.02150300112029

Comparison of outcome of preformed silo with traditionally constructed silo among neonates with gastroschisis

 Nusrat Jahan Mona,  Samiul Hasan,  Ashrarur Rahman Mitul

Department of Neonatal and Paediatric Surgery, Bangladesh Shishu Hospital and Institute, Dhaka, Bangladesh

Received: 10/01/2025

Accepted: 10/01/2025

Published: 10/04/2025

Cite this article: Mona NJ, Hasan S, Mitul AR. Comparison of outcome of preformed silo with traditionally constructed silo among neonates with gastroschisis. *Surg Child*. 2025;2(2):54-59.

Corresponding Author: Nusrat Jahan Mona, dr.nusratjahancnrs@gmail.com

ABSTRACT

Aims: The study aimed to observe the usefulness of a preformed silo (PFS) made of Alexis wound protector/ retractor in comparison to those patients undergoing traditionally constructed silo under general anesthesia (GA).

Methods: This is a quasi-experimental study conducted in the Division of Paediatric Surgery, Bangladesh Shishu Hospital, from October 2022 to March 2024. Patients were randomly divided into group A and group B respectively. Group A, PFS using a Alexis wound retractor/protector was done without the use of GA, and the bowel was gradually allowed to reduce into the abdominal cavity. For group B, the traditional silo was constructed using a urobag under GA after doing the proper preoperative preparations. Outcome variable arrival to procedure time, bowel reduction time, initiation of first enteral feed, survival duration, and mortality were recorded and compared.

Results: A total of 60 patients with gastroschisis were enrolled in this study, of which 30 patients in each group. Both groups had a mean age of 1 day with a gestational age of 36 weeks respectively. The PFS group showed a longer survival duration of around 22 days and with the traditional silo around 12 days respectively. Initiation of enteral feed after closure was on the 5th postoperative day (POD) for the traditional silo and the 3rd POD for the PFS. Mortality of 48% in the PFS group and 52% in the traditional silo group.

Conclusion: PFS (Alexis wound retractor) can bypass the hazards of GA and can become a safer alternative to traditionally constructed silo, particularly in resource-limited settings.

Keywords: Gastroschisis, preformed silo, Alexis wound retractor, traditional silo

INTRODUCTION

Gastroschisis is one of the most common congenital abnormalities, with a growing global incidence. This anomaly of the newborn causes the intestinal loops to protrude out through an abnormal opening in the abdominal wall. This opening commonly occurs on the right of the umbilicus. The incidence is about 2 to 5 per 10,000 live births, with a male preponderance.¹ Worldwide, there is a significant variation in the outcomes for newborns with gastroschisis. Most low and middle-income countries (LMICs) report higher mortality rates between 75% and 100% compared with <4% in high-income countries (HICs).² This difference in mortality is mainly due to the advanced management protocol strategies of resuscitation followed by HICs. The strategies include maintenance of temperature, fluid and electrolyte balance, nutritional support, cardiopulmonary support, and ventilation support. Initial bowel coverage, and subsequently visceral reduction by primary or delayed staged closure, along with total parenteral nutrition (TPN) support which is easily available in

the developed countries, unlike the low-income countries like Bangladesh.² Many researchers encountered promising results, with reduced mortality by implementing the use of preformed silo (PFS) in low-income countries.³

Gastroschisis patients require immediate and urgent care due to their prematurity and large exposed area of bowel which warrants close attention. The unprotected bowel requires preservation of heat, prevention of loss of fluid along with respiratory support. The earlier the bowel is covered and allowed to reduce, the less chance of bowel edema and fibrinous coating to accumulate over the open intestinal loops.⁴ To minimize heat loss, and water loss and to prevent the intestinal injury from worsening, the first line of treatment should focus on rapid and immediate repositioning and covering of the exposed bowel in order to reduce the mortality.⁵

Treatment options for the reduction of contents of gastroschisis vary greatly. Traditionally, the most common techniques



include primary closure or delayed staged closure by traditional silos under anesthesia or delayed staged closure using a PFS at the patient's bedside without general anesthesia (GA).⁶ With a reported success rate ranging from 50% to 83%, immediate primary closure can be carried out in the operating room under GA in places where facilities are available. Immediate closure has benefits such as preventing further intestinal inflammation from mechanical irritation and exposure, allowing for earlier enteral feed initiation, reducing the duration of hospital stay (LOS), and lowering the risk of wound infection. But primary closure comes with drawbacks such as the requirement for GA, an increase in ventilator days, the need for nutritional care like TPN, NICU support, and complications like abdominal compartment syndrome (ACS).⁷

Nevertheless, primary closure is not always possible, in the case of patients who have excessively thickened, distended bowel (matting), small abdominal domain, or complex gastroschisis (atresia, necrosis, perforation) which may require delayed staged closure, which can be further achieved by either by a traditional or PFS. A traditional silo is mostly constructed using a Saline bag or a Urobag sutured around the defect, requiring preoperative preparation and GA. The delayed staged closure lowers the risk of ACS, and decreased ventilator days. Both primary closure and traditional silo require GA, neonates account for about 55% of anesthetic-related cardiac arrests, with 82% occurring during induction. Respiratory risks of anesthesia include laryngospasm, airway obstruction, and difficult intubation. Recent studies have also revealed the toxic effects of general anesthetics on pediatric patients. Neonates are also more prone to developing malignant hyperthermia.⁸

A PFS, consists of a transparent silastic bag with a spring-loaded ring, which has been introduced and extensively used over the past 20 years, to avoid GA. Others have used bedside reduction without anesthesia (Bianchi Procedure)⁹, Alexis wound protector/ retractor as PFS used for staged reduction at the cot side, in places where spring loaded PFS is not available or is expensive.¹⁰ Advantages of PFSs include, their low technological needs, ability to be administered at the bedside by any health care professional, and avoidance of newborn anesthesia and surgery. PFS negates the need for critical care due to less chance of developing ACS.¹¹

PFS has also been shown to be successful in decreasing mortality in low-resource environments where there are insufficient frameworks, inconsistent availability of pediatric surgeons, and inadequate staffing and training for newborn anesthesia and surgery.² Bangladesh, being a developing country, suffers from, a lack of resources, social prejudice, and insufficient information. Along with inexperienced health workers and medical personnel, who play a vital role in the management of patients with gastroschisis.¹² The PFS may help bypass the modulates that are scarce in developing countries like Bangladesh.¹³ The use of Alexis wound protector/retractor as PFS in places where PFS was not easily available has helped to reduce mortality in countries with limited logistics.¹⁴

This study aims to observe the usefulness of a PFS made of Alexis wound protector/retractor and to compare its outcome with those of babies undergoing traditionally constructed silo under GA.

METHODS

The study was carried out with the permission of the Ethical Committee Bangladesh Shishu Hospital and Institute (Date: 25.10.2022, Decision No: 2023702). All procedures were carried out in accordance with the ethical rules and the principles of the Declaration of Helsinki.

This was a quasi-experimental study. This study approach was selected as random assignment of neonates to either treatment modality was not feasible due to ethical concerns and clinical protocols. The study was conducted in Bangladesh Shishu Hospital and Institute (BSHI), Dhaka, Bangladesh, from October 2022 to March 2024.

Neonates with gastroschisis admitted to Bangladesh Shishu Hospital and Institute (BSHI) were included in this study. Complicated gastroschisis (with atresia, perforation, volvulus) and patients with any major congenital anomalies like (diabetes mellitus, and congenital cyanotic heart disease) were excluded from this study.

Convenient purposive sampling was done. Total of 60 patients were included in this study and divided into two groups, group A (PFS) and group B (Traditionally constructed) silo with 30 patients in each group. Out of the study population, the individual sample units were selected according to clinical judgment until the desired sample size was attained. The sample size was determined based on a power analysis conducted during the study design phase. Using G*power software, we estimated that a sample size of 60 would provide adequate statistical power (0.80) to detect a medium effect size (Cohen's $d=0.5$) with a significance level (α) of 0.05 for an independent t-test comparing the two groups. Practical considerations, such as the availability of participants and resource constraints were also taken into account.

The demographic data was extracted using pre-designed questionnaire. The outcome parameters included the arrival to procedure time, bowel reduction time, initiation of enteral feed after closure, survival duration, and mortality. Patients in each group were not under any regular prenatal follow up detection of defect was at birth for both groups. No post-discharge follow-up was included in this study.

Study Procedure

All gastroschisis patients were managed under the close guidance of pediatric surgeons.

Initial management: Bowel protection with a translucent bag, for temperature regulation and homeostasis, and for prevention of evaporative losses. The presence of hypoxia, hypothermia, and dehydration was recorded. The presence of hypoxia was measured by using a pulse oximeter, temperature was measured using an electronic thermometer. The presence of dehydration was noted by the presence of depressed fontanelle, sunken eyes, skin pinch, and urine output.

An initial examination of the intestines was done to exclude obvious findings of atresia, necrosis, perforation, or volvulus. Bowel condition, presence of edema, matting, perforation, or necrosis recorded. A nasogastric tube was placed for gut decompression and a urinary catheter was introduced to

empty the urinary bladder. Intravenous access was acquired for fluid resuscitation. Patient weight recorded. Patients who met the inclusion criteria were randomly divided into group A and group B respectively.

For group A: PFS (Alexis wound retractor/protector) made of a colorless transparent polyurethane film wrap, having two rings at both ends was used. The bottom ring is blue and is flexible, the upper ring is white and folds on itself. After the initial management, the intestinal loops were placed inside via the bottom ring into the silo, the bottom end of the PFS was then carefully introduced into the abdominal cavity, in the operative room without the use of GA and mechanical ventilation (**Figure 1**). The upper ring was then twisted and the opening was tied with a gauge or suture material, then the ring was folded on itself to keep slight pressure on the abdominal wall (**Figure 2**). Analgesic was given per-rectally. The silo was checked daily, and the reduction time is recorded (**Figure 3**). After Application of PFS (Alexis wound retractor).



Figure 1. After application of traditional silo



Figure 2. Application of preformed silo (Alexis wound retractor), after insertion of the bottom ring within abdominal cavity, the upper ring twisted and tied with suture material to allow pressure to be exerted on the abdominal wall

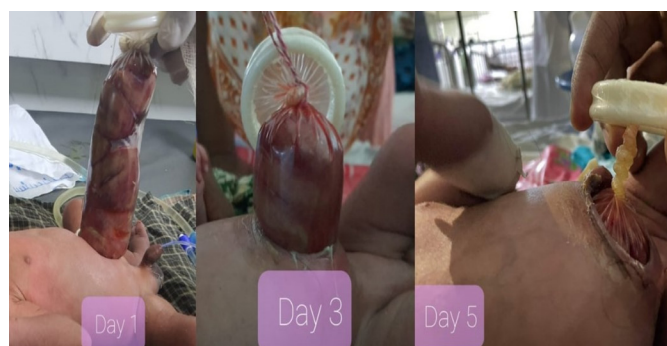


Figure 3. Serial reduction with preformed silo

For group B: Preoperative anesthetic investigations done. Traditional silo constructed under GA. After induction of the anesthesia, a sterile Urobag was taken and cut to an appropriate size according to the gastroschisis defect, the Urobag was then sutured around the defect. The silo was checked daily and the reduction time was recorded.

After the procedure: The daily decrease of the bowel content within the silo inspected, the days required for reduction recorded.

After Complete Reduction

Group A: Closure was done in the operation theatre without GA.

Group B: Closure was done in the operation room under GA.

After closure, the postoperative day (POD) of initiation of the first enteral feed was recorded.

A complete blood count with peripheral blood, C-reactive protein, and blood culture was sent when sepsis was suspected.

Statistical Analysis

Data were coded, entered, and analyzed in a computer. The statistical analysis was conducted using SPSS (Statistical Package for Social Science) version 26 statistical software. The findings of the study were presented by frequency, and percentage in tables. Mean and standard deviation for continuous variables and frequency distribution for categorical variables were used to describe the characteristics of the total samples. Associations of continuous data were assessed using student t-tests. Fisher's exact test was used for mortality. The value of $p < 0.05$ was considered significant.

Operational Definitions

- 1. Arrival time:** Time taken in hours for the patient to reach the hospital after birth.
- 2. Reduction time:** Time taken in days for the eviscerated bowel to return to the abdomen and be ready for closure.
- 3. Survival duration:** Time in days from procedure to discharge or death.

RESULTS

In both groups A and B, most patients presented at the age of 1 day, with no significant difference (**Table 1**). Both groups A and B had similar gestational age of 36 weeks, with a p -value of 0.819 which was not significant (**Table 1**). In both group A and B.

Table 1. Comparison of demographic variables between group A and group B

Variables	Group A n=30 f (%)	Group B n=30 f (%)	p-value
Age (days) Mean $\pm$ SD (range)	1.5 $\pm$ 0.51 (1-2)	1.4 $\pm$ 0.57 (1-3)	0.600
Gestational age (weeks) mean $\pm$ SD (range)	36.4 $\pm$ 0.86 (33-37)	36.2 $\pm$ 0.82 (34-38)	0.819
Weight at birth (kg) mean $\pm$ SD (range)	2.16 $\pm$ 0.37 (1.5-3.0)	2.22 $\pm$ 0.44 (1.5-3.2)	0.126
Hospital arrival time (hours) mean $\pm$ SD (range)	8.70 $\pm$ 1.60 (6-12)	8.97 $\pm$ 1.90 (5-12)	0.344

SD: Standart deviation



The weights for both group A and group B were also similar about 2 kg, with no significant difference (**Table 1**). In both groups, the maximum arrival time was 12 hours, with a p-value of 0.344 showing no significant difference (**Table 1**).

For group A arrival to procedure time was a maximum of 3 hrs and for group B, the maximum time taken from arrival to procedure was 11 hours. The results were statistically significant (p-value: 0.0001) (**Table 2**). In group A most patients required a mean of 5 days for complete reduction and in group B most patients required a mean of 8 days for complete reduction. The difference between the groups was not statistically significant (**Table 2**).

Table 2. Comparison of variables between group A and group B

	Group A (preformed silo) (n)	Group B (traditional silo) (n)	p-value
Arrival to procedure (hours)	2.33±0.48 (30)	9.43±1.07 (30)	0.0001
Bowel reduction time (days)	5.33±0.48 (30)	7.80±0.55 (30)	0.796
Start of enteral feed after closure, POD (days)	2.73±0.45 (26)	5.14±0.36 (21)	0.034
Survival duration (days)	22.97±3.56 (30)	12.47±3.50 (30)	0.479
Mortality	48% (26)	52% (28)	0.268
Cost of procedure	4000 BDT	12000 BDT	

POD: Postoperative day

The enteral feed initiation was possible on the 3rd post-operative day for group A. Whereas for group B enteral feed initiation was possible on the 5th the post-operative day. The number of patients varied in each group as some patients expired before enteral feed could be started. The results were significant with a p-value of 0.034 (**Table 2**).

Survival duration for group A was around 22 days, and in group B survival duration was around 12 days. This difference between the two groups was not significant with a p-value of 0.479 (**Table 2**). Mortality was 48% in group A and 52% in group B respectively (**Table 2**). The main cause mortality was sepsis which was determined by blood culture. In both groups, onset of sepsis was detected on 3rd to 5th day after procedure, the cause of sepsis was determined to be nosocomial infection in few of the cases but most cases the cause remained undetermined, detailed determinant of sepsis was not included in this study.

DISCUSSION

This study was carried out in order to compare whether the PFS (Alexis wound retractor) can be used as an alternative to Traditionally constructed silo.

In this study, the arrival to procedure time in the PFS group, which was placed without GA, was significantly lower ranging from 2-3 hours compared to traditionally constructed silo which was done under GA and yielded a range of 8-11 hours, the time taken from arrival to the procedure was statistically significant in this study. Previous research showed PFS allowed for faster application, bypassing GA, operation, and avoiding

hand sewing of silo, unlike traditional silo construction which is in agreement with this study.^{3,15,16} The requirement for GA, and endotracheal tube insertion leads to an increased risk of neurotoxicity, the PFS can help bypass these added risks.^{13,17}

The present study result reflects that the bowel reduction time was shorter in the PFS group, compared to the bowel reduction time in the traditionally constructed silo group, even though the results were statistically not significant, these findings are in parallel with the research which demonstrated shorter bowel reduction time was achievable with the use of PFS which leads to the early establishment of enteral feed.¹⁸ The PFS allowed equal dissemination of pressure along the abdominal wall due to the presence of the uniform circular ring unlike the traditionally constructed silo where hand sewing is required, this differences in pressure distribution may have contributed to the difference in results.¹⁸

In this current study, the initiation of enteral feed after closure was earlier in the PFS group with the highest frequency occurring on the 3rd POD compared to the traditionally constructed silo group where the highest frequency occurred on the 5th POD, these results were statistically significant and are in alignment with conclusions made in studies by.^{13,19} Who reported earlier first feed was achievable with the use of PFS. Application of PFS followed by delayed closure in infants with gastroschisis was associated with rapid return of bowel function was also reported in a study by.²⁰ Early initiation of enteral feed can lead to better outcomes as reported in previous studies.²¹ As no GA or surgical trauma was inflicted for the PFS, no delay in reversal of bowel movement was encountered, which may have contributed to the similarities in results.

In this study, the survival duration for the patients with gastroschisis treated with the PFS was higher compared to the patients treated with traditionally constructed silo but these results were not statistically significant. There were no similar studies comparing survival duration between preformed and traditional silos. However, some studies have concluded that the survival duration was not dependent on the type of intervention, in the case of gastroschisis management.²²

This study reports showed that the mortality was similar in both groups, with 48% mortality in the PFS group and 52% mortality in the traditionally constructed silo respectively. The main cause of death was found to be sepsis in both groups. This can be explained by the deficiency in the neonatal friendly environment, non-availability of NICU, improper immediate primary resuscitation, and absence of TPN. The type of intervention did not play a role in the mortality of patients in both groups, these findings were in conjunction with other study findings.²³ Unhygienic health delivery systems including maternal and neonatal health, poor transportation facilities without stabilization, delay in presentation, and late referrals make the situation worse and result in poor outcomes. Nosocomial infection, antimicrobial resistance, sharing wards, and equipments have all been identified as the source of sepsis for neonates leading to a high mortality in developing countries.²⁴ According to reports it is suggested that gastroschisis can become an important indicator in reflecting the ability of an institute to provide proper and adequate healthcare.²⁵

In the present study, both groups presented with a gestational age of 36 weeks mostly preterm, with a high frequency of birth



weight of 2 kg, with a male predominance of 68% similar to other studies.^{26,27}

These previous authors have concluded that gastroschisis with a lower birth weight can lead to poor outcomes due to their increased susceptibility to infection and need for nutritional support.

Most of the patients in this study, similar to previous research, had a delay in hospital arrival with a maximum of 12 hours in each group but this was not statistically significant in this study.²⁸ Even though it was not statistically significant this delay in hospital arrival with inadequate resuscitation may eventually contribute to unfavourable outcomes. Late presentation to the tertiary centers without adequate resuscitation is a major problem in the management of gastroschisis in low-resource framework was reported.²⁹

The cost of PFS in comparison to traditionally constructed was quite low as reflected in the present study. Thus with the limited logistics, the PFS can be used in place of the traditional use of primary closure or traditional silo, avoiding the need for neonatal anesthesia and surgery, thus helping to improve the mortality in developing countries like Bangladesh.¹⁴

Limitations

- Birth to arrival time was not under the control of the researcher.
- The neonatal friendly environment could not be ensured in all cases.
- Long term follow-up and post-discharge data was not included in this study.

Recommendation

- Alexis wound retractor can be used as an alternative for traditionally constructed silos.
- Further comparative study between both these procedures with optimal perioperative conditions should be carried out.

CONCLUSION

As an alternative to traditionally constructed silos, PFSs can avoid the risks associated with GA and can be a more affordable and safer option, particularly in resource constraints environment. However, to greatly increase these patients' chances of survival, enhanced perioperative neonatal care is needed.

ETHICAL DECLARATIONS

Ethics Committee Approval

The study was carried out with the permission of Ethical Committee Bangladesh Shishu Hospital and Institute (Date: 25.10.2022, Decision No: BSHI/2022/2645).

Informed Consent

Written informed consent was obtained from the parents of all patients.

Referee Evaluation Process

Externally peer-reviewed.

Conflict of Interest Statement

The authors have no conflicts of interest to declare.

Financial Disclosure

The authors declared that this study has received no financial support.

Author Contributions

All of the authors declare that they have all participated in the design, execution, and analysis of the paper, and that they have approved the final version.

REFERENCES

1. Gonzalez DO, Cooper JN, St. Peter SD, Minneci PC, Deans KJ. Variability in outcomes after gastroschisis closure across U.S. children's hospitals. *J Pediatr Surg.* 2018;53(3):513-520. doi:10.1016/j.jpedsurg.2017.04.012
2. Wright NJ, Langer M, Norman IC, et al. Improving outcomes for neonates with gastroschisis in low-income and middle-income countries: a systematic review protocol. *BMJ Paediatr Open.* 2018;2(1):e000392. doi:10.1136/bmjpo-2018-000392
3. Zalles-Vidal C, Peñarrieta-Daher A, Bracho-Blanchet E, et al. A Gastroschisis bundle: effects of a quality improvement protocol on morbidity and mortality. *J Pediatr Surg.* 2018;53(11):2117-2122. doi:10.1016/j.jpedsurg.2018.06.014
4. Davenport M, Haugen S, Greenough A, Nicolaides K. Closed gastroschisis: antenatal and postnatal features. *J Pediatr Surg.* 2001;36(12):1834-1837. doi:10.1053/jpsu.2001.28856
5. Chesley PM, Ledbetter DJ, Meehan JJ, Oron AP, Javid PJ. Contemporary trends in the use of primary repair for gastroschisis in surgical infants. *Am J Surg.* 2015;209(5):901-906. doi:10.1016/j.amjsurg.2015.01.012
6. Zani A, Rutenstock E, Davenport M, Ade-Ajayi N. Is there unity in Europe? First survey of EUPSA delegates on the management of gastroschisis. *Eur J Pediatr Surg Off J Austrian Assoc Pediatr Surg Al Z Kinderchir.* 2013;23(1):19-24. doi:10.1055/s-0032-1326954
7. Dekonenko C, Fraser JD. Approaches for closing gastroschisis. *Adv Pediatr.* 2020;67:123-129. doi:10.1016/j.yapd.2020.03.005
8. Butterworth J, Mackey D, Wasnick J. Morgan and Mikhail's Clinical Anesthesiology, 7th Edition. McGraw Hill / Medical; 2022.
9. Emami CN, Youssef F, Baird RJ, et al. A risk-stratified comparison of fascial versus flap closure techniques on the early outcomes of infants with gastroschisis. *J Pediatr Surg.* 2015;50(1):102-106. doi:10.1016/j.jpedsurg.2014.10.009
10. Ade-Ajayi N, Wright N, Zani A. Epidemiology, management and outcome of gastroschisis in Sub-Saharan Africa: results of an international survey. *Afr J Paediatr Surg.* 2015;12(1):1. doi:10.4103/0189-6725.150924
11. Allotey J, Davenport M, Njere I, et al. Benefit of preformed silos in the management of gastroschisis. *Pediatr Surg Int.* 2007;23(11):1065-1069. doi:10.1007/s00383-007-2004-9
12. Rashid M, Hoque M, Mamun A, Kabir A, Ibrahim M, Khan A. Management of gastroschisis in a tertiary care hospital. *Bangladesh J Child Health.* 2013;36(3):133-138. doi:10.3329/bjch.v36i3.14276
13. Schlatter M, Norris K, Uitvlugt N, DeCou J, Connors R. Improved outcomes in the treatment of gastroschisis using a preformed silo and delayed repair approach. *J Pediatr Surg.* 2003;38(3):459-464. doi:10.1053/jpsu.2003.50079
14. Ross AR, Eaton S, Zani A, Ade-Ajayi N, Pierro A, Hall NJ. The role of preformed silos in the management of infants with gastroschisis: a systematic review and meta-analysis. *Pediatr Surg Int.* 2015;31(5):473-483. doi:10.1007/s00383-015-3691-2
15. Ogasawara Y, Okazaki T, Kato Y, Lane GJ, Yamataka A. Spontaneous sutureless closure of the abdominal wall defect in gastroschisis using a commercial wound retractor system. *Pediatr Surg Int.* 2009;25(11):973-976. doi:10.1007/s00383-009-2450-7
16. Dariel A, Poocharoen W, De Silva N, Pleasants H, Gerstle J. Secondary plastic closure of gastroschisis is associated with a lower incidence of mechanical ventilation. *Eur J Pediatr Surg.* 2014;25(1):34-40. doi:10.1055/s-0034-1395487
17. Andropoulos DB. Effect of anesthesia on the developing brain: infant and fetus. *Fetal Diagn Ther.* 2018;43(1):1-11. doi:10.1159/000475928
18. Gomes Ferreira C, Lacreuse I, Geslin D, et al. Staged gastroschisis closure using Alexis wound retractor: first experiences. *Pediatr Surg Int.* 2014;30(3):305-311. doi:10.1007/s00383-013-3440-3
19. Bhat V, Moront M, Bhandari V. Gastroschisis: a state-of-the-art review. *Children.* 2020;7(12):302. doi:10.3390/children7120302



20. Kunz SN, Tieder JS, Whitlock K, Jackson JC, Avansino JR. Primary fascial closure versus staged closure with silo in patients with gastroschisis: a meta-analysis. *J Pediatr Surg*. 2013;48(4):845-857. doi:10.1016/j.jpedsurg.2013.01.020
21. Aljahdali A, Mohajerani N, Skarsgard ED. Effect of timing of enteral feeding on outcome in gastroschisis. *J Pediatr Surg*. 2013;48(5):971-976. doi:10.1016/j.jpedsurg.2013.02.014
22. Allotey J, Davenport M, Njere I, et al. Benefit of preformed silos in the management of gastroschisis. *Pediatr Surg Int*. 2007;23(11):1065-1069. doi:10.1007/s00383-007-2004-9
23. Hasan S, Rahman A. Role of surgery in the outcome difference of gastroschisis between high-income and lowmiddle- income countries: a review. *Dhaka Shishu Child Hosp J*. 2023;38(2):103-106. doi:10.3329/dshj.v38i2.70595
24. Rahman Mitul A. Surgical neonatal sepsis in developing countries. *J Neonatal Surg*. 2015;4(4):41.
25. Ford K, Poenaru D, Moulot O, et al. Gastroschisis: bellwether for neonatal surgery capacity in low resource settings? *J Pediatr Surg*. 2016; 51(8):1262-1267. doi:10.1016/j.jpedsurg.2016.02.090
26. Carnaghan H, Baud D, Lapidus-Krol E, et al. Effect of gestational age at birth on neonatal outcomes in gastroschisis. *J Pediatr Surg*. 2016;51(5): 734-738. doi:10.1016/j.jpedsurg.2016.02.013
27. Charlesworth P, Njere I, Allotey J, et al. Postnatal outcome in gastroschisis: effect of birth weight and gestational age. *J Pediatr Surg*. 2007;42(5):815-818. doi:10.1016/j.jpedsurg.2006.12.034
28. Oyinloye A, Abubakar A, Wabada S, Oyeboji L. Challenges and outcome of management of gastroschisis at a tertiary institution in North-Eastern Nigeria. *Front Surg*. 2020;7:8. doi:10.3389/FSURG.2020.00008
29. Wesonga AS, Fitzgerald TN, Kabuye R, et al. Gastroschisis in Uganda: opportunities for improved survival. *J Pediatr Surg*. 2016;51(11):1772-1777. doi:10.1016/j.jpedsurg.2016.07.011

Respiratory functions after lung surgeries in children: review of the literature

 Atilla Şenaylı,  Sevgi Ulusoy Tangül

Department of Pediatric Surgery, Faculty of Medicine, Yozgat Bozok University, Yozgat, Türkiye

Received: 17/09/2023

Accepted: 04/03/2024

Published: 10/04/2025

Cite this article: Şenaylı A, Ulusoy Tangül S. Respiratory functions after lung surgeries in children: review of the literature. *Surg Child.* 2025;2(2):60-65.

Corresponding Author: Atilla Şenaylı, atillasenayli@gmail.com

ABSTRACT

In pediatric surgery, the operation of the lung parenchyma tissue is generally performed with limited essential evaluations both before and after the operation. Decisions to be taken are formed around some basic examination methods. For this reason, this review aims to determine the routine and more specific methods in the literature, which are evaluated in pre- and postoperative processes related to lung parenchyma surgeries, and to examine how these methods are applied to which diseases. In the study, pre- and postoperative examination methods of the lung were primarily examined. Later, these evaluations were classified into two separate groups: congenital and acquired diseases. Rare surgeries were excluded from the study. Among the congenital diseases, congenital pulmonary airway malformation, congenital lobar emphysema, and primary ciliary dyskinesia are discussed. Among acquired diseases, infections and cancer are discussed. In general, it has been determined that there is no regular follow-up and no standard practice. The reason for this was that the evaluation processes were not based on a particular algorithm; personal approaches came to the fore, and it was determined that the individual characteristics of the patients were very variable. The literature regarding pre- and postoperative evaluations of patients undergoing lung parenchymal surgery is impoverished. There is an information gap where multicenter systematization studies can be made of the decision-making mechanisms initiated on an institutional basis regarding those who have these surgeries.

Keywords: Lung, function, surgery, children, congenital, acquired

INTRODUCTION

Evaluating lung functions before and after surgeries in children usually needs to be appropriately formed. At first sight, lung surgeries in childhood are not as common as in adulthood. The main reason is the low incidence of lung disease in children. In addition, authors operating on children's lung diseases have their style of pre-, per-, and postoperative. Because of all these facts, concluding the functions of remaining lung tissue after surgery is controversial.¹

There needs to be more studies evaluating lung functions after surgeries to solve this controversy.² There are reasons for this fact. First, infrequent lung operations in the childhood age group need more studies.^{3,4} Second, due to the specific physiological changes at different ages in childhood, postoperative complications may show variations that make postoperative evaluations complex.⁴ Third, physiotherapy procedures performed after surgeries must be standardized.⁴ Since it will affect the level of recovery, physiotherapy standards are essential to make proper evaluations. Finally, different designations of lung surgeries in different institutions may affect the functions and exercise capacity differently.²

Despite foreseeing that pulmonary functions after lung surgeries seem complicated, it is well known that respiratory functions usually reach satisfactory levels.⁵ Satisfying results are revealed at diagnosis, surgery technique, and postoperative care differences.⁵ Nevertheless, a decrease in oxygen saturation and thoracic and lung compliance is expected in young children in the early periods of thoracic surgery.⁴

This study aims to examine the functions of the rest of the lung tissue postoperative in childhood in detail as soon as possible. Organ and tissue surgeries in the thoracic region, such as the esophagus, mediastinum, costae, and diaphragm, were excluded, and only lung parenchymal operations were evaluated.

METHODS

Literature was reviewed to prepare the manuscript. For this purpose, keywords such as "lung," "function," "surgery," "children," "congenital," and "acquired" were used. Nowadays, in English literature, there are 113.000 articles and publications in Google Scholar, but when keyword combination is



evaluated, only 19 articles are found to complete the aim of this study. Other literature in different languages should have been searched. When “surgery” was excluded, 209.000 publications were found, and when “children” was excluded, 241.000 publications were found.

The articles that did not define surgery, lung parenchyma, and evaluation techniques were excluded. Also, articles accomplishing the keywords but consisting of rare problems were excluded. Therefore, a few articles achieved the goal of this study.

First, preoperative and postoperative general aspects were investigated in these articles.

PREOPERATIVE LUNG FUNCTION GENERAL ASPECTS

Predictable postoperative pulmonary function may be necessary for the quality of life of a patient undergoing primary pulmonary resection.⁶ Barikbin et al.⁷ stated that early lung function tests could be helpful when deciding on surgery. In their study, the timing of the operations is essential, and when surgery is delayed, lung functions of sick children would be more limited in those whose operations were delayed.⁷ Also, there were some other studies to calculate the possible loss amount of lung tissue to predict the postoperative conditions of patients. Werner et al.¹ formulated this amount and their predicted loss of function formulation, and this was expressed as Predcorr (corrected prediction)=(predicted for healthy subjectX% of lung remaining)/100.

In another study, carbon monoxide diffusion capacity measurement is a method to calculate the possible surgical resection area.⁶ It is advised that it will be helpful to measure the diffusion capacity of carbon monoxide with FEV₁ in the lung to predict postoperative lung function, especially in lung cancers.⁶

Preoperative radio diagnostic evaluations are essential in our practice, but investigating lung capacity is only sometimes possible due to a lack of staff. There are few pediatric lung disease specialists working in a few amounts of hospitals. As their lung capacity evaluations preoperatively depend primarily on pediatric pulmonology practice, we operated on our patients without this type of support.

POSTOPERATIVE LUNG FUNCTION- GENERAL ASPECTS

Regardless of the method used, one of the most essential conditions to evaluate respiratory function as a standard criterion after surgery is a period without respiratory infection.¹ In addition, attention should be paid to the height and weight of the patient to compare them with average results appropriate for the patient's age.¹ It has been stated that ventilation, respiratory rate, oxygen consumption, carbon dioxide production, and respiratory parameters can be measured with maximal work capacity in patients older than five years of age who are thought to be able to comply with and meet height criteria.⁹

Postoperative evaluations are easier than preoperative assessments. Nevertheless, the principal evaluations are physical examinations, plain chest graphs, and computerized

tomography. If these evaluations are insufficient to find results, then scintigraphy is planned. However, we seldom use other procedures as technical, and staff is generally lacking.

POSTOPERATIVE LUNG FUNCTION MEASUREMENT METHODS

Physical Examination

Physical examinations other than weight and height monitoring should be present in the literature. No scientific study has been found where physical examinations were carried out with long-term follow-up for postoperative lung surgeries.

Chest X-Rays

A chest X-ray is often helpful for understanding the condition of lung diseases before operations.¹¹ Evaluating the chest radiographs may also be helpful for postoperative periods.¹ In this examination, detecting rib changes, pleural reactions, and decreased pulmonary vascularization findings in the lung resection area can be meaningful.¹ Other X-ray examinations that can be performed are bronchography and high-resolution CT (HRCT).¹⁰ Unfortunately, no systematic and explanatory study is present using the evaluation of these lung graphs in the literature.

Chest X-rays are the most used evaluation instrument for experienced surgeons especially. Combining the images with clinical symptoms and findings with X-ray images seems enough for most authors. Numeric findings might be less valuable because of this point of view.

Ventilation-Perfusion Ratio (V/Q)

A V/Q examination is performed to show whether the disease causes lung collapse, consolidation, aplasia, or severe hypoplasia, especially in patients with a closed lung.¹¹ The features and functions of the remaining tissue after lung surgery can also be examined with this method.¹¹ However, it is not a preferred method in the first place. If scintigraphy is chosen for this investigation, it would be better to use Xenon 133 inhalation, especially to examine the ventilation.¹ This method can evaluate breathing, equalization, and wash-out situations.¹ For perfusion, Technetium 99m macro-aggregate is one of the most used methods.¹

In our country, it is only sometimes possible to perform this technique. However, it can be a well-structured follow-up device for these patients where available.

Lung Function Tests

Spirometry results of congenital problems were presented to be significantly lower in those who were operated on than those who were followed up without surgery.⁸ According to another study, reliable spirometry can be performed at the age of 8.⁸ Respiratory capacity measurements can also be performed with portable ergo spirometry systems.²

The measurement of total lung capacity (TLC), forced vital capacity (FVC), and forced expiratory volume in 1st second (FEV₁) are among the indicators of lung growth.^{2,9,12}

When lung capacity is 80% or less or when the FEF25-75% value is below 70%, it is necessary to consider that lung functions are insufficient.^{2,9,12}



When there is a restrictive defect, TLC will be detected below 80%.²

In hyperinflation, the residual volume (RV) will be expected to be greater than 135, and the RV/TLC ratio to be greater than 0.35.²

Performing function tests is restricted to a small age group for pediatric surgery. Some studies reported that children older than eight years old may be evaluated. Children must be older to have optimum results in our regions. Also, being an adolescent sometimes makes cooperation difficult. Therefore, function tests, if performed sparingly, may need to be more conclusive. Another restriction in performing lung function tests is, again, availability. So, routine practice may only be possible for some units.

Cardiopulmonary Exercise (Treadmill) Tests

Treadmill tests can be performed on children whose age is appropriate regarding endurance.^{1,2} In this study, FVC, FEV₁, FEV₂₅₋₇₅%, TLC, RV, and functional residual capacity (FRC) can be evaluated.¹³ Maximum work capacity (power) and maximal oxygen uptake (VO₂-max) are also determined in this process.¹² One of the features that can be evaluated is “durability”.¹ It is evaluated whether there is a moment of desaturation during exercise.¹

During the treadmill procedure, reactions such as heart rate, blood pressure, transcutaneous oxygen saturation, flushing, and sweating must be appraised.¹ The treadmill can be accelerated from 2.7 km/h to 6.0 km/h with a speed increase of 2% per minute.² Comfortable breathing is allowed with a mask suitable for the face.² The procedure can be continued until the heart rate reaches 85% of its maximum value for age.² To calculate the maximum heart rate, the “heart rate=220-age (years)” equation can be used.² These data can be compared according to standard references of gender and body surface area.¹²

For authors, it is unlikely for thoracic and pediatric surgeons to use this technique in preoperative and postoperative evaluations. The main reasons are obtaining the device, sustaining staff, and, for children, the need for age selections.

Nitrogen Wash-out Test

Nitrogen measurements with multiple-breath nitrogen wash-out in 100% oxygen are worthy for assessing respiratory inhomogeneity in preschool children.¹³ Lung clearance index (LCI) of 5%, LCI of 2.5%, average resistances at 8 Hz, and average reactance at 8 Hz are evaluated.¹³

We have no experience with this test. Also, there needs to be more literature on it. Therefore, the nitrogen wash-out test needs much evaluation before clinical practice.

Other Wash-out Techniques

Another method to measure the effect of surgeries on lung volume is to measure heptafluoropropane (HFP) level with multiple breath wash-out (MBWO) techniques.¹⁴ It has been indicated that this method can be used, especially immediately after surgery.¹⁴ HFP-MBWO examination shows the effects on FRC and V_I measurements.¹⁴ Patient-specific trace gases can also be produced from nitrogen, helium, or sulfur hexafluoride.¹⁴

During the evaluation of all these tests, there is no reference information on postoperative respiratory functions for preschool and school-age children, so it seems necessary to use the “global lung function initiatives” scores.¹³ According to these scores, the detection of 80 percent function can be considered normal.¹³

As nitrogen wash-out tests, other wash-outs are hardly available, and they are not used in practice in our country.

POSTOPERATIVE LUNG FUNCTIONS ACCORDING TO DISEASES

Lung functions after operations can be evaluated according to the origin of the diseases. So, they can be classified as postoperative functions of congenital and acquired diseases.

Postoperative Lung Features in Congenital Pulmonary Diseases

Congenital diseases can demonstrate clinical findings in any period of life.³ The noticeable characteristic of this group of diseases is high morbidity and mortality.^{7,13} Depending on their development mechanisms, these diseases may cause clinical progressions varying from preterm labor to cardiopulmonary insufficiency, pulmonary haemorrhage, and pneumothorax.^{7,13}

The controversy becomes complicated if the patient is asymptomatic for some years.⁹ How to treat these congenital diseases is contradictory.⁹ Some centers advocated that the asymptomatic period of life is a disadvantage, and they said that congenital diseases should be operated on in the early period because it is thought that complications such as pulmonary infection, pneumothorax, and malignancy will be prevented.^{9,13} However, most individuals with congenital diseases who underwent surgery in early childhood are not followed up for respiratory functions later in life. So, it will be hard to compare and predict the postoperative functional pulmonary capacity when an asymptomatic patient becomes indicative. Surgeons operate on congenital diseases mainly because of symptoms, lesion sizes, and imaging results.^{7,8} As most patients recover their complaints after surgeries, further functional evaluations are ignored.

Werner et al.¹ emphasized in their article published in 1993 that the stamina in exercise of the remaining lung is essential. They wrote that exercise tolerance is good after lung resections in children.¹ On the contrary, others wrote that exercise tolerance was low in those operated on.⁸ According to those who thought functions were ruined, TLC is lower on body plethysmography after early operations.⁸ In addition, it was shown in the bronchodilator reversibility test that only 2 out of 19 patients included in the study had significant improvement in lung functions.⁸

One problem that Werner et al.¹ found was the decrease in FEV₁, FEV₂₅₋₇₅, and mid-expiratory time in patients operated on for congenital reasons. This situation is also expressed in other similar studies.¹ Some authors have linked this situation to losing the elastic coil property.¹ It is also stated that it can be explained by the cessation of parenchymal growth.¹

A significant disorder was also detected in the perfusion distribution of these patients.¹ It has been stated that this may



be the deterioration of the vascular structure or the formation of dead spaces.¹

Congenital pulmonary diseases were followed up with their clinical wellness and with X-rays in general. As surgeons only sometimes consult these postoperative patients with pediatric thoracic disease specialists, further systematic evaluations were not done in our country.

Congenital Pulmonary Airway Malformations (CPAM)

CPAM is a cystic disease that can be noticed during fetal life and can affect part or all of the lung lobes.¹² It is among the most common causes of congenital pulmonary diseases.¹⁵ It occurs in 1 in 10-35.000 live births.^{12,15} It is primarily fixed in size; or may be reduced.¹² The way it gives clinical symptoms and its course over time is variable.¹⁵ It is generally accepted that symptomatic CPAMs require resection, but there is no consensus for asymptomatic ones.^{12,15} Excision may be recommended to prevent infection, pneumothorax, and, albeit to a lesser extent, to prevent malignancy.^{12,15} Various studies have been carried out in order to perform parenchyma-sparing surgery for better and more effective respiratory recovery in the postoperative period of the patients.⁷ These studies may prevent behavioral, cognitive, and language problems that the neonatal or infant may experience in the future.⁷

In preoperative pulmonary function tests, respiratory compliance and respiratory patterns in CPAM are significantly reduced.⁷ As pressure changes of the esophagus and the chest wall compliance measurements are necessary to find a precise result, it is difficult to measure the compliance of the lungs in patients with pulmonary cyst diseases such as CPAM.⁷ Since chest wall compliance is deficient in newborns compared to lung compliance, it may be convenient in this respect.⁷

Pulmonary compliance usually increases in CPAM after operations.⁷ Suppose improvement in lung functions appears after surgery. In that case, it is controversial whether this is due to the distension of the remaining lung tissues or the increase in the alveoli in the lung parenchyma.¹ In animal models, the number of alveoli increases, and it is also known that alveoli numbers increase in humans.¹ In addition, the increase in the ratio of RV/TLC is predicted to be inexplicable with over-distention alone.¹

The age of surgery is controversial for CPAM. In a study, when newborn and non-neonatal CPAM patients were compared, it was stated that surgery in newborns had a worse prognosis.¹⁵ Nevertheless, in another study, it was said that babies operated on under one year old were better than those operated on older than one year because the alveolar tissue continues to develop.¹³ Finally, a third study showed that age during lobectomy does not affect pulmonary function or maximum work capacity.¹² Nevertheless, in this last study, the authors revealed the same conclusion as others: an increase in lung functions and exercise capacity immediately after surgery, even in the lung that experienced pneumonia.¹² Interestingly, Naito and colleagues¹² found that patients who underwent right-sided lobectomy had a significantly higher TLC than left lobectomy. A study examining respiratory functions showed that while there was a moderate decrease in vital capacity, TLC, and forced expiratory volume (1st second) in long-term follow-ups, the FRC level could be higher than usual.⁷

Werner et al.¹ stated that the “endurance” of all operated patients, regardless of the age of surgery, was good, and their saturation did not decrease during exercises. However, only 2 of 14 patients reached an expected level for pulmonary functions in all criteria.¹

Werner et al.¹ also discussed the weight and the volume of the lost lung part as a primary factor for postoperative improvement. However, further clinical investigations could not be found on weight.

Airway resistance measured by body plethysmography partially increases, but body plethysmography in patients with pulmonary cysts may not be sufficient to measure all compressible gas volume, as stated above.⁷

Blood gas values are insignificant for CPAMs before and after surgery.⁷ These values show that the limitations in the lungs do not cause any impairment in gas exchange.⁷

Despite all these findings, there is little information in the literature about the course of pulmonary functions in CPAM patients who underwent this surgery.¹³ There is no reliable guideline.¹⁵

Congenital Lobar Emphysema

Inadequate parenchymal growth or residual tissue remnants cause air trapping in congenital lobar emphysema.¹ Tissue anomalies are the reason for respiratory function disorders. Therefore, breathing is not homogeneous in patients with congenital lobar emphysema.¹ Very few studies examine the lung's functional properties after congenital lobar emphysema surgeries. One study showed that the lungs increased volume after surgery, and expiratory flow rates decreased.² These entities normalized within two years.² It has been stated that the cause of the hyperinflation may be related to the increase in the RV/TLC ratio.²

Primary Ciliary Dyskinesia

Primary ciliary dyskinesia is often seen with cystic fibrosis. Cystic fibrosis may cause bronchiectasis, so that lung resections may be required.¹⁶ In a multicenter cohort study, Kouis et al.¹⁶ showed that operations due to primary ciliary dyskinesia were mostly in childhood and more in girls. In this study, it was stated that gender is an essential factor.¹⁶ Apart from this, it was seen that there was no difference in the comparisons of FVC, FEV₁, and BMI between patients who were operated on and those who did not.¹⁶ Loss of lung function was observed in both patient populations.¹⁶ *Pseudomonas* infection was detected more in those who had surgery.¹⁶

Rarely, cystic fibrosis may affect only one lobe or limited segments.¹⁷ These patients may benefit from the surgical procedure.¹⁷ Lucas et al.'s¹⁷ study showed that the lung functions of 7 patients with localized primary ciliary dyskinesia operated on were preserved or improved. However, the operations' long-term effects could not be thoroughly evaluated in this study.¹⁷ Some publications show no improvement in lung functions after cystic fibrosis surgeries.¹⁶

Other Diseases

Studies investigating postoperative respiratory functions of bronchopulmonary dysplasia (BPD) and particularly



sequestration cases are scarce. The literature states that the ventilation is normal but slightly reduced perfusion.¹

In those with BPD, it was observed during spirometry that there was a statistically significant decrease in FEV1 values at the age of 9 and 15 years.¹⁸ If the respiratory function is good in the first two years after birth, it can improve.¹⁸

LUNG FEATURES IN OPERATIONS PERFORMED FOR ACQUIRED REASONS

Infections and cancers can be evaluated in this part. Lung operations according to acquired disease with destroyed lungs can be performed at any age.¹⁰ Many authors prefer delayed surgery as much as possible.¹⁹ Generally, 13-14 years of age are considered the best surgery period.¹⁰ Also, being asymptomatic before surgery is a good predictive factor for an effective treatment.¹⁰

Infections

The most common operations for acquired diseases are infectious-based diseases such as bronchiectasis, lung abscess, necrotizing pneumonia, fungal infections, and tuberculosis.^{3,11,19} Factors like foreign body, stricture, and bronchial suppression can be encountered in these diseases, and surgical decisions may be required.¹⁰

The most common lung problem among acquired diseases is bronchiectasis.¹⁹ A bronchiectasis is a clinical form of lung injury mostly related to infection development.³ Publications show no functional benefit in the surgeries of acquired bronchiectasis, but it has been shown that these surgeries reduce the use of antibiotics.¹⁶ Surgery varies from segmentectomy to pneumonectomy in these patients. Indications to perform pneumonectomy must be absolute, such as life-threatening clinical circumstances.¹⁰ In these patients, respiratory functions are, therefore, somewhat restrictive.¹⁰ The advanced enlargement of the remaining lung for compensation always results in some amount of herniation.¹⁰ In long-term follow-up, it was stated that the lungs were still emphysematous, and the lung volume was more extensive than expected.¹⁰ The reason for staying enlarged in this way may be related to the increase in the number of alveoli or the increase in volume.¹⁰ Nevertheless, there is no significant change in FVC values in these patients' postoperative respiratory function tests.³ After surgery, it is reported that the patients continue their lives usually.¹⁰ However, proper lung total excisions have a specific problem. In right pneumonectomy, if the expansion in the opposite lung is significant, this will cause the bronchus to be under pressure by pushing the mediastinum counterclockwise. As a result, respiratory distress and even death may occur.¹⁰ This condition is called "postpneumonectomy syndrome".¹⁰

Surgery can also be performed in case of persistent lobar atelectasis after infection.³ It has been shown that complications resulting in restrictive defects developed in 3 out of 5 patients who were operated due to lung infections.² Hydatid cyst is a type of infectious disease.³ Unfortunately, no specific research examines lung functions after the operations of hydatid cysts in the lungs and persistent lobar atelectasis after infections.

We know that infectious diseases are the most followed-up patients. Although some tests mentioned above, like treadmill, are not used in routine tests like lung functions are used.

The reason for this strongly depends on pediatric thoracic specialists or even on general pediatricians. These health providers perform evaluations of operated infectious patients and non-surgical patients without consultant surgeons.

Lung Cancer

Another acquired problem requiring surgical procedures to the lung tissue is lung cancer.⁶ The stage of cancer is essential to investigate respiratory function. Severe respiratory problems may be seen in low-stage cancer operations due to the mechanisms that affect pulmonary functions.⁶

Studies for lung functions are generally in adult patients. A prospective study demonstrated that lung functions returned to an expected level one month after cancer surgery.⁶ It is also shown in the same study that the functions continue to improve until three months after the operation and after a year. At the same time, the FEV₁ value increased up to 84% and remained constant, and the CO diffusion value increased to 97%.⁶ Recovery is better in those operated with VATS.⁶ Less pain, unaffected respiratory muscles, and no damage to the thoracic wall are the most relevant reasons for postoperative functional differences between VATS and open surgery.⁶

Chemotherapy may affect pulmonary functions. Ten percent of lung cancer patients receiving chemotherapy have pulmonary toxicity.⁶ This toxicity may be due to pneumonitis or interstitial lung disease.⁶ While there was no significant change in FEV₁ values in patients who did not receive adjuvant chemotherapy compared to those who received adjuvant chemotherapy, it was shown that CO diffusion was at 98% in those who did not receive adjuvant chemotherapy and almost did not change at all in adjuvant patients.⁶

Lung cancer is one of the rarest diseases in the pediatric age group. Therefore, finding fewer postoperative evaluation articles for this entity is not surprising. In our country, this result is not different.

CONCLUSION

As respiratory functions improve significantly after surgeries, lung function improvements during the follow-up period do not attract much attention. The examinations are usually done with small groups of patients and using only one technique. As a result, more than the literature is needed to find determinative conclusions. The literature on the subject needs to be increased, and a detailed examination should be done.

In the process leading to the surgery of lung diseases in children, the most significant complaint is shortness of breath and severe impairment in respiratory functions detected with it. Although the clinical condition of the patients generally improves rapidly after surgery, it is stated that some children may not change. However, the evaluation protocol for postoperative lung functions is not standard, and factors such as the patient's height and age may need to be carefully considered in studies with such discourses. As a result, postoperative follow-up protocol must be developed for these children.

The methods used to evaluate respiratory functions in the postoperative period are generally spirometry/whole body plethysmography, forced oscillation technique, and multiple-breath nitrogen wash-out method in 100% oxygen. Some less preferred methods can also be found in the literature.



These techniques can hardly be used in confident children's age groups. So, it seems essential to find a way to ease these laboratory examinations.

ETHICAL DECLARATIONS

Referee Evaluation Process

Externally peer-reviewed.

Conflict of Interest Statement

The authors have no conflicts of interest to declare.

Financial Disclosure

The authors declared that this study has received no financial support.

Author Contributions

All of the authors declare that they have all participated in the design, execution, and analysis of the paper, and that they have approved the final version.

REFERENCES

- Werner HA, Pirie GE, Nadel HR, Fleisher AG, LeBlanc JG. Lung volumes, mechanics, and perfusion after pulmonary resection in infancy. *J Thorac Cardiovasc Surg.* 1993;105(4):737-742. doi:10.1016/S0022-5223(19)34202-3
- Sritippayawan S, Treerojanapon S, Sanguanrungrasirikul S, Deerojanawong J, Prapphal N. Pulmonary function and exercise capacity in children following lung resection surgery. *Ped Surg Int.* 2012;28(12):1183-1188. doi:10.1007/s00383-012-3187-2
- Choudhury SR, Chadha R, Mishra A, Kumar V, Singh V, Dubey NK. Lung resections in children for congenital and acquired lesions. *Ped Surg Int.* 2007;23(9):851-859. doi:10.1007/s00383-007-1940-8
- Kaminski PN, Forgiarini LA Jr, Andrade CF. Early respiratory therapy reduces postoperative atelectasis in children undergoing lung resection. *Respir Care.* 2013;58(5):805-809. doi:10.4187/respcare.01870
- Nakajima C, Kijimoto C, Yokoyama Y, et al. Longitudinal follow-up of pulmonary function after lobectomy in childhood-factors affecting lung growth. *Ped Surg Int.* 1998; 13: 341-345. 1998;13(5-6):341-345. doi:10.1007/s003830050334
- Kim HK, Lee YJ, Han KN, Choi YH. Pulmonary function changes over 1 year after lobectomy in lung cancer. *Respir Care.* 2016;61(3):376-382. doi:10.4187/respcare.04284
- Barikbin, P, Roehr CC, Wilitzki S, et al. Postnatal Lung function in congenital cystic adenomatoid malformation of the lung. *Ann Thorac Surg.* 2015;99(4):1164-1169. doi:10.1016/j.athoracsur.2014.11.018
- Hijkoop A, van Schoonhoven MM, van Rosmalen J, et al. Lung function, exercise tolerance, and physical growth of children with congenital lung malformations at eight years of age. *Ped Pulmonol.* 2019;54(8):1326-1334. doi:10.1002/ppul.24345
- Beres A, Aspirot A, Paris C, et al. A contemporary evaluation of pulmonary function in children undergoing lung resection in infancy. *J Ped Surg.* 2011;46(5):829-832. doi:10.1016/j.jpedsurg.2011.02.012
- Blytha DF, Buckels NJ, Sewsunker R, Soni MA. Pneumonectomy in children. *Eur J Cardiothorac Surg.* 2002;22(4):587-594. doi:10.1016/s1010-7940(02)00404-9
- Gordon I, Helms P. Investigating the small lung: which imaging procedure? *Arch Dis Child.* 1982;57(9):696-701. doi:10.1136/adc.57.9.696
- Naito Y, Beres A, Lapidus-Krol E, Ratjen F, Langer JC. Does earlier lobectomy result in better long-term pulmonary function in children with congenital lung anomalies? A prospective study. *J Ped Surg.* 2012; 47(5):852-856. doi:10.1016/j.jpedsurg.2012.01.037
- Tocchioni F, Lombardi E, Ghionzoli M, Ciardini E, Noccioli B, Messineo A. Long-term lung function in children following lobectomy for congenital lung malformation. *J Ped Surg.* 2017;52(12):1891-1897. doi:10.1016/j.jpedsurg.2017.08.059
- Proquitt H, Freiburger O, Yilmaz S, et al. The effect of surgery on lung volume and conventional monitoring parameters in ventilated newborn infants. *Eur Respir J.* 2010;35(5):1072-1078. doi:10.1183/09031936.00058009
- Dukleska, K, Teeple EA, Cowan SW, Vinocur CD, Berman L. Outcomes in children undergoing surgery for congenital pulmonary airway malformations in the first year of life. *J Am Coll Surg.* 2018;226(3):287-293. doi:10.1016/j.jamcollsurg.2017.12.014
- Kouis P, Goutaki M, Halbeisen FS, et al. Prevalence and course of disease after lung resection in primary ciliary dyskinesia: a cohort & nested case-control study. *Respir Res.* 2019;20(1):212. doi:10.1186/s12931-019-1183-y
- Lucas J, Connett GJ, Lea R, Rolles CJ, Warner JO. Lung resection in cystic fibrosis patients with localized pulmonary disease. *Arch Dis Child.* 1996;74(5):449-451. doi:10.1136/adc.74.5.449
- Filippone M, Bonetto G, Cherubin E, Carraro S, Baraldi E. Childhood course of lung function in survivors of bronchopulmonary dysplasia. *JAMA.* 2009;302(13):1418-1420. doi:10.1001/jama.2009.1419
- Kosar A, Orki A, Kiral H, Demirhan R, Arman B. Pneumonectomy in children for destroyed lung: evaluation of 18 cases. *Ann Thorac Surg.* 2010;89(1):226-231. doi:10.1016/j.athoracsur.2009.10.007

Anesthetic management in pediatric cardiac surgery

 Ayşe Gizem Saraçoğlu,  Nevriye Salman

Department of Anesthesiology and Reanimation, Ankara Bilkent City Hospital, Ankara, Türkiye

Received: 18/09/2024

Accepted: 27/10/2024

Published: 10/04/2025

Cite this article: Saraçoğlu AG, Salman N. Anesthetic management in pediatric cardiac surgery. *Surg Child.* 2025;2(2):66-71.

Corresponding Author: Ayşe Gizem Saraçoğlu, gizemsaracoglu90@gmail.com

ABSTRACT

Pediatric cardiac anesthesia poses significant challenges due to the complexity of congenital heart diseases (CHDs) and the distinct physiological considerations in children. Despite advancements in surgical and anesthetic techniques, many regions still struggle to provide adequate cardiac care, while some areas perform a high volume of pediatric cardiac surgeries annually. This review explores the development and physiology of the cardiovascular system, focusing on congenital heart defects and their impact on anesthesia management. It discusses the roles of cardiopulmonary bypass (CPB) and cardiac catheterization in diagnosis and treatment, as well as essential aspects of preoperative, intraoperative, and postoperative care. Emphasis is placed on effective anesthesia strategies, early extubation, and optimal recovery practices. Ongoing advancements in technology and clinical practice are vital for enhancing safety and outcomes in pediatric cardiac anesthesia.

Keywords: Anesthesia, cardiac, pediatric, congenital heart disease, cardiopulmonary bypass

INTRODUCTION

Pediatric cardiac surgeries pose significant challenges for anesthesiologists due to the complexity of congenital heart diseases (CHDs) and the unique physiological considerations in children. Because of the improvements in surgical techniques, diagnosis, perfusion, and anesthesia; anesthesiologists encounter patients with cardiac diseases more. These patients can have both cardiac and non-cardiac procedures. But still, almost 90% of a million children worldwide can't have adequate medical care.¹ The World Health Organization (WHO) recommends that 300-500 pediatric cardiac surgeries be performed annually per 2 million population.² In our country, over 4.000 pediatric congenital heart surgeries are conducted annually.³

Congenital heart disease, present at birth, includes functional, structural, or positional heart defects. Severe forms can be 12 times more fatal during infancy and occur in 5-14 per 1.000 live births.⁴ Untreated, these defects can lead to serious complications such as infective endocarditis, multi-organ dysfunction, and pulmonary hypertension.² Cardiac anomalies may be genetic, non-genetic, or multifactorial, while acquired cardiac diseases often result from conditions like endomyocardial fibrosis, rheumatic heart disease, Chagas disease, and Kawasaki disease. Without proper medical treatment, these will be fatal, too.⁵

Common congenital heart conditions include hypoplastic left-heart syndrome, tetralogy of Fallot, ventricular septal defect, atrioventricular septal defects, and cardiomyopathy. Patients with these conditions often require both cardiac and non-cardiac procedures.⁶ When administering anesthesia to pediatric patients with CHDs, it is crucial to understand the underlying pathology, the specific procedure, and the effects of anesthetics on the cardiovascular system.

CARDIOVASCULAR DEVELOPMENT

The human embryo relies on uteroplacental diffusion in the first two weeks. After that, the embryo becomes a bilaminar disk that formed from the epiblast and hypoblast. The cardiovascular system is the first to develop, starting in the third week. The embryo transforms into a trilaminar disc through gastrulation, forming all three germ layers; ectoderm, mesoderm, and endoderm. In the epiblast, cardiac progenitor cells migrate to the lateral plate mesoderm cranially and laterally after gastrulation. This is divided into somatic (dorsal) and splanchnic (ventral) layers. The somatic layer generates the pericardium, while the splanchnic layer generates the myocardium, dependent on signals from the endoderm. These embryonic cells are generated not only from the heart mesoderm but also from the cardiac neural crest and proepicardium.⁷

FETAL CIRCULATION

Fetal circulation is significantly different from that of adults. This system helps the fetus obtain oxygenated blood and nutrients from the placenta via two umbilical arteries and one umbilical vein. The lungs are bypassed via the ductus arteriosus, and the liver via the ductus venosus. Blood passes from the right atrium to the left atrium via the foramen ovale. Normal fetal heart rate is 100-160 beats per minute. Fetuses have decreased ventricular filling and reduced contractility compared to adults. After birth, this circulation goes under a rapid change. First, it changes the shunt's way in ductus arteriosus to the left to right and then closes both the shunt and the foramen ovale. If these changes do not occur properly, defects arise.⁸

Congenital heart diseases often result from these shunts not closing properly after birth. These abnormalities can alter blood circulation, leading to cyanotic or acyanotic defects. Cyanotic heart defects arise from right-to-left shunts, where deoxygenated blood bypasses the lungs and enters systemic circulation. Acyanotic heart diseases result from left to right shunts, which occur after birth. If not properly corrected, these shunts can reverse over time, leading to Eisenmenger syndrome (Table 1).⁸

Table 1. Cyanotic and acyanotic heart diseases

Cyanotic heart diseases	Acyanotic heart diseases
Tetralogy of fallot (TOF)	Ventricular septal defect (VSD)
Transposition of the great arteries (TGA)	Atrial septal defect (ASD)
Tricuspid atresia	Patent ductus arteriosus (PDA)
Total anomalous pulmonary venous connection (TAPVC)	Atrioventricular septal defect (AVSD)
Pulmonary atresia	Coarctation of the aorta
Hypoplastic left heart syndrome	Pulmonary stenosis
Ebstein's anomaly	Aortic stenosis
	Mitral valve prolapse

Cardiopulmonary bypass (CPB) is used in major heart or vessel surgeries to isolate the cardiopulmonary system, creating an ideal surgical field. The bypass must fulfill the functions of the heart and lungs, providing oxygen to the blood, removing carbon dioxide, ensuring adequate perfusion to all organs, and minimizing harm.⁹

Some selected cases can be done off pump without the requirement of a CPB as known as beating heart surgery. Although there are more studies on adults, it can be applicable in pediatrics, too (Table 2).¹⁰

Table 2. On-pump and off-pump pediatric cardiac procedures

On-pump cardiac surgeries	Off-pump cardiac surgeries
TOF	PDA
VSD	Coarctation of the aorta
ASD	Blalock-taussing shunt
AVSD	Pulmonary artery banding
TGA	Closed mitral commissurotomy

TOF: Tetralogy of fallot, VSD: Ventricular septal defect, ASD: Atrial septal defect, AVSD: Atrioventricular septal defect, TGA: Transposition of the great arteries, PDA: Patent ductus arteriosus

Cardiac catheterization is used both for diagnostics and treatments, such as repairing atrial or ventricular septal defects, coil embolization of systemic collaterals, addressing coarctation of the aorta, placing stents in stenotic pulmonary vessels, performing balloon valvuloplasty, and conducting radiofrequency ablation. However, less invasive diagnostic methods like echocardiography or magnetic resonance imaging are increasingly preferred. Anesthetists should review these reports to manage patient care effectively.¹¹

PREOPERATIVE CARE

In preoperative care selecting appropriate patients for the team and facility conditions is crucial. The success of perioperative processes depends on this period to foresee potential risks and prepare accordingly. Evaluating the patient's medical history, comorbidities, physical examination, specifics of the cardiac defect, previous surgeries or interventions, and diagnostic tests is essential. Preparation for intra- and postoperative requirements must begin early. It should be remembered that all these processes are multidisciplinary. Routine care such as dental care, immunizations, and medication management should be addressed.⁵

Patients with restricted cardiac reserve may exhibit symptoms like tachypnea, dyspnea, frequent lung infections, feeding difficulties, and sweating, older children may show difficulty in activities with peers, exercise intolerance, tiredness, orthopnea, chest pain, palpitation, or syncope Figure 1.¹² In heart failure, symptoms can include tachypnea, chest retraction, nasal flaring, accessory muscle breathing, pedal edema, jugular vein distention, hepatomegaly, and rales. Peripheral pulses and blood pressure should be checked in all extremities, along with pulse oxygen saturation. Recent upper or lower respiratory infections can cause airway reactivity for 2-8 weeks.¹³

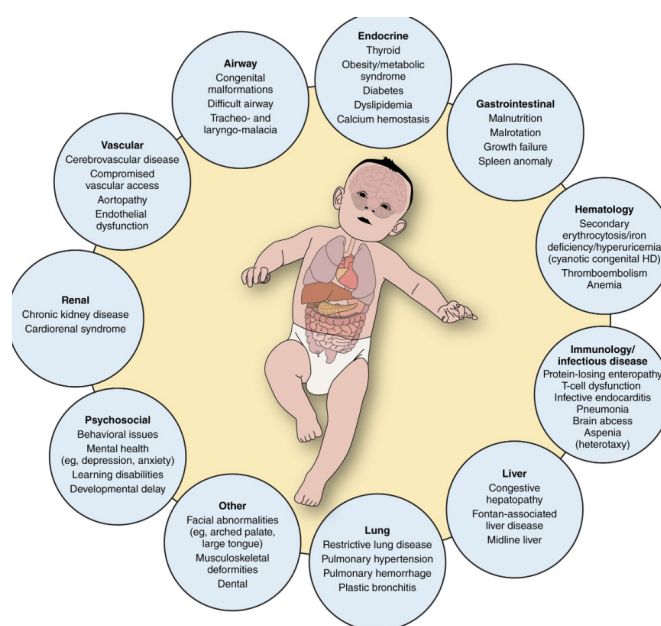


Figure 1. Noncardiac conditions in patients with congenital heart disease
HD: Heart disease

Premedication is important in children to reduce anxiety and ease separation from their parents. But deep sedation should be avoided because it may lead to hypoventilation-related hypercarbia which can result in pulmonary hypertension.



Oxygen saturation should be tracked monitored throughout. Midazolam can be administered at 0.5 mg/kg (max 20 mg) orally or 0.05-0.2 mg/kg intravenously. Alternatives include 50-75 mg/kg pedikloril, ketamine (1-2 mg/kg intravenous, 5 mg/kg oral, 2-5mg/kg intramuscular).¹³ Additionally, dexmedetomidine can be considered. For transnasal administration, the dosage ranges between 0.2 and 4.0 µg/kg. However, a dose of 0.5 µg/kg is typically used in children under 6 months, while 1.0 µg/kg is applicable in older children. For oral administration, a dose of 2 µg/kg is recommended.^{14,15}

INTRAOPERATIVE CARE

To avoid hypothermia, the operating room temperature, heated surgical table pads, and blankets should be prepared before surgery. Catheters, cannulas, perfusion pumps, and appropriate infusion lines should be ready, ensuring air bubbles are removed. Blood products should be prepared in anticipation of potential bleeding.³

In addition to standard ASA monitors (ECG, noninvasive blood pressure, heart rate, peripheral arterial saturation) invasive blood pressure, capnography, central venous pressure catheter, pulmonary artery pressure, and body temperature monitors may be required. Also, cerebral oxygenation (NIRS), bispectral index (BIS), and advanced hemodynamic monitors should be used. In these patients, wide intravenous access is often needed. Ultrasound guidance is mandatory for these procedures. Transesophageal echocardiography is also utilized.

The induction method depends on the child's age: inhalation induction is useful for younger children, while intravenous induction is preferred for older ones. Sevoflurane is the first choice for inhalation induction and maintenance. For IV induction, propofol or ketamine can be used. It should be kept in mind that the altered systemic and pulmonary vascular resistance can affect shunts and cardiac output during this process. Neuromuscular blockers are needed for tracheal intubation, and fentanyl (1-2 µg/kg) can suppress hemodynamic responses.¹³

Remifentanyl can be used for both induction (1.5 µg/kg) and maintenance (0.2-0.5 µg/kg/min, continuously injected by infusion pump), which also allows for fast-track anesthesia. Opioid usage in cardiac anesthesia is preferred for their analgesic effects and minimal impact on hemodynamics.¹⁶

Transesophageal echocardiography is one of the most important cardiovascular imaging methods. Since the esophagus is close to the heart and major vessels, it is a perfect ultrasonic window. It provides more accurate information in many specific diagnostic and catheterization procedures than transthoracic echocardiography (TTE). Results are examined after anesthesia induction, before entering CPB, during surgery, and after coming off CPB. In this way, the need for reoperation is reduced.³

High inspiratory oxygen concentrations can decrease pulmonary vascular resistance and cause an increase in the left to right shunts. It is convenient to use a mixture of oxygen and air.¹³

For arterial cannulation, using 24-gauge catheters for patients under two years, and 22-gauge for those over two years would be appropriate. Central venous catheters typically vary according to weight: a 4 French (4F) double lumen for patients under 5 kg, a 5 French (5F) triple lumen for 5-20 kg, and a 7 French (7F) triple lumen for over 20 kg. Ultrasound guidance would be beneficial for these procedures. Finally, for temperature tracking, a nasopharyngeal probe can be chosen.³

During surgeries involving a CPB machine, it is crucial to maintain an activated clotting time (ACT) of over 480 seconds. Heparin sodium, administered at a dose of 3-4 mg/kg, ensures this. Cardioplegia is administered to arrest the heart, a process that should be repeated systematically over time. Regular monitoring of arterial blood gas analyses, electrolytes, and hemoglobin levels is essential. Hypothermia, cardioplegia, and hemodilution techniques are employed in open-heart surgeries to preserve myocardial viability and facilitate its return to normal function following the removal of the cross-clamp. Research indicates that cooling the myocardium to temperatures between 28-32°C does not disrupt myocardial metabolism and maintains optimal function compared to normothermic conditions (37°C). Cardioplegia not only arrests the heart but also provides essential nutrients that meet the myocardium's energy demands. This approach reduces anaerobic metabolism and protects the heart during ischemic periods. While there is no universal consensus in the literature regarding the best cardioplegia solution, various formulations are used in clinical practice.³

Hypothermia is when body core temperature is below 35°C. In cardiac surgery there are four stages of hypothermia as mild (32°C-35°C), moderate (31-26°C, deep (25-20°C), and profound (<20°C). There are some new approaches where cpb done under normothermia (35°C up to 37°C). In a study on av defects normothermic patients showed lower lactic acidosis, better coagulation results, less inotrop usage, and better postoperative period.¹⁷

In a metaanalysis, there is reduced serum lactate, creatine levels and cpb duration time in normothermic group. However it suggests no strong evidence about adverse events and mortality.¹⁸

It is evident that there is no single approach in medicine. Therefore the temperature management should be done according to selected surgery and patient.

Inotropic agents like epinephrine, milrinone, dobutamine, and dopamine, and vasopressors like norepinephrine, may be needed.¹⁹

To prevent nausea and vomiting, intravenous antiemetics such as dexamethasone (0.2-0.5 mg/kg) or ondansetron (0.1 mg/kg, max 4 mg) can be administered.

Preemptive nerve blocks are used for postoperative analgesia. They additionally reduce intraoperative opioid consumption and enhance hemodynamic stability.²⁰ These blocks also attenuate proinflammatory cytokine release during surgery.²¹ Intercostal nerve blocks are used for this purpose. Various approaches such as pectointercostal fascial block (PIFB) and transversus thoracic plane block (TTPB) have been developed to optimize outcomes. These blocks, administered with ultrasound guidance, target anterior chest wall pain by depositing anesthetic (typically ropivacaine, bupivacaine, or lidocaine) between specific muscle layers—either between the pectoralis major and intercostal muscles (PIFB) or between the internal intercostal and transversus thoracis muscles (TTPB).²² The other blocks that can be beneficial are pectoral nerve (PECS) blocks, erector spinae plane block (ESPB), retrolaminar block (RLB), and paravertebral block (PVB). The PECS block (I and II) targets the pectoral and serratus planes, utilizing agents like ropivacaine or bupivacaine to alleviate pain in the breast and upper chest. The ESPB injects anesthetic (e.g., ropivacaine or bupivacaine) between the erector spinae muscle and transverse processes, offering extensive thoracic analgesia suitable for both acute and chronic pain management. The RLB uses bupivacaine along the vertebral lamina for effective thoracic pain relief, particularly in traumatic cases. Lastly,

the PVB administers anesthetic, such as ropivacaine or levobupivacaine, near the vertebrae for unilateral chest and abdominal wall analgesia, making it ideal for various surgical procedures in pediatric cardiac care.²³

Tracking blood glucose is crucial, especially in infants. It is essential to carefully control fluid infusion rates, which aids in calculating blood loss and managing fluids. Renal failure is a significant concern during these processes, particularly due to the nephrotoxic effects of contrast agents used in catheterization. Risk factors include pre-existing renal disease, diabetes mellitus, heart failure, and concurrent use of other nephrotoxic agents. Despite precautions, renal failure remains a potential complication. To mitigate risks, it is important to avoid dehydration and exercise caution with contrast agents.¹³ However, excessive fluid administration can worsen cardiac function, highlighting the necessity of maintaining a delicate balance.

The choice of fluids is also important **Figure 2**.¹³ Mostly preferred ones in clinical use are crystalloids, balanced crystalloids, and human albumin **Figure 3**.¹³ The administration of HES during surgery is not recommended because it increases the risk of mortality, renal dysfunction, and postoperative bleeding.²⁴

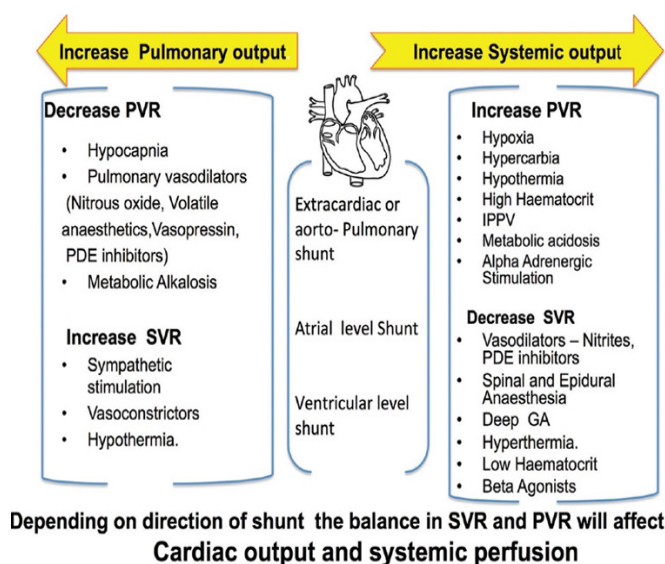


Figure 2. Flow distribution dynamics
PVR: Pulmonary vascular resistance, SVR: Systemic vascular resistance

Anaesthetic drug	SVR	PVR	Dose
Potent volatile agents	↓	↓	0.5-1 MAC
Nitrous oxide	-	↑	
Opioids			
Fentanyl	↓		1-2 µg/kg 0.5-2 µg/kg/h
Morphine	↓		0.05-0.2 mg/kg 0.02-0.2 mg/kg/h
Midazolam	-	-	0.5 mg/kg PO 0.1 mg/kg IV 0.02-0.3 mg/kg/h
Ketamine (with glycopyrrolate)	↑	No effect (if CO ₂ normal)	3-4 mg/kg PO 1-2 mg/kg IV 5-20 µg/kg/min
Propofol	↓	-	2-3 mg/kg 100-300 µg/kg/min
Dexmedetomidine	↑-↓	↓	1 µg/kg (over 10 min) 0.2-1 µg/kg/h

IV – Intravenous; SVR – Systemic vascular resistance; PVR – Pulmonary vascular resistance; MAC – Minimum Alveolar Concentration; ↓ – lowers; ↑ – increases

Figure 3. Anesthetic drug effects and doses

POSTOPERATIVE CARE

After surgery, most patients are transferred to the intensive care unit (ICU) while still intubated, monitored, and receiving continued perfusions. The goals of postoperative care include planning for early extubation, mobilization, and recovery, with a focus on minimizing ICU and hospital stays to reduce morbidity and mortality. Dedicated critical care is crucial at this stage. The impact of postoperative pain cannot be ignored. We know that we deal with opioid-related side effects in some patients. Efficient pain management is essential for enhanced recovery strategies which can improve the outcomes. Intravenous opioid administration is still the main treatment in many practices due to the risk of epidural hematoma or bleeding. Utilizing peripheral nerve blocks can be beneficial in reducing opioid use. Multimodal approaches will help to reduce our concerns. It will also help with intraoperative reduced opioid usage, early extubation, etc. In pediatric cardiac surgery, there is ongoing debate on strategies for enhanced recovery after surgery. The American Association for Thoracic Surgery suggests considering regional anesthesia techniques for optimal outcomes.^{5,25}

It's crucial to recognize that spending extended periods in such environments is particularly challenging for pediatric patients. Therefore, optimizing conditions and minimizing this time are paramount. Early extubation plays a pivotal role in achieving these goals.

Early extubation is associated with improved left heart function post-CPB, as mechanical ventilation can decrease preload during inspiration, increasing intrathoracic pressure.²⁶ Therefore with early extubation, improvement in the blood circulation is a significant outcome after cardiac surgery.²⁷ Fast-track anesthesia facilitates in-room extubation for suitable patients, potentially improving postoperative outcomes. Pulmonary hypertension remains a significant risk factor for postoperative mortality, exacerbated by surgery and anesthesia-induced factors.²⁸ Managing hypoxia, hypercarbia, and acidosis to prevent pulmonary vasoconstriction is crucial. Techniques like applying PEEP (approximately 6 cm H₂O) and alveolar recruitment maneuvers aim to mitigate atelectasis during the perioperative period.²²

Prior to surgery, cardiac catheterization plays a crucial role in diagnosis and disease management. While advancements in techniques and equipment have improved outcomes, understanding and managing relative risks remain essential. Children may require ICU care before or after catheterization, with common adverse effects including the need for mechanical ventilation and hypotension necessitating vasoactive medications.²⁹

In cardiac surgery, antifibrinolytics like tranexamic acid (TXA), ε-aminocaproic acid (EACA), and aprotinin have demonstrated significant benefits. These medications primarily reduce postoperative bleeding and diminish the requirement for blood product transfusions.³⁰ However, there remains a notable gap in large-scale randomized controlled trials investigating the effects and potential side effects of these drugs specifically in pediatric patients.

It should be kept in mind that there can be some complications during these procedures (**Table 3**).³¹

**Table 3.** Cardiac procedure complications

Arrhythmias	• Sepsis
Low cardiac output syndrome	• Coagulopathy
Heart failure	• Anemia
Pericardial effusion or tamponade	• Thrombosis
• Pulmonary hypertension	• Acute kidney injury (AKI)
• Atelectasis	• Electrolyte imbalances
• Respiratory failure	• Necrotizing enterocolitis (NEC)
• Stroke	• Feeding intolerance
• Seizures	• Delayed awakening
• Neurodevelopmental delays	• Airway complications
• Wound infection	• Post-traumatic stress disorder (PTSD)
• Endocarditis	• Developmental delays

CONCLUSION

In pediatric cardiac surgery, the anesthesiologist plays a pivotal role in ensuring optimal outcomes for patients undergoing complex procedures. This specialty faces significant challenges due to the potential for mortality and morbidity in this vulnerable population. A comprehensive understanding of the unique physiological challenges—such as altered hemodynamics, respiratory mechanics, and complications arising from congenital heart defects—is essential. A qualified multidisciplinary team, involving surgeons, cardiologists, and anesthesiologists, is paramount for effectively managing these cases and tailoring the anesthetic plan to each patient's specific needs.

Recommended medications for anesthesia induction and maintenance include agents like propofol, sevoflurane, and fentanyl, each selected for their safety profiles and pharmacokinetic properties in children. Adjuvant medications, such as dexmedetomidine, can enhance sedation and analgesia while reducing opioid requirements, improving patient comfort during and after surgery. Careful selection of neuromuscular blockers is also critical, particularly in patients with pre-existing neuromuscular compromise, to prevent residual paralysis postoperatively.

Effective hemodynamic monitoring during surgery is crucial, as congenital heart defect patients present unique challenges that can impact cardiac output and systemic perfusion. The use of invasive monitoring techniques, such as arterial lines and central venous catheters, allows for accurate assessment and timely intervention when necessary. Anesthesiologists must remain vigilant in recognizing and managing intraoperative complications, including arrhythmias, hypotension, or changes in oxygenation.

Advances in antenatal diagnosis have enabled earlier identification and intervention for these patients, underscoring the importance of comprehensive care and early intervention. The focus must remain on employing the best anesthetic techniques to ensure patient safety and optimal surgical outcomes. Fast-tracking surgery through early extubation and enhancing perioperative pain management with regional blocks are critical strategies for improving recovery and minimizing postoperative discomfort.

In conclusion, the anesthesiologist's involvement in pediatric cardiac surgery extends beyond the operating room to include postoperative care, pain management, and sedation strategies. Continuous evaluation of current literature is essential for staying informed about emerging evidence regarding anesthetic techniques, drug efficacy, and patient safety protocols. By integrating the latest findings with clinical practice, anesthesiologists can enhance the quality of care delivered to this vulnerable patient population, ultimately improving surgical outcomes and long-term prognoses.

ETHICAL DECLARATIONS

Referee Evaluation Process

Externally peer-reviewed.

Conflict of Interest Statement

The authors have no conflicts of interest to declare.

Financial Disclosure

The authors declared that this study has received no financial support.

Author Contributions

All of the authors declare that they have all participated in the design, execution, and analysis of the paper, and that they have approved the final version.

REFERENCES

- Hodges SC, Walker IA, Bosenberg AT. Paediatric anaesthesia in developing countries. *Anaesthesia*. 2007;62(Suppl 1):26-31. doi:10.1111/j.1365-2044.2007.05294.x
- Yacoub MH. Establishing pediatric cardiovascular services in the developing world: a wake-up call. *Circulation*. 2007;116(17):1876-1878. doi:10.1161/CIRCULATIONAHA.107.726265
- Yüzkat N, Çeğin MB, Polat V, Soyoral L, Göktaş U, Kunt AS. Pediatrik Konjenital Kalp Cerrahisinde Anestezi Deneyimlerimiz: İlk Sonuçlar. *GKDA Derg*. 2015;21(4):168-173. doi:10.5222/GKDAD.2015.168
- Tchervenkov CI, Jacobs JP, Bernier PL, et al. The improvement of care for paediatric and congenital cardiac disease across the world: a challenge for the World Society for Pediatric and Congenital Heart Surgery. *Cardiol Young*. 2008;18(Suppl 2):63-69. doi:10.1017/S1047951108002801
- Raj N. Regional anesthesia for sternotomy and bypass-beyond the epidural. *Paediatr Anaesth*. 2019;29(5):519-529. doi:10.1111/pan.13626
- Faraoni D, Zurakowski D, Vo D, et al. Post-operative outcomes in children with and without congenital heart disease undergoing noncardiac surgery. *J Am Coll Cardiol*. 2016;67(7):793-801. doi:10.1016/j.jacc.2015.11.057
- Kussman BD, Miller-Hance WC. Development of the cardiovascular system and nomenclature for congenital heart disease. In: Dean B, Stayer S, Emad B, Wanda C, eds. *Anesthesia for Congenital Heart Disease*. 3rd ed. Houston: Wiley Online Library; 2015.
- Remien K, Majmundar SH. Physiology, fetal circulation. StatPearls Publishing; 2021. Available from: <https://www.ncbi.nlm.nih.gov/books/NBK539710/>
- Nasr VG, DiNardo JA. Cardiopulmonary bypass. In: *The Pediatric Cardiac Anesthesia Handbook*. 1st ed. New York: John Wiley & Sons; 2017.
- Ma J, Li XH, Yan ZX, Liu AJ, Zhang WK, Yang LN. Effect of myocardial protection during beating heart surgery with right sub-axillary approach. *Chin Med J (Engl)*. 2009;122(2):150-152. doi:10.3760/cma.j.issn.0366-6999.2009.02.007
- Öygen Ö, Salık F. Pediatrik kardiyak anestezi. In: Haspolat YK, Baysal Yıldırım Z, eds. *Pediatrik Anestezi*. 1st ed. Ankara: Türkiye Klinikleri; 2023.
- Nasr VG, Markham LW, Clay M, et al. Perioperative considerations for pediatric patients with congenital heart disease presenting for noncardiac procedures: a scientific statement from the American Heart Association. *Circ Cardiovasc Qual Outcomes*. 2023;16(1):e000113. doi:10.1161/HCQ.0000000000000113
- Junghare SW, Desurkar V. Congenital heart diseases and anaesthesia. *Indian J Anaesth*. 2017;61(9):744-752. doi:10.4103/ija.IJA_415_17



14. Kiski D, Malec E, Schmidt C. Use of dexmedetomidine in pediatric cardiac anesthesia. *Curr Opin Anaesthesiol*. 2019;32(3):334-342. doi:10.1097/ACO.0000000000000731
15. Faritus SZ, Khazaee-Koohpar M, Ziyaefard M, Mehrabani MJ. Oral dexmedetomidine versus midazolam as anesthetic premedication in children undergoing congenital heart surgery. *Anesth Pain Med*. 2015; 5(3):e25032. doi:10.5812/aapm.5(3)2015.25032
16. Huang Q, Lin LY, Lin XZ. Comparison of remifentanyl-based fast-track and fentanyl-based routine cardiac anesthesia for intraoperative device closure of atrial septal defect (ASD) in pediatric patients. *Med Sci Monit*. 2019;25:1187-1193. doi:10.12659/MSM.913387
17. Amer GF, Elawady MS, ElDerie A, Sanad M. Normothermia versus hypothermia during cardiopulmonary bypass in cases of repair of atrioventricular septal defect. *Anesth Essays Res*. 2020;14(1):112-118. doi: 10.4103/aer.AER_123_19
18. Xiong T, Pu L, Ma YF, et al. Safety of normothermic cardiopulmonary bypass in pediatric cardiac surgery: a system review and meta-analysis. *Front Pediatr*. 2021;9:757551. doi:10.3389/fped.2021.757551
19. Baehner T, Kiefer N, Ghamari S, et al. A national survey: current clinical practice in pediatric anesthesia for congenital heart surgery. *World J Pediatr Congenit Heart Surg*. 2020;11(3):257-264. doi:10.1177/2150135120902122
20. Padala S, Badhe AS, Parida S, Jha AK. Comparison of preincisional and postincisional parasternal intercostal block on postoperative pain in cardiac surgery. *J Card Surg*. 2020;35(7):1525-1530. doi:10.1111/jocs.14651
21. Bloc S, Perot BP, Gibert H, et al. Efficacy of parasternal block to decrease intraoperative opioid use in coronary artery bypass surgery via sternotomy: a randomized controlled trial. *Reg Anesth Pain Med*. 2021; 46(8):671-678. doi:10.1136/rapm-2020-102207
22. Yamamoto T, Schindler E. Regional anesthesia as part of enhanced recovery strategies in pediatric cardiac surgery. *Curr Opin Anaesthesiol*. 2023;36(3):324-333. doi:10.1097/ACO.0000000000001262
23. Demir AZ, Ozgok A, Balci E, Karaca OG, Şimşek E, Günaydin S. Preoperative ultrasound-guided bilateral thoracic erector spinae plane block within an enhanced recovery program is associated with decreased intraoperative lactate levels in cardiac surgery. *Perfusion*. 2024;39(2):324-333. doi:10.1177/02676591221140754
24. Rizza A, Romagnoli S, Ricci Z. Fluid status assessment and management during the perioperative phase in pediatric cardiac surgery patients. *J Cardiothorac Vasc Anesth*. 2016;30(4):1085-1093. doi:10.1053/j.jvca.2015.11.007
25. Mitnacht AJC, Shariat A, Weiner MM, et al. Regional techniques for cardiac and cardiac-related procedures. *J Cardiothorac Vasc Anesth*. 2019;33(2):532-546. doi:10.1053/j.jvca.2018.09.017
26. Mahle WT, Jacobs JP, Jacobs ML, et al. Early extubation after repair of tetralogy of Fallot and the Fontan procedure: an analysis of the society of thoracic surgeons congenital heart surgery database. *Ann Thorac Surg*. 2016;102(3):850-858. doi:10.1016/j.athoracsur.2016.03.013
27. Ovroutski S, Kramer P, Nordmeyer S, et al. Early extubation is associated with improved early outcome after extracardiac total cavopulmonary connection independently of duration of cardiopulmonary bypass. *Eur J Cardiothorac Surg*. 2018;54(5):953-958. doi:10.1093/ejcts/ezy179
28. van der Griend BF, Lister NA, McKenzie IM, et al. Postoperative mortality in children after 101,885 anesthetics at a tertiary pediatric hospital. *Anesth Analg*. 2011;112(6):1440-1447. doi:10.1213/ANE.0b013e318213be52
29. Stein ML, Bilal MB, Faraoni D, et al. Selected 2022 highlights in congenital cardiac anesthesia. *J Cardiothorac Vasc Anesth*. 2023;37(7): 1095-1100. doi:10.1053/j.jvca.2023.03.032
30. Schertz K, Karam O, Demetres M, Mayadunna S, Faraoni D, Nellis ME. Prophylactic use of antifibrinolytics during pediatric cardiac surgery with cardiopulmonary bypass on postoperative bleeding and transfusion: a systematic review and meta-analysis. *Pediatr Crit Care Med*. 2022;23(11):e517-e529. doi:10.1097/PCC.0000000000003049
31. Murni IK, Djer MM, Yanuarso PB, et al. Outcome of pediatric cardiac surgery and predictors of major complication in a developing country. *Ann Pediatr Cardiol*. 2019;12(1):38-44. doi:10.4103/apc.APC_146_17

Chest wall deformities: diagnosis, classification, and treatment approaches

 Ayşe Uğurum Yüccemen¹,  Bülent Mustafa Yenigün²,  Cihan Genco Çetinkanat³

¹Department of Thoracic Surgery, Sivas Numune Hospital, Sivas, Türkiye

²Department of Thoracic Surgery, Faculty of Medicine, Ankara University, Ankara, Türkiye

³Department of Thoracic Surgery, Citrusmed Ltd. Co., Ankara, Türkiye

Received: 19/11/2024

Accepted: 18/01/2025

Published: 10/04/2025

Cite this article: Uğurum Yüccemen A, Yenigün BM, Çetinkanat CG. Chest wall deformities: diagnosis, classification, and treatment approaches. *Surg Child.* 2025;2(2):72-78.

Corresponding Author: Cihan Genco Çetinkanat, genco.cetinkanat@citrusmed.com.tr

ABSTRACT

Chest wall deformities encompass a range of structural abnormalities that may be congenital or acquired, impacting various aspects of the thoracic structure and function. This document provides an extensive review of primary chest wall deformities, including pectus excavatum (PE), pectus carinatum (PC), Poland syndrome (PS), Jeune syndrome (JS), and sternal cleft (SC). PE, the most prevalent deformity, is characterized by the depression of the anterior chest wall and is often linked to genetic syndromes such as Marfan and Noonan. Its clinical implications, such as exercise intolerance and cardiovascular effects, necessitate careful evaluation through physical examination, imaging, and pulmonary function tests. For severe cases, the Nuss and Ravitch surgical procedures offer corrective options, each with specific indications, benefits, and complications. PC is typified by the protrusion of the sternum and is generally considered a cosmetic issue. However, in severe cases, patients may experience functional impairments. Conservative treatment, including a dynamic compression system, is suitable for mild to moderate cases, while severe cases may require surgical intervention via the modified Ravitch or Abramson procedures. PS involves unilateral absence or hypoplasia of the pectoralis major muscle and may affect rib structures. Treatment focuses on both functional and cosmetic correction, with surgical options including custom implants and reconstructive techniques. JS, primarily congenital, leads to a restricted thoracic volume, posing a significant respiratory risk. Surgical expansion of the thorax is critical in severe cases, utilizing methods like rib grafting and vertical expandable titanium rib placement. SC, a rare condition with incomplete fusion of the sternum, requires early surgical intervention to prevent complications like respiratory distress and mediastinal shift. Across these conditions, individualized treatment plans are essential, balancing surgical benefits with risks, and considering each patient's unique anatomical, functional, and psychosocial needs. The complexity of chest wall deformities necessitates multidisciplinary management to optimize outcomes and enhance quality of life for affected individuals.

Keywords: Pectus excavatum, pectus carinatum, Poland syndrome, Jeune syndrome, sternal cleft, thoracic surgery techniques

INTRODUCTION

Chest wall deformity can be seen in one-third of live births. There is a possibility that the deformity will progress during growth. Nuss stated that patients lose their chance of treatment because of the belief of pediatricians that “There is no need to worry, it will resolve itself over time” for these deformities.¹

Chest wall abnormalities may be either congenital or acquired. They are categorized into three main groups. The major conditions are sternal depression (pectus excavatum) and sternal protrusion (pectus carinatum), caused by an imbalance in the development of the costochondral region. The second group includes anomalies that disrupt the normal development

of the chest wall, including bifid sternum, pentalogy of Cantrell, and Poland syndrome (PS). The last group includes defects that develop due to trauma.¹⁻³

PECTUS EXCAVATUM

Pectus excavatum (PE) is a malformation of the anterior chest wall marked by variable degrees of sternal depression. Its prevalence is 1 in every 400 to 1000 live births^{4,5}, with a higher incidence in males than females.⁶ The deformity typically manifests in the early years of life and tends to worsen over time.⁷

Several hypotheses exist regarding the pathogenesis of PE, but none have received conclusive proof.⁸ It is thought that the deformity may arise from an imbalance in the development of the costochondral regions.

PE is generally sporadic⁹, nevertheless, genetic factors likely play a significant role, as familial inheritance has been sometimes shown.^{10,11} PE is also linked to connective tissue disorders (e.g., Marfan syndrome), genetic syndromes (e.g., Noonan syndrome, Turner syndrome), and neuromuscular diseases (e.g., spinal muscular atrophy).^{6,12,13}

Diagnosis

The most common symptoms in patients with PE include exercise intolerance, chest pain, poor endurance, and shortness of breath. As a result of distortion and displacement of the heart, stroke volume may decrease, and tachycardia may occur.¹⁴ When evaluating these patients, consider the severity of the deformity, its impact on the cardiopulmonary system, and psychosocial factors.

During the physical examination, the degree of sternum depression is determined by inspection. The deformity may be either centralized or asymmetric. In asymmetric cases, the sternum is often rotated, and breast hypoplasia can be observed in female patients. 10–39% of patients with PE report having scoliosis.⁵ Patients with respiratory complaints or those being considered for surgery should undergo pulmonary function testing. Patients with suspected heart issues should undergo a cardiac evaluation, which includes exercise testing, electrocardiography, and echocardiography. Patients who are not candidates for surgery should be followed up periodically, especially during adolescence.

For imaging, anteroposterior and lateral chest radiographs are typically obtained. In patients with moderate or severe PE and/or cardiopulmonary symptoms, computed tomography (CT) is used to assess the severity of the deformity. However, it is noted that symptom severity correlates poorly with the degree of deformity seen on CT.¹⁵

The Haller index (HI), also known as the pectus severity index, defines the depth of the defect. It is calculated by dividing the lateral diameter (A-B) of the chest by the distance between the sternum and vertebra at the point of maximum depression (C-D) on inspiratory CT (**Figure 1a**).¹⁶ A normal HI is equal to or less than 2.5.¹⁷ PE correction index (PCI), a horizontal line is drawn from the front of the vertebra. Then, the maximum distance (AB) between the inner edge of the most anterior part of the rib cage and the horizontal line is measured. The minimum distance (CD) between the inner edge of the most depressed point of the sternum and the horizontal line is measured (**Figure 1b**). The percentage of the patient's missing chest depth is calculated using the formula ((maximum distance - minimum distance)/maximum distance)x100.¹⁸

Patients with moderate to severe PE or respiratory symptoms should undergo pulmonary function testing. Although many patients with PE report respiratory symptoms, less than one-third have abnormal pulmonary function. There is little link between symptoms and pulmonary function.^{19,20} Patients with moderate to severe PE or significant respiratory symptoms should undergo exercise testing, as it is more sensitive in detecting cardiopulmonary abnormalities.^{9,21} Cardiovascular abnormalities are prevalent in individuals with PE and related

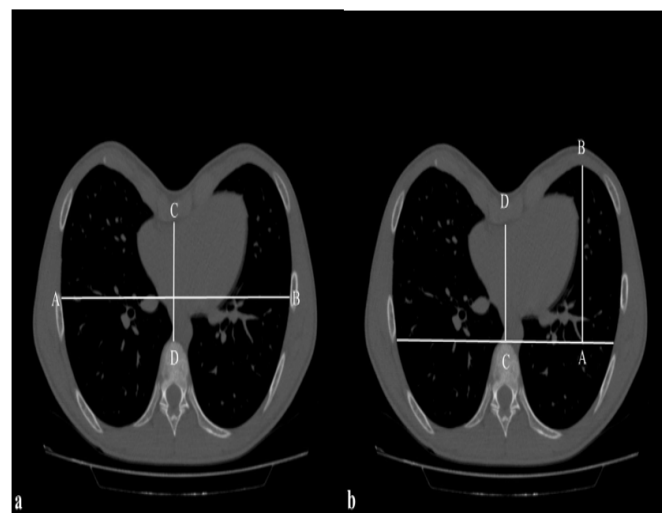


Figure 1. (a) Haller index: AB/CD. (b) Pectus excavatum correction index: ((AB-CD)/AB)x100

disorders.¹² Functional systolic murmurs may be observed in 18% of cases, while mitral valve prolapse occurs in 7-20% of instances.^{5,6}

Kelly's criteria state that surgery is indicated for symptomatic and severe cases of PE if at least two of the following criteria are met: HI>3.25, cardiac and/or pulmonary compression demonstrated by CT or ECHO, restrictive breathing pattern in pulmonary function tests, presence of mitral valve prolapse or arrhythmia, history of unsuccessful corrective surgery, and severe discomfort with body image.²²⁻²⁴

Treatment

Depending on the severity of the malformation, treatment may be conservative or surgical. Surgeons frequently recommend surgical intervention prior to the complete ossification of the chest wall, ideally during late childhood or early adolescence.²³ In mild cases, or preoperatively in severe cases, vacuum bell therapy may be applied. Studies have shown that applying a vacuum bell can reduce the defect.²⁵⁻²⁷

The Ravitch procedure is an open technique. The intervention is performed by a transverse or vertical incision/ sternotomy incision on the sternum. The pectoralis major muscle is dissected to reveal the sternum. The defective cartilages (except for the second and third) are bilaterally resected subperichondrially (**Figure 2**). If the bony segment is affected, subperiosteal resection is also performed. Intercostal bands are freed, and the xiphoid process is resected. The sternum is mobilized from the xiphoid to the angle of Louis. The posterior manubrium and anterior corpus receive transverse osteotomy and wedge resection. The removed segments are placed in the osteotomy lines, bringing the sternum into its normal anatomical position. Kirschner wires are passed beneath the sternum and fixed to both sides of the chest wall.²⁸

The most commonly used technique is the minimally invasive repair of PE (MIRPE), also known as the Nuss procedure. The points where the chest begins to depress inward are marked bilaterally as thoracic entry and exit at the most depressed point of the deformity. In addition, intercostal spaces at the intersection of the same line with the anterior axillary line bilaterally are marked as the incision site. The distance between both anterior axillary lines is measured to determine the length of the bar. The aluminum bar used as a mold is given the



Figure 2. The intraoperative view of subperichondrially resected abnormal cartilages. (Resource: Department of Thoracic Surgery, Ankara University Faculty of Medicine, Ankara, Türkiye)

desired shape of the rib cage. Incisions are made at the marked intercostal spaces along the bilateral anterior axillary lines. A thoracoscope is inserted through the right hemithorax, and then intrathoracic air is infused. A tunnel is created over the ribs. A thoracoscope assists in passing the introducer through the parasternal intercostal space from the right hemithorax, advancing it over the pericardium, and removing it from the opposite hemithorax. The chest wall is shaped by lifting the introducer from both ends. The nylon type attached to the introducer is pulled and passed through the tunnel. Then the nylon type is removed from the introducer and attached to the pectus bar. The nylon-type fixed bar is passed through the tunnel with its convexity facing backward. The pectus bar is rotated 180 degrees and attached to a stabilizer on the left side. The bar is fixed to the ribs with absorbable monofilament pericostal sutures on the right side. The bar is removed between 2-4 years.²⁴

Postoperatively, patients who receive the Nuss surgery generally report greater pain compared to those who undergo the Ravitch operation.²⁹ Secondary thoracic dystrophy is the most serious complication after the Ravitch procedure.³⁰ Pneumothorax and recurrence are other potential complications.³¹

During MIRPE, rare intraoperative complications include injuries to the heart, major vascular structures, and lungs, as well as severe bleeding. Early postoperative complications include spontaneous pneumothorax, pneumothorax requiring drainage, temporary Horner syndrome due to epidural analgesia, wound infection, pneumonia, pericarditis, and pleural effusion. Late complications (after one month) include bar displacement, overcorrection, allergic reactions to the metal bar, severe wound infections, pericardial effusion and pericarditis, and persistence of the deformity.^{6,32-36}

PECTUS CARINATUM

Pectus carinatum (PC), also referred to as pigeon chest, is characterized by the protrusion of the sternum and costal cartilage.³⁷ It is more common in males than females.⁴ Abnormal costal cartilage and sternum growth are believed to be the pathogenesis of PC, although the exact cause is unknown.^{38,39} PC may be associated with various genetic syndromes and disorders affecting connective tissue.⁴⁰

Classification is based on the anatomical region where the protrusion is most prominent. In chondro-gladiolar deformity, the middle and lower parts of the sternum show protrusion along with the costal cartilages, which may be symmetrical or asymmetrical. The costo-manubrial deformity, also known as pectus arcuatum (PA), is characterized by protrusion of the manubriosternal junction and adjacent costae.⁴¹ The mixed type exhibits both PC and PE.

There is a special and rare form of the costo-manubrial variant called Currarino Silverman. Premature fusion and ossification of the manubriosternal joint and sternal segments cause this condition. The distal third of the sternum is short and thick, the 2nd to 5th ribs are angulated, and there is a high arcuatum appearance on the chest.⁴²

Diagnosis

Typical PC deformity is usually recognized in early adolescence and worsens over time.⁴³ Patients are usually asymptomatic; however, symptoms such as respiratory problems and exercise intolerance may be observed in severe cases.⁴¹ In untreated cases with isolated PC deformity, morbidity or mortality due to deformity is usually not observed.⁴⁴

The anteroposterior diameter of the chest is enlarged, and chest wall mobility is typically restricted during physical examination.⁴¹ The type and symmetry of the deformity are determined. It is checked whether the deformity improves when pressure is applied to the deformity. Direct radiographs are used to assess the severity of the deformity, and CT is performed on patients for whom surgery is planned.

Patients with PC typically have normal cardiovascular function. However, genetic syndromes associated with PC may increase the risk of mitral valve prolapse and aortic root dilatation, thereby recommending evaluation with ECHO.⁴⁵

Treatment

PC is considered more of a cosmetic problem, and therefore there are no precise criteria for treatment. The patient's age, the severity, and the progression of the deformity determine the treatment options and timing.

Conservative treatment can be applied in mild and moderate cases. This treatment uses a dynamic compression system to correct the deformity by applying pressure to the rib cage before bone development is complete.⁴⁶ This method is applied for 12-18 months at daily intervals. Disadvantages include the fact that it cannot be used in adult patients and difficulty in patient compliance with treatment.⁴⁷ In order to increase treatment compliance, treatment can be started in the early period of puberty. This method is not effective in cases of P.⁴⁸

Surgical treatment may be more effective in patients with severe PC, PA, moderate or severe sternal asymmetry, loss of chest wall elasticity, or in cases that do not comply with conservative treatment.⁴⁹

The modified Ravitch procedure serves as the open surgical method for PC treatment. This procedure involves the resection of costal cartilages with abnormal angulation, while maintaining the perichondrium intact. A wedge osteotomy is performed on the protruding part of the sternum, and the resected portion is utilized as a graft to remodel the sternum. Absorbable or synthetic materials may also be used as grafts.⁵⁰



The minimally invasive repair method is known as the Abramson procedure or modified Nuss procedure. In this method, an aluminum bar of appropriate size is shaped by applying pressure to the sternum at the point where the sternum is most protruding. The pectus bar is also shaped according to the aluminum bar. The intercostal space is reached through incisions made in the bilateral mid-axillary lines on the line where the protrusion is most prominent. Subperiosteally, two consecutive ribs are prepared, and fixation plates are fixed with pericostal wires. In this method, the pleural cavity is not entered. A chest tube with a trocar is passed through the tunnel opened in the presternal region. The trocar is removed, and the pectus bar is passed through the tube. The drain is retracted through the tunnel together with the bar. The bar is fixed to stabilizers on both sides and presses the sternum. The bar is usually removed after two years. Complications of this procedure include skin adhesion to the bar, seroma, bar breakage, infection, pain, and pneumothorax.⁵¹

POLAND SYNDROME

The absence of the sternocostal head of the pectoralis major muscle and hypoplasia or absence of the pectoralis minor muscle characterizes PS. The syndrome may also include aplasia or hypoplasia of the second to fifth costal cartilages, hypoplasia or absence of the breast and nipple, and ipsilateral brachydactyly and syndactyly. The presence and severity of the components are variable.⁴

PS is seen in 1/10.000 to 1/100.000 live births.⁵² Most cases are sporadic, with rare reports of familial cases.^{53,54} Although its etiology is still unclear, there is a hypothesis that it may have developed due to damage to the subclavian artery.⁵⁴

In 2018, Romanini et al.⁵⁵ proposed a classification of PS to precisely identify patients, as the phenotype is significantly distinct. Type 1, also called the minimal form, has only pectoral muscle deficiency. Upper extremity or costal anomalies accompany the pectoral muscle defect in type 2, the partial form. Type 2a classifies upper extremity anomalies, while type 2b classifies costal anomalies. Type 3 represents the complete form, where both upper extremity and costal anomalies accompany pectoral muscle deficiency.⁵⁵

In 2016, Romanini et al.⁵⁶ proposed the TBN classification to grade the severity of the defect, taking into account thoracic defects separately. T stands for thoracic defects, B for breast defects, and N for nipple-areola complex defect.

Diagnosis

These patients may have respiratory distress due to paradoxical respiratory movement, lung herniation, rarely spontaneous pneumothorax, bullous emphysema, and congenital diaphragmatic hernia.⁵⁷

The most characteristic feature of PS is a partial or complete absence of the pectoralis major muscle.⁵⁴ Ultrasound is used to evaluate the pectoralis major muscle, breast tissue, and costae. Direct radiographs can be taken for costal anomalies; CT and 3D reconstructed CT are preferred in patients with planned surgery.⁵⁵

Furthermore, for surgical planning in patients, a detailed evaluation of the internal thoracic artery is required.⁵⁸ ECHO

and abdominal USG are recommended in terms of other accompanying pathologies.⁵⁵

Treatment

Thoracic defects in PS are repaired for functional and cosmetic reasons.⁵⁴ Studies have shown that performing surgeries during the growth period improves quality of life more.⁵⁹ Surgical intervention may be performed in the presence of asymmetric depression in the chest wall, paradoxical respiration, poor protection of the heart and lungs, the presence of breast hypoplasia or aplasia in female patients, and cosmetic concerns.⁵³

A midline anterior thoracic incision is made for anterior chest wall defects; in addition to this short incision, additional incisions can be made in the axillary line for costal anomalies. This incision allows for the resection of the PC-forming cartilages on the opposite side of the PS. Sternal osteotomy can be added if necessary. Cases with a single costal anomaly do not require surgery, whereas Gore-Tex mesh is used in cases with two costal anomalies and titanium bars are used in cases with two or more costal anomalies. Gore-Tex mesh should be placed between the titanium bar and the skin. A tissue expander or pectoral/breast prosthesis can be placed over this mesh layer. In recent years, CT-scanned and 3D-printed models of the rib cage have been used through these surgical incisions. If PS is accompanied by PE and PC, the classic PC or PE treatment is used.^{55,60}

JEUNE SYNDROME

Jeune syndrome (JS) may be congenital or acquired. Acquired JS develops due to the loss of cartilage growth centers following excessive resection during prepubertal chest wall reconstruction surgeries. Congenital JS, also known as asphyctic thoracic dystrophy, is an autosomal recessive polychondrodysplasia. The incidence is between 1/100.000 and 1/130.000. In this syndrome, the costae are short and horizontal, and the costochondral joints are irregular. The thorax is narrowed and has reduced elasticity, resulting in decreased thoracic volume and reduced lung ventilation.

The severe form is characterized by pulmonary hypoplasia and pulmonary hypertension and presents soon after birth. The moderate form is seen in the first years after birth. Both forms are associated with respiratory failure and pneumonia. In the mild form, patients may have low exertional capacity, and their respiratory problems tend to decrease as they reach adolescence. There are also patients with normal lung function.^{61,62}

Diagnosis

Other characteristic features of the syndrome include short arms and short legs. Kidney, liver, pancreatic, and ocular abnormalities may occur.^{61,62} The diagnosis is made with clinical and radiologic findings. CT with 3-dimensional reconstruction is useful for surgical planning. ECHO and cardiologic evaluations are essential. Pulmonary hypertension and cor pulmonale may result from respiratory failure, making surgery contraindicated.

Treatment

Surgery for JS should be performed soon after birth or in early childhood. However, for stable patients and mild forms,

it is thought that waiting a few years to allow the costae to develop may increase the success of surgery. The aim of surgical treatment is to increase thoracic volume by expanding the thorax. The presence of recurrent respiratory infections, respiratory distress, growth retardation, and pulmonary hypertension determines the indication and timing of surgery.

There are various approaches to surgery.^{63,64} One technique achieves thoracic expansion by placing retractors or grafts after a median sternotomy. The lateral thoracic expansion (LTE) technique involves performing a thoracotomy and splitting the 4th to 9th ribs at their midpoints. The ribs to be operated on are dissected free from the surrounding muscular tissue. The 5th and 6th, as well as the 7th and 8th ribs, are then cut and approximated towards the midline, where they are fixed with titanium bars. This method provides two longer, oblique ribs instead of two horizontal ones.⁶⁵ Davis recommends conducting a second surgery on the contralateral hemithorax after 6 months⁶⁵, but some authors prefer bilateral surgery in the same session.⁶⁶ Another technique, vertical expandable titanium rib (VEPTR), allows for vertical rib cage expansion. In this procedure, a titanium bar is placed vertically and secured to the ribs with locking cradles at both the top and bottom. The apparatus adapts to the patient's growth by sliding along the ribs.⁶⁷

STERNAL CLEFT

Sternal cleft (SC) is a partial or complete fusion defect of the two halves of the sternum.⁵³ SC appears in 1 in 50.000–100.000 live births.⁶⁸ The etiology of SC is unknown. There are 4 main types, including superior and inferior SC, subtotal, and total SC. Superior SC with cleft mandible and median SC are rare types. The most common forms are superior and subtotal.⁵³ Inferior SC is frequently associated with Cantrell pentalogy.⁶⁹ Sternal defects are classified as thoracic ectopia cordis, cervical ectopia cordis, thoracoabdominal ectopia cordis (Cantrell pentalogy), and SC.⁷⁰

Diagnosis

USG can diagnose SC during the intrauterine period, and magnetic resonance imaging (MRI) confirms the diagnosis. Fetal ECHO should also be performed for the presence of concomitant Cantrell's pentalogy.⁶⁹ Direct radiography, CT, and 3D reconstructed CT are used in patients with a surgical plan.

Treatment

Surgical intervention in the treatment of SC may be performed for reasons such as paradoxical respiratory movement and related complications (mediastinal shift, right ventricular overload, arrhythmia), dyspnea, frequent respiratory tract infections, vulnerability of the heart and great vessels to trauma, risk of dermopericardial fistula development, possibility of enlargement of the defect over time, and cosmetic discomfort caused by the external view of the heartbeat.⁷¹ The most suitable time for surgery is the neonatal period, when the chest wall is most flexible and the risk of compression of the underlying structures is minimal.

In SC surgery, intervention is performed by making an anterior thoracic incision along the midline. A V-shaped incision is made in the intact sternum to enable primary repair in cases

with partial SC (Figure 3a). The sternal halves are approximated using nonabsorbable sutures (Figure 3b, 3c). Approximation during primary repair may cause increased mediastinal pressure, which could compromise venous return and cause hemodynamic disturbances.^{72,73} To minimize this risk, Torre et al.⁷² recommend complete or partial removal of the thymus. In cases of large defects or in older patients where the sternal halves cannot be easily approximated due to poor compliance, multiple chondrotomies may be performed to ease the closure and reduce mediastinal pressure. Periosteal flaps, dissected from the sternal bars, are rotated inward and sutured at the midline to form the posterior wall of the newly reconstructed sternum. Any residual gap can be filled with cartilage grafts or, alternatively, synthetic prostheses. If primary repair cannot be achieved, materials such as Gore-Tex, or bone grafts can be used to bridge the gap.^{10,74,75}

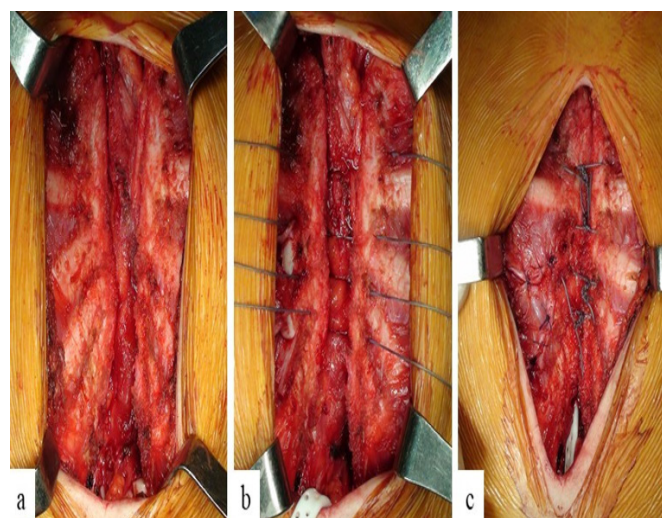


Figure 3. The intraoperative view of sternal cleft. (a) The view of separated sternal halves. (b) Sternal halves were prepared for primary repair. (c) Both sternal halves were sutured with interrupted nonabsorbable sutures. (Resource: Department of Thoracic Surgery, Faculty of Medicine, Ankara University Ankara, Türkiye)

CONCLUSION

Chest wall deformities present with varying degrees of functional and psychological challenges. There is a weak correlation between the severity of the deformity and the symptoms. Therefore, each patient should be evaluated individually, and a treatment plan should be developed through a multidisciplinary approach, considering both conservative and surgical options. Recent advancements in minimally invasive techniques and prosthetic applications have contributed to reduced complications and improved cosmetic and functional outcomes.

ETHICAL DECLARATIONS

Referee Evaluation Process

Externally peer-reviewed.

Conflict of Interest Statement

The authors have no conflicts of interest to declare.

Financial Disclosure

The authors declared that this study has received no financial support.

Author Contributions

All of the authors declare that they have all participated in the design, execution, and analysis of the paper, and that they have approved the final version.

REFERENCES

- Nuss D, Kelly RE. Chest wall deformities. In: Puri P, Höllwarth M, eds. *Pediatric Surgery*. Springer; 2009. doi:10.1007/978-3-540-69560-8_25
- Shamberger RC, Welch KJ. Surgical repair of pectus excavatum. *J Pediatr Surg*. 1988;23(7):615-622. doi:10.1016/s0022-3468(88)80629-8
- Croitoru DP, Kelly RE, Goretsky MJ, Lawson ML, Swoveland B, Nuss D. Experience and modification update for the minimally invasive Nuss technique for pectus excavatum repair in 303 patients. *J Pediatr Surg*. 2002;37(3):437-445. doi:10.1053/jpsu.2002.30851
- Fokin AA, Steuerwald NM, Ahrens WA, Allen KE. Anatomical, histologic, and genetic characteristics of congenital chest wall deformities. *Semin Thorac Cardiovasc Surg*. 2009;21(1):44-57. doi:10.1053/j.semtcv.2009.03.001
- Fonkalsrud EW. 912 open pectus excavatum repairs: changing trends, lessons learned: one surgeon's experience. *World J Surg*. 2009;33(2):180-190. doi:10.1007/s00268-008-9793-4
- Williams AM, Crabbe DCG. Pectus deformities of the anterior chest wall. *Paediatr Respir Rev*. 2003;4(3):237-242. doi:10.1016/s1526-0542(03)00053-8
- Obermeyer RJ, Goretsky MJ. Chest wall deformities in pediatric surgery. *Surg Clin North Am*. 2012;92(3):669-684. doi:10.1016/j.suc.2012.03.001
- Park CH, Kim TH, Haam SJ, Lee S. Rib overgrowth may be a contributing factor for pectus excavatum: evaluation of prepubertal patients younger than 10 years old. *J Pediatr Surg*. 2015;50(11):1945-1948. doi:10.1016/j.jpedsurg.2015.07.010
- Fokin AA, Steuerwald NM, Ahrens WA, Allen KE. Anatomical, histologic, and genetic characteristics of congenital chest wall deformities. In: *Seminars in Thoracic and Cardiovascular Surgery*. Vol 21. Elsevier; 2009. <https://www.sciencedirect.com/science/article/pii/S1043067909000318>
- Leung AK, Hoo JJ. Familial congenital funnel chest. *Am J Med Genet*. 1987;26(4):887-890. doi:10.1002/ajmg.1320260416
- Creswick HA, Stacey MW, Kelly RE, et al. Family study of the inheritance of pectus excavatum. *J Pediatr Surg*. 2006;41(10):1699-1703. doi:10.1016/j.jpedsurg.2006.05.071
- Behr CA, Denning NL, Kallis MP, et al. The incidence of Marfan syndrome and cardiac anomalies in patients presenting with pectus deformities. *J Pediatr Surg*. 2019;54(9):1926-1928. doi:10.1016/j.jpedsurg.2018.11.017
- Billar RJ, Manoubi W, Kant SG, Wijnen RMH, Demirdas S, Schnater JM. Association between pectus excavatum and congenital genetic disorders: a systematic review and practical guide for the treating physician. *J Pediatr Surg*. 2021;56(12):2239-2252. doi:10.1016/j.jpedsurg.2021.04.016
- Saxena AK, Willital GH. Valuable lessons from two decades of pectus repair with the Willital-Hegemann procedure. *J Thorac Cardiovasc Surg*. 2007;134(4):871-876. doi:10.1016/j.jtcvs.2007.06.008
- Ewert F, Syed J, Kern S, Besendörfer M, Carbon RT, Schulz-Drost S. Symptoms in pectus deformities: a scoring system for subjective physical complaints. *Thorac Cardiovasc Surg*. 2017;65(1):43-49. doi:10.1055/s-0036-1584355
- Haller JA, Kramer SS, Lietman SA. Use of CT scans in selection of patients for pectus excavatum surgery: a preliminary report. *J Pediatr Surg*. 1987;22(10):904-906. doi:10.1016/s0022-3468(87)80585-7
- Malek MH, Fonkalsrud EW, Cooper CB. Ventilatory and cardiovascular responses to exercise in patients with pectus excavatum. *CHEST*. 2003;124(3):870-882. doi:10.1378/chest.124.3.870
- St. Peter SD, Juang D, Garey CL, et al. A novel measure for pectus excavatum: the correction index. *J Pediatr Surg*. 2011;46(12):2270-2273. doi:10.1016/j.jpedsurg.2011.09.009
- Bay V, Farthmann E, Naegele U. Unoperated funnel chest in middle and advances age: evaluation of indications for operation. *J Pediatr Surg*. 1970;5(6):606-609. doi:10.1016/s0022-3468(70)80004-5
- Redding GJ, Kuo W, Swanson JO, et al. Upper thoracic shape in children with pectus excavatum: impact on lung function. *Pediatr Pulmonol*. 2013;48(8):817-823. doi:10.1002/ppul.22660
- Malek MH, Berger DE, Marelich WD, Coburn JW, Beck TW, Housh TJ. Pulmonary function following surgical repair of pectus excavatum: a meta-analysis. *Eur J Cardio-Thorac Surg Off J Eur Assoc Cardio-Thorac Surg*. 2006;30(4):637-643. doi:10.1016/j.ejcts.2006.07.004
- Nuss D, Obermeyer RJ, Kelly RE. Pectus excavatum from a pediatric surgeon's perspective. *Ann Cardiothorac Surg*. 2016;5(5):493-500. doi:10.21037/acs.2016.06.04
- Nuss D, Kelly RE. Indications and technique of nuss procedure for pectus excavatum. *Thorac Surg Clin*. 2010;20(4):583-597. doi:10.1016/j.thorsurg.2010.07.002
- Nuss D, Obermeyer RJ, Kelly RE. Nuss bar procedure: past, present and future. *Ann Cardiothorac Surg*. 2016;5(5):422-433. doi:10.21037/acs.2016.08.05
- Haecker FM, Sesia S. Vacuum bell therapy. *Ann Cardiothorac Surg*. 2016;5(5):440-449. doi:10.21037/acs.2016.06.06
- Haecker FM, Sesia S. Non-surgical treatment of pectus excavatum. *J Vis Surg*. 2016;2(3):63. doi:10.21037/jovs.2016.03.14
- Obermeyer RJ, Cohen NS, Kelly RE, et al. Nonoperative management of pectus excavatum with vacuum bell therapy: a single center study. *J Pediatr Surg*. 2018;53(6):1221-1225. doi:10.1016/j.jpedsurg.2018.02.088
- Kelly RE. Pectus excavatum: historical background, clinical picture, preoperative evaluation and criteria for operation. *Semin Pediatr Surg*. 2008;17(3):181-193. doi:10.1053/j.sempedsurg.2008.03.002
- Papic JC, Finnell SME, Howenstein AM, Breckler F, Leys CM. Postoperative opioid analgesic use after Nuss versus Ravitch pectus excavatum repair. *J Pediatr Surg*. 2014;49(6):919-923. doi:10.1016/j.jpedsurg.2014.01.025
- Haller JA, Colombani PM, Humphries CT, Azizkhan RG, Loughlin GM. Chest wall constriction after too extensive and too early operations for pectus excavatum. *Ann Thorac Surg*. 1996;61(6):1618-1624. doi:10.1016/0003-4975(96)00179-8
- Antonoff MB, Erickson AE, Hess DJ, Acton RD, Saltzman DA. When patients choose: comparison of Nuss, Ravitch, and Leonard procedures for primary repair of pectus excavatum. *J Pediatr Surg*. 2009;44(6):1113-1119. doi:10.1016/j.jpedsurg.2009.02.017
- Hebra A, Kelly RE, Ferro MM, Yüksel M, Campos JRM, Nuss D. Life-threatening complications and mortality of minimally invasive pectus surgery. *J Pediatr Surg*. 2018;53(4):728-732. doi:10.1016/j.jpedsurg.2017.07.020
- Park HJ, Lee SY, Lee CS, Youm W, Lee KR. The Nuss procedure for pectus excavatum: evolution of techniques and early results on 322 patients. *Ann Thorac Surg*. 2004;77(1):289-295. doi:10.1016/S0003-4975(03)01330-4
- Cheng YL, Lin CT, Wang HB, Chang H. Pleural effusion complicating after Nuss procedure for pectus excavatum. *Ann Thorac Cardiovasc Surg Off J Assoc Thorac Cardiovasc Surg Asia*. 2014;20(1):6-11. doi:10.5761/atcs.0a.12.02067
- Park HJ, Lee SY, Lee CS. Complications associated with the nuss procedure: analysis of risk factors and suggested measures for prevention of complications. *J Pediatr Surg*. 2004;39(3):391-395. doi:10.1016/j.jpedsurg.2003.11.012
- Moss RL, Albanese CT, Reynolds M. Major complications after minimally invasive repair of pectus excavatum: case reports. *J Pediatr Surg*. 2001;36(1):155-158. doi:10.1053/jpsu.2001.20039
- Cw L. Pigeon breast (pectus carinatum) and other protrusion deformities of the chest of developmental origin. *Ann Surg*. 1953;137(4):482-489. doi:10.1097/0000658-195304000-00008
- Park CH, Kim TH, Haam SJ, Jeon I, Lee S. The etiology of pectus carinatum involves overgrowth of costal cartilage and undergrowth of ribs. *J Pediatr Surg*. 2014;49(8):1252-1258. doi:10.1016/j.jpedsurg.2014.02.044
- Haje SA, Harcke HT, Bowen JR. Growth disturbance of the sternum and pectus deformities: imaging studies and clinical correlation. *Pediatr Radiol*. 1999;29(5):334-341. doi:10.1007/s002470050602
- Kotzot D, Schwabegger AH. Etiology of chest wall deformities—a genetic review for the treating physician. *J Pediatr Surg*. 2009;44(10):2004-2011. doi:10.1016/j.jpedsurg.2009.07.029
- Fonkalsrud EW. Surgical correction of pectus carinatum: lessons learned from 260 patients. *J Pediatr Surg*. 2008;43(7):1235-1243. doi:10.1016/j.jpedsurg.2008.02.007
- Gritsiuta AI, Bracken A, Beebe K, Pechetov AA. Currarino-Silverman syndrome: diagnosis and treatment of rare chest wall deformity, a case series. *J Thorac Dis*. 2021;13(5):2968. doi:10.21037/jtd-20-3472
- Fonkalsrud EW, Beanes S. Surgical management of pectus carinatum: 30 years' experience. *World J Surg*. 2001;25(7):898-903. doi:10.1007/s00268-001-0048-x
- Shamberger RC, Welch KJ. Surgical correction of pectus carinatum. *J Pediatr Surg*. 1987;22(1):48-53. doi:10.1016/s0022-3468(87)80014-3
- Port E, Hunter CJ, Buonpane C, et al. Echocardiography reveals heart abnormalities in pediatric pectus carinatum. *J Surg Res*. 2020;256:364-367. doi:10.1016/j.jss.2020.06.061
- Kravarusic D, Dicken BJ, Dewar R, et al. The Calgary protocol for bracing of pectus carinatum: a preliminary report. *J Pediatr Surg*. 2006;41(5):923-926. doi:10.1016/j.jpedsurg.2006.01.058
- Martinez-Ferro M, Fraire C, Bernard S. Dynamic compression system for the correction of pectus carinatum. *Semin Pediatr Surg*. 2008;17(3):194-200. doi:10.1053/j.sempedsurg.2008.03.008
- Abdellaoui S, Scalabre A, Piolat C, et al. Pectus arcuatum: a pectus unlike any other. *J Pediatr Surg*. 2023;58(9):1679-1685. doi:10.1016/j.jpedsurg.2023.03.013
- Stephenson JT, Bois JD. Compressive orthotic bracing in the treatment of pectus carinatum: the use of radiographic markers to predict success. *J Pediatr Surg*. 2008;43(10):1776-1780. doi:10.1016/j.jpedsurg.2008.03.049



50. Robicsek F, Watts LT, Fokin AA. Surgical repair of pectus excavatum and carinatum. *Semin Thorac Cardiovasc Surg.* 2009;21(1):64-75. doi:10.1053/j.semtcv.2009.03.002
51. Abramson H, D'Agostino J, Wuscovi S. A 5-year experience with a minimally invasive technique for pectus carinatum repair. *J Pediatr Surg.* 2009;44(1):118-123. doi:10.1016/j.jpedsurg.2008.10.020
52. Yiyit N, Işıtmangil T, Öksüz S. Clinical analysis of 113 patients with Poland syndrome. *Ann Thorac Surg.* 2015;99(3):999-1004. doi:10.1016/j.athoracsur.2014.10.036
53. Fokin AA. Thoracic defects: cleft sternum and Poland syndrome. *Thorac Surg Clin.* 2010;20(4):575-582. doi:10.1016/j.thorsurg.2010.06.001
54. Bavinck JNB, Weaver DD, Opitz JM, Reynolds JF. Subclavian artery supply disruption sequence: hypothesis of a vascular etiology for Poland, Klippel-Feil, and Möbius anomalies. *Am J Med Genet.* 1986;23(4):903-918. doi:10.1002/ajmg.1320230405
55. Romanini MV, Calevo MG, Puliti A, et al. Poland syndrome: a proposed classification system and perspectives on diagnosis and treatment. *Semin Pediatr Surg.* 2018;27(3):189-199. doi:10.1053/j.sempedsurg.2018.05.007
56. Romanini MV, Torre M, Santi P, et al. Proposal of the TBN classification of thoracic anomalies and treatment algorithm for Poland syndrome. *Plast Reconstr Surg.* 2016;138(1):50. doi:10.1097/PRS.0000000000002256
57. Fokin AA, Robicsek F. Poland's syndrome revisited. *Ann Thorac Surg.* 2002;74(6):2218-2225. doi:10.1016/S0003-4975(02)04161-9
58. Cochran JHJ, Pauly TJ, Edstrom LE, Dibbell DG. Hypoplasia of the latissimus dorsi muscle complicating breast reconstruction in Poland's syndrome. *Ann Plast Surg.* 1981;6(5):402. doi:10.1097/0000637-198105000-00010
59. Baldelli I, Santi P, Dova L, et al. Body image disorders and surgical timing in patients affected by Poland syndrome: data analysis of 58 case studies. *Plast Reconstr Surg.* 2016;137(4):1273. doi:10.1097/PRS.0000000000002018
60. Torre M, Palo F, Infant M. Open-surgery repair of congenital malformation of the chest: indications, technical tips and outcomes. *Pediatr Med.* 2019;2:48. doi:10.21037/pm.2019.08.04
61. Keppler-Noreuil KM, Adam MP, Welch J, Muilenburg A, Willing MC. Clinical insights gained from eight new cases and review of reported cases with Jeune syndrome (asphyxiating thoracic dystrophy). *Am J Med Genet A.* 2011;155A(5):1021-1032. doi:10.1002/ajmg.a.33892
62. de Vries J, Yntema JL, van Die CE, Crama N, Cornelissen EA, Hamel BCJ. Jeune syndrome: description of 13 cases and a proposal for follow-up protocol. *Eur J Pediatr.* 2010;169(1):77-88. doi:10.1007/s00431-009-0991-3
63. Phillips JD, van Aalst JA. Jeune's syndrome (asphyxiating thoracic dystrophy): congenital and acquired. *Semin Pediatr Surg.* 2008;17(3):167-172. doi:10.1053/j.sempedsurg.2008.03.006
64. Conroy E, Eustace N, McCormack D. Sternoplasty and rib distraction in neonatal Jeune syndrome. *J Pediatr Orthop.* 2010;30(6):527-530. doi:10.1097/BPO.0b013e3181e03a08
65. Davis JT, Ruberg RL, Leppink DM, McCoy KS, Wright CC. Lateral thoracic expansion for Jeune's asphyxiating dystrophy: a new approach. *Ann Thorac Surg.* 1995;60(3):694-696. doi:10.1016/0003-4975(95)92703-0
66. Muthialu N, Mussa S, Owens CM, Bulstrode N, Elliott MJ. One-stage sequential bilateral thoracic expansion for asphyxiating thoracic dystrophy (Jeune syndrome). *Eur J Cardio-Thorac Surg Off J Eur Assoc Cardio-Thorac Surg.* 2014;46(4):643-647. doi:10.1093/ejcts/ezu074
67. Waldhausen JHT, Redding GJ, Song KM. Vertical expandable prosthetic titanium rib for thoracic insufficiency syndrome: a new method to treat an old problem. *J Pediatr Surg.* 2007;42(1):76-80. doi:10.1016/j.jpedsurg.2006.09.059
68. Ashok Raja J, Mathevan G, Mathiarasan K, Ramasubramaniam P. Closing the cleft over a throbbing heart: neonatal sternal cleft. *BMJ Case Rep.* 2014;2014:bcr2014204529. doi:10.1136/bcr-2014-204529
69. Williams AP, Marayati R, Beierle EA. Pentalogy of cantrell. *Semin Pediatr Surg.* 2019;28(2):106-110. doi:10.1053/j.sempedsurg.2019.04.006
70. Shamberger RC, Welch KJ. Sternal defects. *Pediatr Surg Int.* 1990;5(3):156-164. doi:10.1007/BF00179653
71. Fokin AA. Cleft sternum and sternal foramen. *Chest Surg Clin N Am.* 2000;10(2):261-276. doi:10.1016/S1052-3359(25)00495-8
72. Torre M, Rapuzzi G, Guida E, Costanzo S, Jasonni V. Thymectomy to achieve primary closure of total sternal cleft. *J Pediatr Surg.* 2008;43(12):e17-e20. doi:10.1016/j.jpedsurg.2008.09.011
73. Sabiston DC. The surgical management of congenital bifid sternum with partial ectopia cordis. *J Thorac Surg.* 1958;35(1):118-122. doi:10.1016/S0096-5588(20)30288-9
74. Biswas G, Khandelwal NK, Venkatramu NK, Chari PS. Congenital sternal cleft. *Br J Plast Surg.* 2001;54(3):259-261. doi:10.1054/bjps.2000.3527
75. Laamiri R, Kechiche N, Hmidi N, et al. Isolated central sterna clefts: a rare congenital malformation. *Afr J Paediatr Surg AJPS.* 2021;18(2):117-118. doi:10.4103/ajps.AJPS_47_20

Evaluation of Yolk sac tumor in a pediatric patient with F-18 FDG PET/CT

¹Güler Silov¹, ²Nilüfer Bıçakcı², ¹Fatih Batı¹, ²Banu Kırtıloğlu²

¹Department of Nuclear Medicine, Faculty of Medicine, Samsun University, Samsun, Türkiye

²Department of Nuclear Medicine, Samsun Training and Research Hospital, Samsun, Türkiye

Received: 25/05/2024

Accepted: 02/07/2024

Published: 10/04/2025

Cite this article: Silov G, Bıçakcı N, Batı F, Kırtıloğlu B. Evaluation of Yolk sac tumor in a pediatric patient with F-18 FDG PET/CT. *Surg Child*. 2025;2(2):79-81.

Corresponding Author: Nilüfer Bıçakcı, niluferbicakci@gmail.com

ABSTRACT

Yolk sac tumors are a form of malignant primitive germ cell tumor that exhibit similarities to the mesenchymal cells of the primitive yolk sac in histological terms. These tumors are frequently observed in pediatric patients and can be found in various locations throughout the body. It is essential to diagnose and treat this condition promptly to minimize the high morbidity and mortality associated with it. The aim of this report is to assess the efficacy of F-18 FDG PET/CT in the initial staging of the disease, evaluating chemotherapy response, and restaging the disease following chemotherapy and a second surgical intervention.

Keywords: Yolk sac tumor, pediatric patient, PET/CT

INTRODUCTION

Malignant germ cell tumor (MGCT), of which yolk sac tumor (YST) is the most common subtype, accounting for only 1-4% of childhood malignancies.¹ YST is thought to arise from primitive germ cells. YST can be of testicular, ovarian or extragonadal origin.² The level of alpha-fetoprotein (AFP) is high in most patients due to the endodermal origin of YST. Therefore, the level of AFP is considered to be an important biomarker.

The outcome of patients with YST is closely related to the stage of the disease. Studies have shown that children with gonadal YST in stage 1 may benefit from surgical intervention. F-18 FDG PET/CT is a system that integrates metabolic imaging with anatomical imaging. F-18 FDG PET/CT is widely used in pediatric lymphoma, neuroblastoma, soft tissue sarcoma and other malignancies. However, the use of F-18 FDG PET/CT in pediatric YST has mostly been reported in case reports.³ In this report, we evaluated the usefulness of F-18 FDG PET/CT in the initial staging of the disease, in the assessment of chemotherapy response, and in the restaging of the disease after chemotherapy and second surgical treatment.

CASE REPORT

A 15-year-old Syrian girl patient underwent surgery for abdominal swelling and pain in her country. A right salpingo-oophorectomy was performed. A mass measuring

220x150x120 mm was removed. Pathology report compatible with an ovarian YST.

Then the patient was admitted to our hospital for treatment. Blood tests showed AFP 426 ng/ml (reference range 0-40 ng/ml), Beta HCG 0.1 mIU/ml (reference range 0-10 mIU/ml). Abdominal and pelvic MRI, F-18 FDG PET/CT imaging and USG were performed. A total of four F-18 FDG PET/CT scans of a pediatric patient were analysed.

F-18 FDG PET/CT images were evaluated retrospectively by two nuclear medicine physicians by consensus. Both were blinded to the clinical and imaging findings. F-18 FDG PET/CT images were interpreted as positive or negative for disease. Any non-physiological focal F-18 FDG uptake on PET was anatomically localised on CT. Semi-quantitative analysis was performed using the maximum standardized uptake value (SUV max).

On evaluation of the first post-operative F-18 FDG PET/CT scan, multiple hypermetabolic lesions were observed on the left side of the pelvis on the MIP image. Cross-sectional imaging at the level of the pelvic inlet showed metastatic foci in the right rectus abdominis muscle and a metastatic focus near the left psoas muscle were hypermetabolic. Pelvic masses anterior and posterior to the uterus and a mass in the anterior wall of the left pelvis were hypermetabolic and pelvic fluid was observed (Figure 1a, 1b). At the level of the pelvic floor, increased F-18

FDG uptake was seen in a hypodense lesion in the left rectus abdominis muscle and in inguinal lymph nodes.

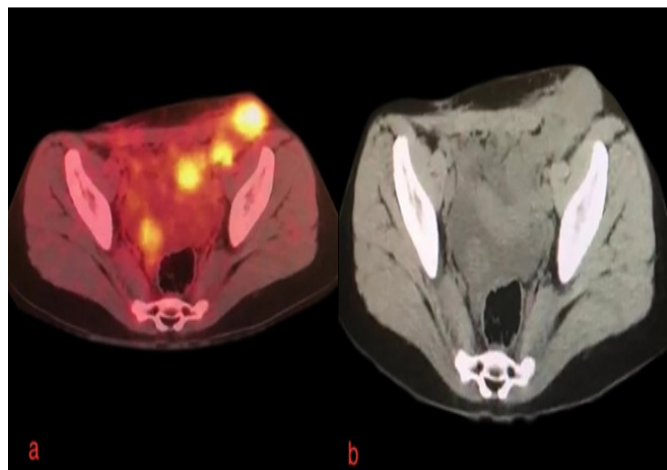


Figure 1. First post-operative F-18 FDG PET/CT images. Fusion image (a) and CT image (b)

The cisplatin-etoposide-bleomycin (PEB) chemotherapy protocol was used. The second follow-up F-18 FDG PET/CT scan was performed after 2 cycles of the PEB protocol. In the last F-18 FDG PET/CT scan, no pathological FDG uptake was observed in the pelvic region on MIP images. Cross-sectional imaging at the level of the pelvic inlet showed a calcified lesion without significant FDG uptake in the right rectus abdominis muscle. No mass was observed in the left lateral psoas muscle. At the level of the uterus, there was no significant FDG uptake in the calcified lesions of the left anterior pelvic wall, and no other lesions of the pelvis were observed (**Figure 2a, 2b**). **Table** shows the temporal change in the number, size and metabolic activity of the lesions after initial surgery, following response to chemotherapy and at the end of treatment by F-18 FDG PET/CT imaging and the decrease in serum AFP levels.

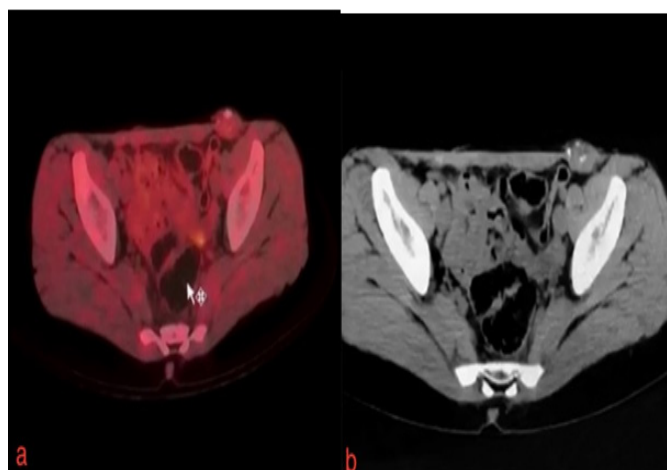


Figure 2. F-18 FDG PET/CT images at the end of chemotherapy. Fusion image (a), CT image (b)

Following MRI findings of a slight increase in the size of a calcified mass in the anterior wall of the pelvis, a decision was made at a consultative meeting to excise the mass and surgery was performed. The patient's current pathology was compatible with a mature teratoma (**Figure 3**). A decrease in AFP level is shown in **Figure 4** and **Table**.

Mature teratomas are considered benign and are not included in the management protocol. The patient is currently stable

Table. The temporal characteristics of the lesions (size, number, location, SUVmax) and serum AFP levels

Study no	LTS (mm)/ no of lesion	Lesion sites	SUVmax	AFP (ng/ml)
1	39x23 mm/5	Pelvic anterior wall	4.2	426
	33x19 mm/6	Pelvic	4.6	
	33x3 mm/4	Inguinal	6.8	
2	30x17 mm/5	Pelvic anterior wall	1.0	21.15
	16x16 mm/1	Pelvic	2.8	
	Ss/multiple	Left external iliac	1.8	
3	27x15 mm/5	Pelvic anterior wall	2.4	1.91
	12 mm/1	Pelvic	1.9	
	Ss/multiple	Left external iliac	3.8	
4	26x16 mm/5	Pelvic anterior wall	-	1.77
	-	Pelvic	-	
	-	Left external iliac, left inguinal	-	

AFP: Alpha-fetoprotein, LTS: Largest tumor size, Ss: Subcentimetric

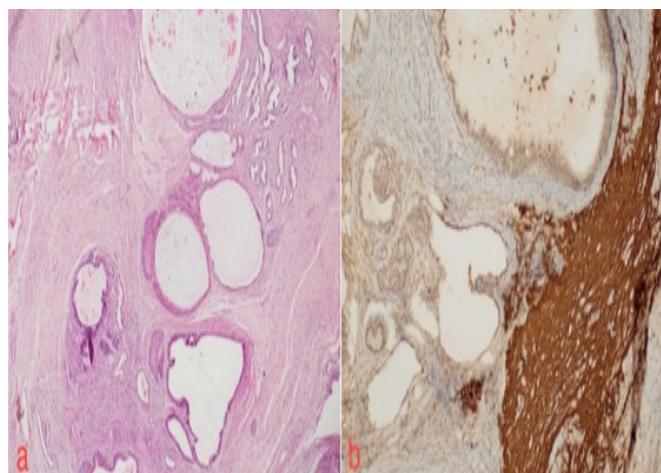


Figure 3. Histopathology compatible with a mature teratoma. Hematoxylin and eosin staining (HEx10) (a). Glial tissue stained positive for GFAP, immunohistochemical staining (IHKx10) (b)

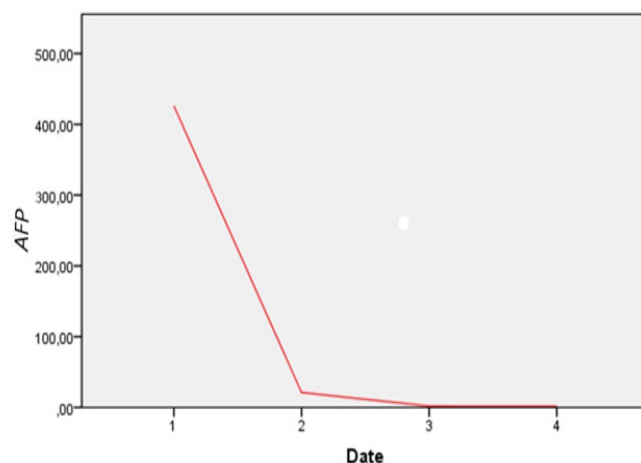


Figure 4. Follow-up AFP measurement levels
AFP: Alpha-fetoprotein

and under close observation. No residual disease was found on the second post-surgery imagings. No distant metastases were observed.

DISCUSSION

YST is a rare tumor that is often included in the MGCT subgroup. Few studies in the literature have focused on YST alone.⁴ Most studies in the literature have focused on MGCT, a heterogeneous group that includes seminoma, dysgerminoma,

embryonal carcinoma, choriocarcinoma, polyembryoma, gonadoblastoma and teratoma.⁵

There are few studies on the usefulness of F-18 FDG PET/CT in MGCT, and most of these have been in the adult population.⁶ There are even fewer studies on the use of F-18 FDG PET/CT in pediatric MGCT. It is important to clarify the usefulness of F-18 FDG PET/CT in pediatric YST.⁷ The value of F-18 FDG PET/CT in preoperative staging of pediatric YST or MGCT is unknown. F-18 FDG PET/CT is not routinely recommended for initial staging of MGCT in adults because of the small additional benefit of F-18 FDG PET/CT compared with CT or MRI.⁸ In our study, although F-18 FDG PET/CT provided additional metabolic information to conventional imaging, the F-18 FDG PET/CT findings did not change the stage of the disease and therefore did not change clinical management in the initial studies. However, last F-18 FDG PET/CT images contributed to the decision to refer the ametabolic pelvic anterior wall mass for surgery. Histopathological examination showed that the lesion was compatible with benign mature teratoma.

CONCLUSION

F-18 FDG PET/CT is an imaging modality with high diagnostic accuracy in demonstrating anatomical and metabolic response to chemotherapy after surgical treatment in patients YST. It can show tumor differentiation at follow-up and has a major impact on therapeutic management.

ETHICAL DECLARATIONS

Informed Consent

The patient's parents signed a free and informed consent form.

Referee Evaluation Process

Externally peer-reviewed.

Conflict of Interest Statement

The authors have no conflicts of interest to declare.

Financial Disclosure

The authors declared that this study has received no financial support.

Author Contributions

All of the authors declare that they have all participated in the design, execution, and analysis of the paper, and that they have approved the final version.

REFERENCES

1. Yin M, Wang T, Yang JX. Yolk sac tumor of the uterus in a 2-year-old girl: a case report and literature review. *J Pediatr Adolesc Gynecol.* 2022; 35(2):177-181. doi:10.1016/j.jpaga.2021.09.005
2. Euscher ED. Unusual presentations of gynecologic tumors: extragonadal yolk sac tumor of the vulva. *Arch Pathol Lab Med.* 2017;141(2):293-297. doi:10.5858/arpa.2016-0151-SA
3. Tang W, Liu Z, Fu H, Li C, Wang H. FDG PET/CT evaluation of pediatric patients with yolk sac tumor. *AJR Am J Roentgenol.* 2019;213(3):676-682. doi:10.2214/AJR.18.20968
4. Sharma C, Shah H, Sisodiya Shenoy N, Makhija D, Waghmare M. Ovarian yolk sac tumour in a girl- case report. *Dev Period Med.* 2017; 21(2):101-103. doi:10.34763/devperiodmed.20172102.101103
5. Euscher ED. Germ cell tumors of the female genital tract. *Surg Pathol Clin.* 2019;12(2):621-649. doi:10.1016/j.path.2019.01.005
6. Jin AF, Luo ZH. 18F-DG PET/CT imaging of ovarian yolk sac tumor in a woman after induction of labor: a case report. *Curr Med Imaging.* 2024;9. doi:10.2174/0115734056260771231019101641
7. Tang W, Liu Z, Fu H, Li C, Wang H. FDG PET/CT evaluation of pediatric patients with yolk sac tumor. *AJR Am J Roentgenol.* 2019;213(3):676-682. doi:10.2214/AJR.18.20968
8. Wang Z, Lu F, Song C, et al. Ultrasonic misdiagnosis of giant pediatric testicular yolk sac tumor: a case report and literature review. *Front Pediatr.* 2022;20(10):1058037. doi:10.3389/fped.2022.1058037