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Features of surgical line managements in Baku

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ABSTRACT

Aims: Central venous catheters (CVC) have become an acceptable form of permanent access in patients with limited access options. Proper management of this procedure is crucial for a successful treatment period. Therefore, we aim to demonstrate our insertion steps and discuss our findings with the literature, highlighting the successful outcomes we have achieved.

Methods: After the approval of the hospital directorate, central catheters were inserted into pediatric patients at a Children's Hospital in Baku through a retrospective review of patient notes over the last two years. Experienced or inexperienced staff, operating theatre or ward, emergent or elective insertions, catheter diameters, catheter dimensions, selected models, number of lumens, reason for insertion, insertion method, fixation methods, dressings, and locations of catheter tips are evaluated. Also, companies that produce catheters are mentioned.

Results: Sixty-three CVC insertions were performed via CVC between 2021 and 2023. Forty-one catheters were inserted by senior staff. Sixty-one were inserted in the operating theatre. Fifty-four were emergent applications. Mainly, 6.5 and 7 French lines were used. Three different international companies supplied the catheters. Half of the catheters used were double-lumen. Oncology diseases were the most common reasons for insertions (n=40). Seldinger's method was the main one chosen for insertions (n=59). Prolene sutures are mainly used to fix catheters. After insertions, iodine dressings (tegaderm®) are commonly used (n=48). The right atrium was the principal place for the lumen tip (n=39).

Conclusion: Our procedures for central venous catheterizations are similar to those in the literature. Nevertheless, the catheter might be positioned at the ending area of the superior vena cava. We observed no harm in the patients with the right atrium location. Also, fixation and trademarks might be matters of evaluation about the effectiveness of procedures.

Keywords: Children, central venous catheters, features

INTRODUCTION

Central venous catheters (CVC) are devices that must be used inevitably when medium or long-term vascular access is required.¹⁻³ Their use has been significantly increased, especially in the last 20 years.² The most crucial factor in this situation has been the emergence of the need for reliable vascular access for modern daily health care.⁴ Central catheters have become preferred over time for their comfort, convenience, cheapness, and long-term use against the possibility of complications.⁵

Especially in intensive care units, emergency rooms, operating rooms, and wards, situations requiring urgent or planned insertion for reasons such as patient diagnosis and treatment frequently occur.^{1,3,6} CVCs are used for various reasons: hemodynamic monitoring, parenteral nutrition, extracorporeal treatments, blood samples, drug and fluid treatments, and supplementation of blood and its products.^{6,7} The reasons for using CVC are generally for treating oncological diseases.⁸ They also form the permanent access option in a limited subset of

dialysis patients. If a CVC is placed in an emergency, removing it after 48 hours is also recommended. The purpose of the use is essential in terms of catheter choice.^{4,7} It is necessary to evaluate which catheter will be preferred.⁷

Placement of central catheters is an application that requires technical expertise.³ Otherwise, it may cause an increase in complications and infections.³ Practitioners' ability to implement the central venous catheter with evidence-based practical information will ensure effectiveness and sustainability.⁹ Although institutional conditions may vary, effective use can be designed by paying attention to basic features.⁹

Differences may occur regarding catheter insertion in emergency or elective situations and the ability to use the catheters. If a central venous catheter is inserted in an emergency, it is also recommended to be removed after 48 hours.⁷



The diameter of the catheters is related to the catheter and catheter location to be used according to the patient’s needs. When choosing a catheter, it must be smaller than the venous diameter, but it is reported that its use will be problematic in case of excessive stenosis.⁷ Therefore, catheters of different diameters are available.

While the evaluation of catheter brands in terms of effectiveness and sustainability is not a widely discussed topic, it is an area that warrants attention. These brands offer various types of catheters, and choosing between silicone or polyurethane catheters can also impact effectiveness. Therefore, future research should consider the role of catheter brands in CVC management.

They are generally defined as tunneled central catheters, peripherally located central catheters, and implanted catheters.⁹ The catheters are generally tunneled, cuffless, or cuffed catheters.⁷ These may also be called subcutaneous ports or Hickman-Broviac.⁸ The purpose of CVCs used with cuffs is to prevent infection by causing fibrosis in the cuff area and to fix the catheter.⁸ Mainly, dual-lumen catheters are used. Dual-lumen tunneled catheters are the preferred form of access for intermediate use. One of the indications for using CVC is to cover the examination and treatment process in emergencies.⁷ Although methods such as a peripheral catheter or intraosseous catheter are at the forefront, placement of CVC is also essential when necessary.⁷

It has been stated that when a central catheter is inserted, fixation without stitches is better and that subcutaneous anchorage, cyanoacrylate adhesive, or a semitransparent dressing can be used.⁷

One of the necessary conditions for central catheters to be used under appropriate conditions is the placement of the catheter with a maximum sterile protection barrier.¹⁰ Catheter dressings can be made with sterile gauze or transparent and semi-permeable materials.¹⁰ In a study conducted in Mexico, it was stated that complications caused by infection will decrease if various parameters are taken into consideration.⁵ In addition to appropriate dressing, hand hygiene, skin asepsis, maximum barrier use, timely catheter removal, and maximum infection prevention in pediatric intensive care units have been identified as the most critical factors.⁵

Improper placement of central catheters is a situation that can be considered a complication.⁵ Methods that can be used to insert the catheter include blind insertion, venography (an old method), ultrasonography, or fluoroscopy.⁷ The tip of the central catheter is expected to be adjacent to the right atrium and superior vena cava.³ It is essential to determine the catheter location.¹¹ For this purpose, USG and X-ray evaluation can be used.¹¹

We evaluated these features in our patients and tried to understand local behavior compared to the literature.

METHODS

The study was carried out with the permission of Ethical Committe Ege Hospital (Date: 17.09.2024, Decision No: 392). All procedures were carried out in accordance with the ethical rules and the principles of the Declaration of Helsinki.

After the hospital directorate approved the study, data from 63 patients hospitalized in the Clinical Medical Center were evaluated. The study evaluated the staff that inserted CVC, the place insertion performed, emergency or elective insertions, line sizes, catheter trademarks, catheter types, reason for insertions, insertion method, surgical sutures used for insertions, dressing types, and catheter tip. Features of the central venous catheter insertions were documented to define our cumulative management strategies retrospectively-all parameters, where available, were discussed with literature.

RESULTS

CVC insertions were performed in 63 patients. All data are defined in the Table 1 and Table 2. Emergent procedure is performed in fifty-four patients, and elective procedure is performed in 11. Senior surgeons performed forty-one insertions, and 22 insertions were performed by residents. Sixty-one were performed in the operating theatre. Fifty-four insertions were performed in emergent conditions. Different catheter sizes were used between 3F and 12F. The most used catheter was 6.6F, and 27 catheter insertions were performed with this size. Seven-French size catheter was the second most used one. Products from three companies were used. Double-lumen catheters were the most needed type, and 29 catheters were in this group. Forty oncology patients received the insertions. Parenteral nutrition, hematology diseases, and antibiotics-related treatment patients followed this group.

Table 1. Features of CVC insertions and number of interventions	
Line inserted by	Senior: 61 Junior: 2 Total: 63
Line insertions	Operating theatre: 61 Ward: 2
Planning	Emergent: 54 Elective: 9
Line size	6.6Fr: 27 7Fr: 23 4.2Fr: 4 5Fr: 4 3Fr: 2 6Fr: 1 10Fr: 1 12Fr: 1
Type of catheters	Double lumen: 29 Single lumen: 8 Hemodialysis: 3 NA: 21
NA:Not available, CVC: Central venous catheters	

Table 2. Features of CVC insertions and number of interventions	
Patient diagnosis	Oncology: 40 Parenteral nutrition: 12 Hematologic: 5 Antibiotics: 3 NA: 3 Total: 63
Insertion technique	Percutaneous: 59 Surgery: 4 Prolene: 15
Fixation	Polydioxanone: 2 Silk: 1 NA: 45 Tegaderm: 43
Dressing	Gel contents:10 Biopatch: 3 Steril sponges: 3 NA: 4
Tip locations	Right atrium junciton: 39 Upper right atrium: 16 Superior vena cava: 6 Middle atrium: 1 NA:1
NA:Not available, CVC: Central venous catheters	



Almost all of them were inserted using Seldinger's method. Fifteen catheters were fixed with prolene sutures; two were with polydioxanone, and one was with silk sutures. Unfortunately, information for surgical sutures for other patients was not found. Forty-three patients were treated with tegaderm; other dressings were biopatch, jelly, and povidon-iodine. Thirty-nine catheter tips were located at the right atrium junction. Sixteen were in the upper atrium. One was in the middle atrium. Six were in superior vena cava.

DISCUSSION

While the problems caused by catheters can be disturbing even in adults, it can be expected that children's compliance with a central venous catheter and the problems may be more psychologically complex.⁸ For this reason, much work is being done worldwide on the placement and maintenance of CVC.⁹ Naturally, in case of systematic central catheter use errors, a review will be mandatory individually or in the healthcare system.⁹ For this reason, the most crucial point for CVC is to observe the effectiveness of catheter placement before treatment.⁹ Studies have stated that the most crucial point for these practices to be effective and sustainable is to be trained in using central catheters and, therefore, to allow people to perform these practices.^{1,9} It is stated that to prevent complications, experienced physicians should perform central catheter placement, or an experienced physician should be present next to the inexperienced physician.⁶ The American Board of Internal Medicine reports that approximately 10-290 catheter insertion procedures must have been performed to gain experience.⁶ In our clinic, 61 of the 63 catheters were made by an experienced surgeon. It has been observed that this approach is compatible with general acceptance. It is essential for physicians working as assistants to be involved in and observe these procedures.⁸ These observations will ensure they understand the device and are familiar with all the procedure features.⁸ For now, two catheter placements have been performed with junior staff.

It has been determined that the area of placement is as essential as catheter placement. Studies have stated that hospitals try to prevent central catheter use outside intensive care units.¹ It has been stated that they aim to prevent possible complications by ensuring that it is used by specific procedures.¹ It was stated that 33 pediatric hospitals work at specific standards to ensure catheter placement with appropriate procedures. All these hospitals are members of an organization called "Children's Hospital Solutions for Patient Safety" and collaborate.¹ In a study conducted in Italy, any central, peripheral, or intraosseous catheter that can be placed in patients' emergencies is expressed as necessary.⁷ Central catheters with or without tunnels are also included. However, if a CVC is inserted in an emergency, it is also recommended to remove it after 48 hours.⁷ It has been determined that central catheters inserted in operating rooms will likely cause bloodstream infections.² In our study, it is seen that most of the catheters are incompatible with the literature because they are inserted in the operating room. Another study determined that the PICU was where the procedure was performed the least.⁹ The reason for the few insertions in the PICU was that the unit was less preferred due to its complexity.⁹

There are different types of CVCs. These are subcutaneous ports, Hickman-Broviac, and non-tunnelled catheters that

can be placed peripherally.⁸ The purpose of CVCs used with cuffs is to prevent infection by causing fibrosis in the cuff area and to fix the catheter.⁸ CVCs are placed for various reasons, such as hemodynamic monitoring, parenteral nutrition, extracorporeal treatments, blood sample collection, drug and fluid treatments, and supplementation of blood and its products.⁶ A study found a lack of peripheral vascular access in 47% and the need for hemodynamic monitoring in 22%.⁶ In the same study, the authors found their reasons to be 28% fluid and drug therapy, 25% extracorporeal therapy, and 25% lack of peripheral vascular access.⁶ The catheters used in our hospital were mainly used for oncological reasons and international use. After this reason, according to the frequency of use, patients used for parenteral nutrition, hematological reasons, and antibiotics come next. What the diseases are is excluded from the purpose of our study. In addition, the majority of catheters used were double-lumen catheters. Single-lumen and hemodialysis catheters follow this type of catheter. It is understood that this choice is compatible with international practice.

Evaluation according to the diameter of catheters is very rare in the literature. One study reported that catheters should be smaller than the venous diameter, but if necessary, they will have problems in case of excessive stenosis.⁷ We used catheter diameters as data in our study, but no study or evaluation criteria evaluate diameters according to cases. However, planning which catheter is most appropriate has been examined.¹ For this purpose, it has been stated that central venous catheter sets are established, and the catheter properties are determined according to the patient's condition.¹ It has been stated that catheter-related complications can be reduced this way, but there is little literature on this type of information for children.¹ For this reason, detailed analysis was not used in our study. The use of 6.6 and 7F catheters as a preference can be expressed as a clinical feature.

There is no comparison and effectiveness analysis of catheter brands. This is understandable. In our study, the brands were taken into account, but instead of expressing them as names, it was deemed appropriate to identify the three most used brands worldwide. We believe that the necessity of performing effectiveness analyses of these brands should be considered a principle in future studies.

In our study, almost all catheter placements were performed using the percutaneous (Seldinger's) method using ultrasonography. No difference was found in complications of central venous catheter complications, especially infection and thrombosis, between using and not using ultrasonography.⁶

It has been stated that when a central catheter is inserted, fixation without stitches is better, and subcutaneous anchorage, cyanoacrylate adhesive, or semitransparent dressing can be used for this.⁷ Our study found that sutures were placed in nearly half of our patients. We have recorded the sutures, but a detailed study needs to be done. We tried to find records of the subjects in our archives for many of our patients. It was decided to follow the issue in our subsequent clinical follow-ups.

A study showed that catheter-related infections were significantly reduced if catheter care was performed with povidone-iodine.² Although there are drawbacks to the



use of chlorhexidine because it causes skin problems, especially in premature babies, it continues to be preferred worldwide.² In our patients, a semitransparent iodine-impregnated drape was used, and other methods were used. This situation was thought to be compatible with the literature.

A study stated that the tips of the catheters could be visualized at a rate of 95% using USG.¹¹ In the same study, care was taken to ensure the catheter tip was placed adjacent to the tracheal bifurcation.¹¹ If the catheter is placed in the cranial direction, problems such as central venous pressure measurement error or spontaneous catheter removal may occur. If it is sent too much toward the heart, problems such as arrhythmias, tears in the atrial or ventricular walls, and hemopericardium may occur. It is known that the optimal placement area is the lower 1/3 of the superior vena cava until its junction with the right atrium.¹¹ The tracheal carina is the best landmark. However, in some studies, it can be expressed as slightly below the carina.¹¹ It was determined that the catheter was located close to the cranial position in one of our patients and that the others were located typically.

CONCLUSION

Our catheter applications in operating room conditions differ from the literature, and when compared, it is understood that we do not have a definitive, standard application. However, paying attention to the practitioner's characteristics, being sensitive about the location of the catheter, and the care shown for the care processes are at world standards. We cannot say whether CVC placement is similar across our country, but the information of our clinic is important as a start.

ETHICAL DECLARATIONS

Ethics Committee Approval

The study was carried out with the permission of Ethical Committee Ege Hospital (Date: 17.09.2024, Decision No: 392).

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

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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.

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Effect of mucous fistula refeeding in neonates with small bowel enterostomy at a low resource center- a prospective interventional study: a single center experience in a Bangladeshi pediatric surgical unit

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ABSTRACT

Aims: Neonates with small bowel enterostomy face substantial challenges in maintaining fluid balance, optimal nutrition and growth, especially in low- and middle-income countries (LMIC) where parenteral nutrition (PN) is not extensively available. Moreover, the discrepancy between proximal and unused distal loop often makes the anastomosis challenging and risky during closure. This study was aimed to describe a simple method of mucous fistula refeeding (MFR) in a resource constrained environment and to see the impact of MFR on growth and nutrition and distal intestine diameter.

Methods: A simplified MFR technique requiring minimal resources was employed, which was taught to and carried out by the caregivers. Distal loopogram was performed before commencement of MFR and before stoma closure. The weight gain and width difference of the distal loop at splenic flexure was measured. Statistical analysis was done in SPSS 26.0 and $p < 0.05$ considered to be significant.

Results: We implemented MFR on ten consecutive neonates. The indications for enterostomy formation were meconium ileus (6), three among them were complicated, ileal atresia (3), and intestinal perforation (1). Mean rate of weight gain was 18 gm/day and mean gut width increment was 0.1 mm/day ($p < 0.05$). One patient (10%) had anastomotic leakage following stoma closure.

Conclusion: Mucous fistula refeeding is a safe method that can reduce nutritional complications and enhance growth, and reduce bowel lumen discrepancy, making the anastomosis safer during stoma closure.

Keywords: Small bowel enterostomy, mucous fistula refeeding, gut width, rate of weight gain, bowel lumen discrepancy

INTRODUCTION

Enterostomy is an essential procedure in pediatric surgery where patients are not in the condition to take the burden of anastomosis or the condition of the distal bowel are not favorable.¹ In complicated meconium ileus, intestinal atresia, necrotizing enterocolitis, bowel perforation, anastomotic leakage, temporary small bowel stoma becomes surgeon's choice for the safety of the patients.²

Though enterostomies are made as a safe procedure with the intention of avoiding mortality and morbidity of the patients, there are some potential complications for small bowel enterostomies.³ Proximal high output small bowel stoma mimics as short gut syndrome and results in inadequate absorption of nutrients and electrolytes, leading to fluid imbalance, dehydration, malnutrition and failure to thrive,

affecting growth, cognitive development and in severe cases survival.⁴ Previous studies indicate that the prolonged impact of nutrient and fluid loss will lead to a decline in growth in the majority of young children with a stoma, prompting to recommend early stoma closure.⁵

To improve nutritional management in neonates, parenteral nutrition (PN) has gained widespread utilization in developed nations.⁶ Though PN has a significant role in promoting growth and nutrition in the management of short gut syndrome, dependency on PN may cause PN related liver disease (PNALD), metabolic derangements and sepsis.^{6,7} Besides, in developing countries, lack of availability, inadequate aseptic environment and the high expense makes PN a not accessible option.⁸

In children with enterostomy, when the distal bowel or mucous fistula no longer experiences direct exposure to the fecal stream, there is a concurrent decrease in contractility and smooth muscle tone in the mucous fistula.⁹ Moreover, in cases of intestinal atresia, there is higher incidence of complications due to the persistent significant discrepancy between the proximal and distal unused gut.¹⁰ The disuse atrophy results in distal and proximal bowel loop diameter discrepancy which increases the risk for anastomotic leak.¹¹

Mucous fistula refeeding (MFR) or feeding enteroclysis or chyme recycling means transferring the proximal loop content into the distal loop, maintaining the normal pathway of chyme and utilizing the whole remaining gut length as absorptive surface. In recent decades, a growing number of centers have adopted the practice of MFR in the neonatal population.¹²

This intervention is believed to prevent atrophy of the inactive gut, promoting regular absorption and digestion of nutrients.¹³ Additionally, it is thought to stimulate mucosal growth, support intestinal adaptation, and reduce the risk of anastomotic leaks.¹¹

There have been several studies, mostly retrospective, performed in developed countries focusing on the effect of MFR on decreased requirement of PN, and a few review articles and one randomized control trial have proved the safety and efficacy of MFR.^{7,12} There is no study available yet regarding the efficacy and safety of MFR in middle- and low-income setting. The aim of this study was to assess the outcome of MFR in our set up in a prospective trial.

METHODS

The study was carried out with the permission of Ethical Committee Bangladesh Shishu Hospital & Institute (Date: 25.10.2022, Decision No: BSHI/2022/2645). All procedures were carried out in accordance with the ethical rules and the principles of the Declaration of Helsinki.

This prospective intervention study was conducted in the Department of Pediatric Surgery at the Bangladesh Shishu Hospital & Institute from October 2022 to October 2023. Neonates with jejunostomy or ileostomy with mucous fistula managed at our Institution were eligible in the study. Distal obstruction, stenosis, multiple stomas or any anastomosis in the distal bowel were exclusion criteria from the study.

After taking informed consent from the parents, patients with small bowel stoma created at our unit or referred to us after stoma creation, were included in the study. Each patient was thoroughly examined, investigated and all relevant information was noted. The data was recorded in pre-designed, semi-structured questionnaire (supplementary material).

MFR was commenced when patient was on oral feed and could manage hydration and electrolyte balance without any parenteral support. Technique was taught to the caregivers. Proximal loop effluent was collected in a customized ostomy pouch. This was either made by a polybag attached with strips of cloth on both ends and tying this around, or by making a hole in wraparound cloth and adjusting a condom into it over the stoma (Figure 1A, 1B). If the effluent was thick, mixing it with normal saline was suggested. The stoma effluent was transferred in the distal loop within 3 hours to avoid bacterial colonization. An 8-10 Fr feeding tube was advanced 3-5 cm

through the distal stoma. The effluent fluid was infused through the feeding tube with a syringe slowly to prevent backflow of the content (Figure 1C). Distal loopogram was performed twice: before starting MFR and before stoma closure. The distal colon width measurement at splenic flexure in antero-posterior view was measured in order to compare the width increased within the intervention period (Figure 2A, 2B). Weight measurement was also taken before starting MFR and before stoma closure to assess the rate of weight gain.



Figure 1. Methods of collecting proximal effluent either into a poly bag [1A] or into a condom [1B] and infusing proximal loop effluent into mucous fistula by a syringe [1C]

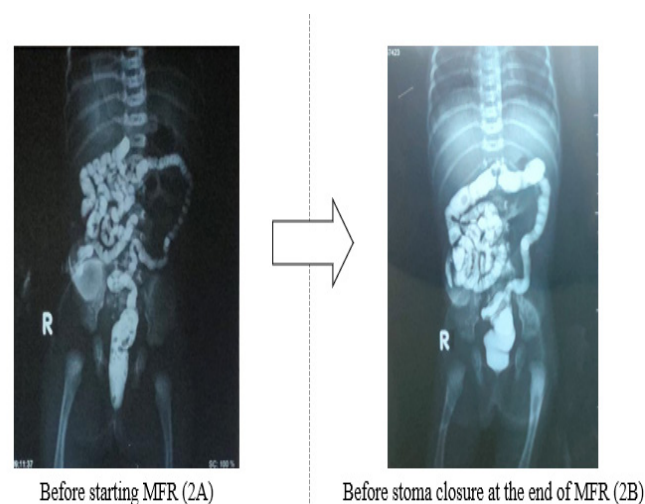


Figure 2. Distal loopogram showing the effect of MFR on distal colonic diameter before starting MFR [2A] and at the end of the intervention before stoma closure [2B]

MFR: Mucous fistula refeeding

Statistical Analysis

The statistical analysis was conducted using SPSS (Statistical package for Social Science) version 26 statistical software. Mean and standard deviation for continuous variables and frequency distribution for categorical variables were used to describe the characteristics of the population. Associations of continuous data were assessed using paired t-test. Associations of categorical data were assessed using Chi-square (X^2) test. Value of $p < 0.05$ was considered significant.

RESULTS

Ten consecutive patients were included. The underlying diagnosis for enterostomy were meconium ileus ($n=3$), complicated meconium ileus ($n=3$) (two with meconium pseudocyst formation and perforation), ileal atresia [3], and intestinal perforation [1] (Figure 3).

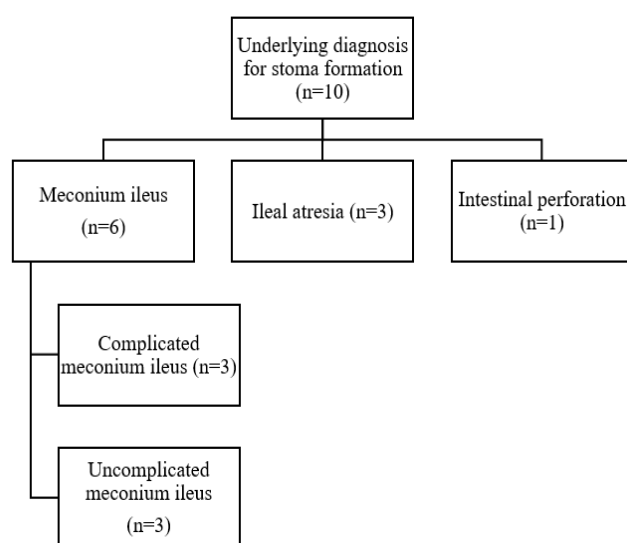


Figure 3. Underlying diagnosis for stoma formation among the participant

Age at start of MFR varied widely, as 4 patients in this series were referred from another hospital, for which refeeding was initiated after 30 days of age. Age at stoma closure also had wide differences, as we delayed the anastomosis where there was satisfactory weight gain and maintenance of hydration and electrolyte balance (Table 1).

Mean weight at start of MFR was 2.49 ± 0.41 kg and mean weight at end of MFR was 3.38 ± 0.61 kg. Mean rate of weight gain was calculated by dividing the weight gain ($W_2 - W_1$) by total days of MFR. 18.18 ± 11.6 gm weight was gained per day during refeeding period (Table 1, Figure 4A).

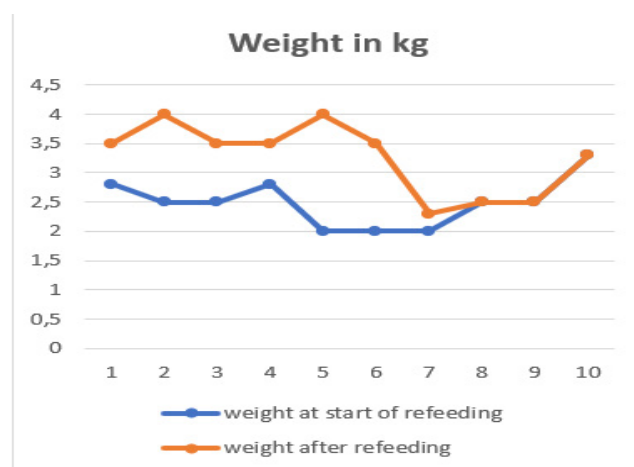


Figure 4A. Increase in weight after MFR

Mean width at splenic flexure of distal loopogram was 6.7 ± 2 mm, which increased to 12 ± 3.4 mm at the end of refeeding. The distal colon width increment rate per day during MFR period was 0.1 ± 0.04 mm (Table 1, Figure 4B). Both weight and distal gut width increment was statistically significant on paired t test ($p < 0.05$) (Table 2).

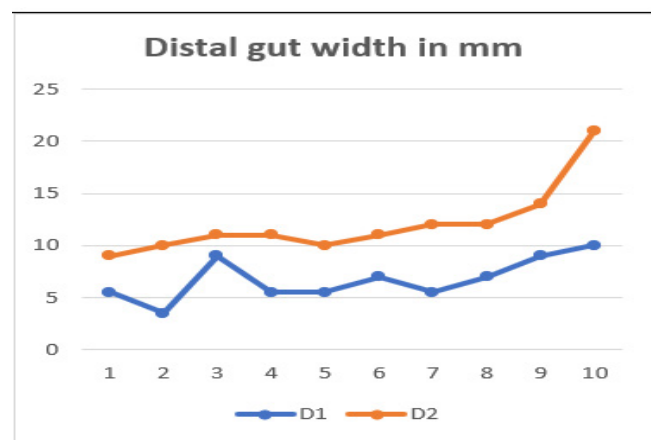


Figure 4B. Distal gut width after MFR

Table 1. Patient's characteristics

Sl no.	Diagnosis	POD ^a of MFR initiation	Total days of MFR	Weight at start of MFR [W1]	Weight after MFR [W2]	D1 ^b	D2 ^c
1	Meconium ileus	30	30	2.8	3.5	5.5	9
2	Complicated meconium ileus	11	56	2.5	4	3.5	10
3	Jejunioleal atresia	8	36	2.5	3.5	9	11
4	Complicated meconium ileus	11	83	2.8	3.5	5.5	11
5	Complicated meconium ileus	30	60	2	4	5.5	10
6	Meconium ileus	40	48	2	3.5	7	11
7	Meconium ileus	14	41	2	2.3	5.5	12
8	Jejunioleal atresia	21	75	2.5	2.5	7	12
9	Jejunioleal atresia	30	42	2.5	2.5	9	14
10	Intestinal perforation	14	60	3.3	3.3	10	21
Mean $\pm$ SD		20.9 $\pm$ 10.91	53.10 $\pm$ 16.993	2.49 $\pm$ 0.41	3.38 $\pm$ 0.61	6.7 $\pm$ 2	12 $\pm$ 3.4

^a Post operative day after stoma formation, ^b Width at splenic flexure at distal loopogram in AP film before MFR [in mm], ^c Width at splenic flexure at distal loopogram in AP film after MFR [in mm], MFR: Mucous fistula refeeding, SD: Standard deviation

**Table 2.** Statistical analysis by paired t test

	Mean	SD	p
Weight gain [W2-W1]	0.89	0.61	.001
Colon width increment [D2-D1]	5.35	2.4	.000

SD: Standard deviation

After stoma closure one patient suffered a complication: a kinking of bowel just distal to the anastomosis was obstructing the flow, which resulted in leakage, relaparotomy was done and double barrel ileostomy was made, and this was successfully closed after 6 weeks. There was no incidence of bowel perforation or stoma prolapse in our study. However, as our customized proximal effluent collection pouch was not leak proof, an extensive excoriation was seen in all patient before stoma closure, but resolved within a week after enterostomy reversal.

DISCUSSION

In this prospective study we have shown that MFR can be safely performed in the context of middle-low-income settings, and that MFR enhance growth and decrease bowel lumen discrepancy, making stoma closure a safer procedure. In 1983 Levy et al.¹¹ first introduced the concept of MFR. However, it was first applied on neonates with ileostomy in 1985 by Puppala et al.¹⁴ where he addressed it as ileostomy drip feeding. There, elemental formula was mixed with proximal loop effluent and was transferred to the distal loop by infusion pump.¹⁴ Since the year 2000, MFR has become a routine practice for managing neonates with mucous fistula in high income countries.¹¹

The majority of studies regarding MFR have been conducted in high-income countries where there is access to extensive manpower and specialized stoma care.¹⁵ Refeeding is widely practiced and has been shown to shorten the duration of dependence on PN in high-income settings.¹⁶ However, there is a lack studies examining the outcomes of MFR in the absence of PN. This study focused on evaluating the outcomes of MFR in infants with small bowel stoma, particularly in situations where PN is not accessible.

In the context of the present study, the predominant etiology contributing to the formation of small bowel enterostomies was meconium ileus and complicated meconium ileus. A review article highlighted necrotizing enterocolitis (NEC) as the most prevalent cause of enterostomy formation among patients undergoing MFR.¹⁵

This study demonstrated a significant rate of weight gain after MFR which aligns with the findings of the previous retrospective researches.^{2,17,18} Their analysis revealed a significantly higher rate of weight gain in the MFR group compared to both the pre-refeeding and those who did not get refeeding. Other studies have corroborated these results, with reported better weight gain in the MFR group compared to non refeeding group.^{19,20}

Wood et al.¹⁶ observed a trend to increased weight gain in the MFR group when compared the result with non refeeding group and found non-significant result. The described insignificant differences in weight gain between the MFR and non-MFR groups may be attributed to the access to PN support which may have contributed to weight gain in control groups.

A retrospective comparative study, showed MFR reduces gut loop discrepancy as well as disuse gut atrophy and postoperative

anastomotic leakage.¹¹ Our study also demonstrated enhancement of gut diameter after refeeding. Likewise, Lee et al.⁷ reported statistically significant larger colonic diameter in the refeeding group compared to the control group. However, upon multivariate analysis accounting for post-menstrual age, Lee et al.⁷ observed that the colon diameter increment rate was insignificant.

One study has reported hazards related to refeeding process which included perforation during tube insertion.²¹ Some authors observed instances of stricture, minor local infections, MF prolapse, and enterocutaneous fistula and stoma bleeding.^{20,22} Though we have not faced this sort of complication, maintaining the consistency of MFR at home has been a great challenge.

Limitations

This was one of the limitations of this study, that the caregivers were unsupervised at home while doing refeeding and we had to rely on their words and consider MFR was being done consistently.

CONCLUSION

MFR might enhance growth and reduce nutritional and fluid balance complications and bowel lumen discrepancy making the anastomosis safer during stoma closure. By establishing wide practice of MFR, it is possible to decrease the morbidity and mortality of neonates with small bowel enterostomy.

ETHICAL DECLARATIONS

Ethics Committee Approval

The study was carried out with the permission of Ethical Committee Bangladesh Shishu Hospital & 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.

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Hypospadias scoring challenges: a pilot study on GMS interpretation

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ABSTRACT

Aims: Hypospadias is one of the most common urological congenital anomalies. Several scoring systems have been established to create a common language for hypospadias because the severity, determined only according to meatus localisation, does not provide an accurate approach. Glans-urethral meatus shaft (GMS) scoring is one of them.

Methods: This pilot study's purpose was to determine how to improve the GMS score (3-12) and if it would be beneficial to divide the three-part structure (mild, moderate, severe) into two groups: high and low scores. Our study was planned and conducted as a one-year prospective study, and 44 cases were included in this study.

Results: Conventional groups were regrouped as the low score group, including GMS 3-7 scores, and the high score group, including GMS 8-12 scores. High score group was statistically significantly higher than low score group regarding the presence of complications ($p=0.021$). No significant correlation was found between glans and penis measurements and development of complications.

Conclusion: Our study suggests that the simpler two-category method we proposed for the evaluation of the GMS score may also be an alternative. It suggests the need for further studies to evaluate and improve the utility of all scoring methods, including the GMS.

Keywords: Hypospadias, classification, score, complication, outcomes

INTRODUCTION

Hypospadias is one of the most common congenital anomalies in boys in which the external urethral meatus is located more proximal than where it should be. The ventral curvature of the penis, developmental defects of the corpus spongiosum, and incomplete prepuce comprise the other components of the presenting anomaly. The assessment of the severity of birth defects can sometimes be quite difficult, and this can be even more complicated in the case of hypospadias. Estimating the severity of hypospadias based solely on the location of the external urethral meatus often leads to inaccurate assessments. There are many factors-such as penis length (PL), glans width (GW), adequacy or shape of the foreskin, penoscrotal changes, degree of curvature, degree of torsion of the penis, urethral plate width (UP) and depth of the urethral groove-used for evaluating the hypospadias.¹⁻⁴ Many scoring methods also have been developed to determine the severity of hypospadias. The motivation for these attempts was the fact that none of the previous classifications could fully describe the degree of the

condition and were inadequate in predicting postoperative possible complications. When we look at all the existing scoring methods today, it is seen that there is no consensus on a scoring system.⁵⁻⁷

Hypospadias scoring involves complex parameters. Considering the need for a common language of hypospadias, many scoring systems have been developed to make this challenging situation more manageable, including anatomical meatus localisation and different features, one of which is the Glans-Urethral Meatus Shaft (GMS) score (Table 1).^{8,9}

The moderate group in the conventional classification is not clearly separated from the mild and severe groups. This grouping method is thought to create difficulties in the surgical approach and evaluation of results. This study aimed to investigate how the GMS score could be improved and whether there would be any advantage in reorganising the three-part structure (mild-moderate-severe) into only two groups, high and low scores.

**Table 1.** GMS scoring sections and criteria**Glans (G) score**

1. Glans good size; healthy urethral plate, deeply grooved
2. Glans adequate size; adequate urethral plate, grooved
3. Glans small in size; urethral plate narrow, some fibrosis or flat
4. Glans very small; urethral plate indistinct, very narrow or flat

Meatus (M) score

1. Glanular
2. Coronal sulcus
3. Mid or distal shaft
4. Proximal shaft, penoscrotal

Shaft (S) score

1. No chordee
2. Mild ($<30^\circ$) chordee
3. Moderate ($30-60^\circ$) chordee
4. Severe ($>60^\circ$) chordee

GMS: Glans-urethral meatus shaft scoring

METHODS

This study was approved by the local ethics committee of the Gülhane Training and Research Hospital, Clinical Researches Ethics Committee (Date: 02.03.2022, Decision No: 2022/25). All procedures were carried out in accordance with the ethical rules and the principles of the Declaration of Helsinki.

We designed this study prospectively by including the patients who would undergo primarily hypospadias surgery after obtaining institutional ethical committee approval and informed consent preparation. We excluded the patients (8 cases) who had undergone circumcision or hypospadias surgery before starting the study. A tubularised incised plate urethroplasty (TIPU) procedure was performed in all cases. In most cases, degloving was sufficient for the straightening of the chordee. All surgical procedures were performed by a single surgeon. A total of 44 paediatric hypospadias cases were included in our study at the end of one year. The age, duration of hospital stay, operation time, GMS scores, GW, UP, PL, and meatus distance (MD) were recorded. The measurement variables were obtained manually and with the help of callipers while the patient was on the operating table just before the operation. Operation time was calculated as the time between incision and dressing. All adverse outcomes which developed during the follow-up period were recorded and categorised as complications. To determine the UP measurement, the widest points of the UP were evaluated horizontally on the glans held stretched before any incision. After the GMS score was obtained, it was grouped conventionally as mild, moderate, and severe. An ROC analysis was applied for the statistical discrimination of the reduction of the GMS grouping to two classes (low score, high score) instead of three (mild, moderate, severe). Accordingly, it was seen that the GMS score had an AUC (Youden index) of 83.6% with a cut-off value (7.50). Thus, scores between 3-7 and between 8-12 were determined as groups. Then the moderate part of the group was taken out and regrouped as only low score (LS) and high score (HS).

The many tests were applied to the statistical analysis. Shapiro-Wilk ($n < 50$) and Skewness-Kurtosis tests were used to determine whether the continuous measurements in the study were normally distributed and parametric tests were applied since the measurements were normally distributed. Independent T-test and One-way Analysis of Variance (ANOVA) were used to compare the measurement values according to categorical

groups. Following the variance analysis, the “Duncan test” was used to determine the different groups. The chi-square test was calculated to determine the relationship between categorical variables. The statistical significance level was taken as $p < 0.05$ and the SPSS (IBM SPSS for Windows, ver.26) statistical package program was used for the analyses.

RESULTS

The mean age (months) of 44 paediatric cases included in the study was 52.14 ± 36.01 . The mean duration of hospital stay (day) of the cases was 9.68 ± 5.52 . The mean follow-up period (months) was 9.7 ± 5.62 . The mean PL (mm) was 24.25 ± 5.75 . The mean GW (mm) was 13.91 ± 3.23 . The mean MD (mm) was determined as 8.14 ± 4.94 . The mean UP (mm) was 6.2 ± 1.8 (Table 2).

Table 2. Means and standard deviations (SD) of continuous variables

	Mean	SD	Min.	Max.
Age (months)	52.14	36.01	13	145
Duration of hospital stay (days)	9.68	5.52	2	27
Follow-up period (months)	9.7	5.62	2	23
Penis length (mm)	24.25	5.75	13	41
Glans width (mm)	13.91	3.23	10	23
Meatus distance (mm)	8.14	9.94	4	70
Urethral plate width (mm)	6.2	1.87	3	10
Operation duration (minutes)	105	44.28	40	300
Glans score	2.39	1.04	1	4
Meatus score	2.18	0.79	1	4
Shaft score	2.09	0.91	1	4
GMS score	6.66	2.24	3	12

Min: Minimum, Max: Maximum, GMS: Glans-urethral meatus shaft

It was determined that the mean Glans score was 2.39 ± 1.04 , the mean Meatus score was 2.18 ± 0.79 , and the mean Shaft score was 2.09 ± 0.91 . The mean total GMS score is 6.66 ± 2.24 . Complications occurred in 12 of 44 cases (27.20%). These complications (Clavien-Dindo 2-3) were fistula in 7 cases, infection in 2 cases, meatal stenosis in 2 cases, and dehiscence in 1 case. There were 23 cases with a GW of 14 mm and above. The number of cases with UP of 8 mm and above was 12. The age and duration of hospital stay in complicated cases were 70 ± 41.39 months and 12.75 ± 6.66 days, respectively. These variables were statistically significantly higher in complicated cases than in uncomplicated cases ($p = 0.042$ and $p = 0.022$, respectively). the Glans score and the Meatus score were statistically significantly higher in complicated cases ($p = 0.001$ for the Glans score and $p = 0.002$ for the Meatus score). These variables were 3.42 ± 0.67 and 2.75 ± 0.62 , respectively. The Shaft score was 2.33 ± 1.07 and did not lead to a statistically significant difference in complicated and uncomplicated cases ($p = 0.285$). GMS score was 8.50 ± 1.78 in complicated cases and 5.97 ± 2.01 in uncomplicated cases. GMS score was detected statistically significantly higher in complicated cases than in uncomplicated cases ($p = 0.001$) (Table 3). When GW was considered as a categorical variable, the number of cases with a GW of 14 mm or above was 23, 6 of which were complicated cases. In cases with GW below 14 mm, 6 of 21 cases were complicated. Thus, it was found that GW above and below 14 mm did not create a statistically significant difference in terms of complications ($p = 0.853$). Similarly, when the UP is taken as a categorical variable, the number of cases with a UP of 8 mm or more is 12, with 2 being complicated cases. In cases with a UP of less than 8 mm, 10 of 32 cases were complicated. Hence, it was found that UP above and below 8 mm did not cause

**Table 3.** Comparison results of variables according to the presence of complications

	Complicated		Non-complicated		*p
	Mean	SD	Mean	SD	
Age (months)	70	41.39	45.44	31.95	0.042
Duration of hospital stay (days)	12.75	6.66	8.53	4.63	0.022
Follow-up period (months)	11.75	5.05	8.94	5.7	0.141
Penis length (mm)	24.33	6.56	24.22	5.53	0.954
Glans width (mm)	14.33	3.85	13.75	3.03	0.600
Meatus distance (mm)	8.67	3.96	7.94	11.46	0.831
Urethral plate width (mm)	6.08	1.73	6.25	1.95	0.796
Operation duration (minutes)	113.75	36.38	101.72	47	0.429
Glans score	3.42	0.67	2	0.88	0.001
Meatus score	2.75	0.62	1.97	0.74	0.002
Shaft score	2.33	1.07	2	0.84	0.285
GMS score	8.5	1.78	5.97	2.01	0.001

*: Significance levels according to independent T-test results, SD: Standard deviation, GMS: Glans-urethral meatus shaft

Table 4. Statistical outcomes of GMS score groups according to the presence of complications

		Complicated		Non-complicated		*p
		n	%	n	%	
Conventional grouping	Mild GMS 3-6	1	4.50	21	95.50	0.001
	Moderate GMS 7-9	7	41.20	10	58.80	
	Severe GMS 10-12	4	80.00	1	20.00	
Recommended grouping	vs.					0.021
	Low score GMS 3-7	5	16.70	25	83.30	
	High score GMS 8-12	7	50.00	7	50.00	

*: Statistical significance level according to chi-square results, GMS: Glans-urethral meatus shaft

a statistically significant difference in terms of complications ($p=0.333$). According to the presence of complications, there was a statistically significant difference between the groups when GMS scores were grouped only as LS (cases with 3-7 points) and HS (cases with 8-12 points). HS was statistically significantly more likely to have complications ($p=0.021$). Also, a statistically significant difference was observed in the conventional grouping (mild, moderate, severe) according to the presence of complications (**Table 4**).

DISCUSSION

In some other studies, the mean age was lower and there was no statistically significant difference. In many articles in the literature, the mean age is lower than in our study. But, in similar studies conducted in Turkey, higher mean age is found more frequently. Whether the attitude of caregivers or the health policy approach causes this is worthy of further research.^{8,10-12} A shorter hospitalisation period was expected however, the fact that patients were not discharged with a catheter in our centre or caregivers' hesitation for post-operative care may have caused this situation. In various other studies, complication rate was found to be 14.1%, 18%, and 28.6%. Among these studies, the complication rates of the studies that mostly included proximal hypospadias cases were higher.^{8,13-15}

UP is one of the parameters used in predicting results and decision-making in hypospadias surgery. There is no statistically significant difference with UP according to the presence of complications, both as a continuous variable and as categorised for 8 mm. Statistical significance was found in some studies, but not in others. Variations in measurements may have been effective in these differences; in some studies, the widest part of the UP was taken while in others the width at the midline of the glans was taken.¹⁶⁻²⁰

However, in some studies, glans size was statistically significant, while in others it was not. Factors such as the age of the patients and preoperative hormone use may be causally associated with this difference.²¹⁻²³ GMS is a scoring system that determines the severity of hypospadias with four points from each section and 12 points in total. Inter-observer reliability tests of GMS scoring have been performed previously, this test has no measurement data and provides practical facilities. It has been in clinical use as a simple hypospadias assessment tool. The three sections are the G score, which evaluates the glans, the M score, which evaluates the meatus, and the S score, which evaluates the shafts. In addition to GW in the G score, the UP and the urethral groove condition are evaluated in the same line. In fact, in a previous study by Arlen et al.⁸ the higher value was chosen when there were controversies about these different parameters that coexist in the G score. Sometimes, these problems were avoided by considering only the GW.^{8,9,24,25}

The conventional grouping method that assesses the severity of hypospadias into three categories: mild, moderate, and severe leads to some challenges. Firstly, the concept of severity of hypospadias is susceptible to misinterpretation (for example, when multiple parameters within the Glans score can lead to discrepancy results, resulting in minor score variations from moderate to severe). The second problem is that if the scoring systems have a moderate group, designed for practical decision-making, forces the surgeon to take the initiative in deciding for cases within the moderate group. Our research addresses these challenges and suggests modifying the grouping system by reducing it from three groups to two which makes the definition clear. This revised approach highlights the distinction between LS and HS groups without emphasising focusing on the severity of the condition.^{16,21,22,26}

After obtaining the GMS score, all cases are grouped as mild, moderate, and severe. GMS and similar scores, which



are created to increase preoperative decision-making and predictions about postoperative results, may cause a relatively difficult situation for the surgeon because they contain an unclear moderate group.^{8,27} Although the shaft score portion of GMS has been associated with complication as an independent variable in some previous studies, it has been seen that there is no relation between shaft score and the presence of complication in our study ($p=0.285$). Other studies suggesting that the glans score can be divided into glans and urethra sections (Glans-Urethra-Meatus-Shaft scoring) have also been included in the literature. Our suggestion is a simplified scoring system in which the surgeons' approach and hypospadias results can be evaluated more simply rather than GUMS or similar systems.^{8,28}

A previous study reported that a GMS score above 6 increased the risk of surgical complications. According to the grouping we recommended in this study, the complication rate was 16.5% ($n=5$) in the LS (GMS 3-7) group and 50.0% ($n=7$) in the HS (GMS 8-12) group (Table 4).

Limitations

The most prominent limitation of this study is the small number of participants and short follow-up time. In the future, prospective studies with more participants may provide even more valuable and detailed insights into similar issues.

CONCLUSION

GMS is one of the most practical scoring systems that can effectively evaluate hypospadias. However, it should not be ignored that there are deficiencies in this scoring system. It seems that the simpler two-group method we proposed for the interpretation of the scoring may be an alternative in terms of ease of use. All these results demonstrate the need for a common language on hypospadias as well as the need for an objective system for the correct approach.

ETHICAL DECLARATIONS

Ethics Committee Approval

This study was approved by the local ethics committee of the Gülhane Training and Research Hospital, Clinical Researches Ethics Committee (Date: 02.03.2022, Decision No: 2022/25).

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.

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Anesthesia management in pediatric bronchoscopy

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ABSTRACT

Aims: The primary aim of this retrospective study is to analyze pediatric bronchoscopy cases performed under general anesthesia, evaluating the indications, findings, and adverse events.

Methods: The study included a total of 31 pediatric patients who underwent rigid and flexible bronchoscopy under general anesthesia at Bozok University Faculty of Medicine Hospital between July 1, 2018, and May 1, 2024. Demographic data, types of anesthesia, anesthetic agents, and length of hospital stay were recorded. Anesthesia management and surgical processes were described in detail.

Results: The study found that an increased duration of bronchoscopy was associated with a higher likelihood of laryngospasm and bronchospasm ($p=0.049$). Additionally, in line with the literature, the average age of patients included in our study who underwent bronchoscopy for diagnostic and therapeutic purposes was determined to be 33 months. The incidence of laryngospasm and bronchospasm was found to be 16.1%, which is consistent with other studies in the literature.

Conclusion: Our study indicates that foreign body aspiration occurs most commonly in male children under three years of age and that prolonged bronchoscopy durations increase the risk of laryngospasm and bronchospasm.

Keywords: Pediatric bronchoscopy, anesthesia, laryngospasm, bronchospasm, procedure duration

INTRODUCTION

Bronchoscopy is an instrument used to visualize the upper and lower airways for diagnostic and therapeutic purposes.¹ It is considered as one of the most important diagnostic and therapeutic procedures in pediatric respiratory diseases.^{2,3} The first report on diagnostic flexible bronchoscopy in children was published in 1978, and this method has revolutionized the management of pediatric respiratory diseases.^{4,5} Over time, bronchoscopy has become an increasingly important tool in children with acute and chronic lung diseases. In 2003, the European respiratory society (ERS) task force published results related to flexible endoscopy of pediatric airways, indicating that this procedure is safe, provided that children are adequately prepared and the procedure is performed by competent and trained personnel.⁶

The ERS pediatric bronchology group conducted a survey in pediatric pulmonology centers in Europe from 2012 to 2014, revealing that a total of 56,145 bronchoscopies were performed in 198 centers, with the vast majority of these procedures conducted under general anesthesia.⁷ This invasive endoscopic procedure is typically performed under deep sedation or general anesthesia to minimize the likelihood of movement in

children and to prevent respiratory defense reflexes.⁸ During bronchoscopy, benzodiazepines (midazolam), intravenous general anesthetics (propofol, etomidate, opioids), inhalational agents (sevoflurane, desflurane), or a combination of these anesthetic medications are generally used.⁹⁻¹¹

Many studies have shown that surgical factors, anesthesia management, and the stability of pediatric patients contribute to unwanted respiratory events.¹² Particularly, patients who experience foreign body aspiration are often younger than 3 years, and it has been reported that the shared use of the airway with the surgical team may increase the risk of complications.^{13,14} One of the most commonly encountered complications in pediatric bronchoscopy is hypoxemia; the literature indicates that prolonged procedure duration increases the frequency of these complications and also raises the risk of laryngospasm and bronchospasm.¹⁵

The primary aim of this retrospective study is to evaluate the indications, findings, and adverse events by analyzing all pediatric bronchoscopy cases performed under anesthesia between 2021 and 2024.



METHODS

Ethics

This study was approved by the Ethics Committee of Bozok University (Date: 06.03.2024, Decision No: GOKAEK-245_2024.06.05_76). This retrospective observational study was conducted at Yozgat Bozok University Faculty of Medicine Hospital, a tertiary healthcare center, this study included pediatric patients younger than 18 years old who underwent bronchoscopy procedure between July 1, 2018, and May 1, 2024. The study was conducted in accordance with the principles of the Helsinki Declaration.

Participants

A total of 31 pediatric patients who underwent rigid and flexible bronchoscopy under general anesthesia during the specified dates were included in the study. Patients under 18 years of age who underwent bronchoscopy for diagnostic or therapeutic purposes were included. Patients with incomplete data were excluded.

Clinical Data

Data on age, sex, weight, ASA physical status classification, admission diagnoses, type of anesthesia, anesthetic agents used, bronchoscopy technique applied, and length of hospital stay were recorded from the hospital automation system and file archive.

Anesthesia Management

No sedation or analgesia was required for premedication in any of the patients. Routine monitoring was performed upon transferring patients to the operating room [electrocardiography (ECG), non-invasive blood pressure, and pulse oximetry (SpO₂)], and a 22-24 gauge IV cannula was placed in the dorsum of the right hand. All patients received a crystalloid infusion (8 ml/kg/hour) throughout the surgical procedure. Preoxygenation was performed with 100% O₂ for 3 minutes. Standard anesthesia induction was applied to all patients [2-3 mg/kg propofol (Propofol 1%, Fresenius®), 0.6-1.2 mg/kg rocuronium (Esmeron, MSD®), and 1 µg/kg fentanyl (Talinat, VEM®)]. After the placement of the laryngeal mask airway (LMA), patients were ventilated in controlled mode with a tidal volume of 8 ml/kg, FiO₂ 0.6, I ratio of 1:2, and ET CO₂ of 30-40 mm Hg. For maintenance of anesthesia, sevoflurane [minimum alveolar concentration (MAC) 0.8-1.3%] was administered with 60% O₂ and 40% air. If rigid bronchoscopy was necessary, an additional 0.1 mg/kg of rocuronium was administered for muscle relaxation. Intraoperatively, supplemental fentanyl (0.5 µg/kg) was used as needed.

All patients received 0.1 mg/kg metoclopramide and 10 mg/kg paracetamol IV (Partemol 1g/100ml, VEM) for postoperative nausea and analgesia within the last 30 minutes of the procedure. At the end of the surgery, for patients who received muscle relaxants, residual neuromuscular block was antagonized with a combination of IV neostigmine (50 µg/kg) and atropine (20 µg/kg).

Statistical Analysis

The SPSS 20.0 software package was used for the analysis. Evaluations were performed using demographic data and general descriptive statistics (descriptive and frequency). The normality of the data distribution was assessed using the

Kolmogorov-Smirnov test, and it was determined that the data did not follow a normal distribution. Analyses of relevant data were performed using the Mann-Whitney U and Kruskal-Wallis tests. A p-value of <0.05 was considered statistically significant.

RESULTS

The analysis revealed that 67.7% (21) of the children who underwent bronchoscopy were male and 32.3% (10) were female. A foreign body was detected in 13 patients (41.9%). Complications developed in 5 patients following bronchoscopy (Table 1). The ages of the patients ranged from 6 months to 132 months, with a mean age of 33 months. Those with longer procedure durations had a higher incidence of bronchospasm and longer hospital stays (Table 2). All patients planned for flexible bronchoscopy underwent LMA placement. In 6 patients, it was determined that the procedure started with flexible bronchoscopy but continued with rigid bronchoscopy (Figure 1). A total of 13 patients (41.9%) had a foreign body detected (Figure 2). The development of desaturation and bronchospasm was more frequent in patients who underwent rigid laryngoscopy. All patients fully recovered during postoperative care. All patients who experienced complications were from the group with detected foreign bodies. Among the 13 patients with foreign bodies, 11 were male and 2 were female (Figure 3).

Table 1. Characteristics and percentage distribution of patients undergoing bronchoscopy

		n	%
Sex	F	21	67.7
	M	10	32.3
ASA	I	19	61.3
	II	10	32.3
	III	1	3.2
	IV	1	3.2
	LMA	19	61.3
Airway; R or LMA	R	8	25.8
	LMA+R	4	12.9
Foreign body	FB(-)	18	58.1
	FB(+)	13	41.9
	F	17	54.8
Type of bronchoscopy	R	8	25.8
	F+R	6	19.4
Bronchospasm/laryngospasm	B/L (-)	26	83.9
	B/L (+)	5	16.1

F: Female, M: Male, ASA: American society of anesthesiologists score, LMA: Laryngeal mask airway, R: Rigid bronchoscopy, F: Flexible bronchoscopy, FB: Foreign body, B/L: Bronchospasm/laryngospasm

DISCUSSION

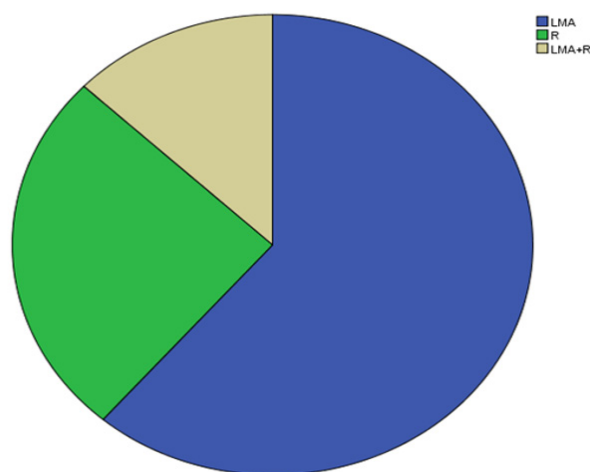
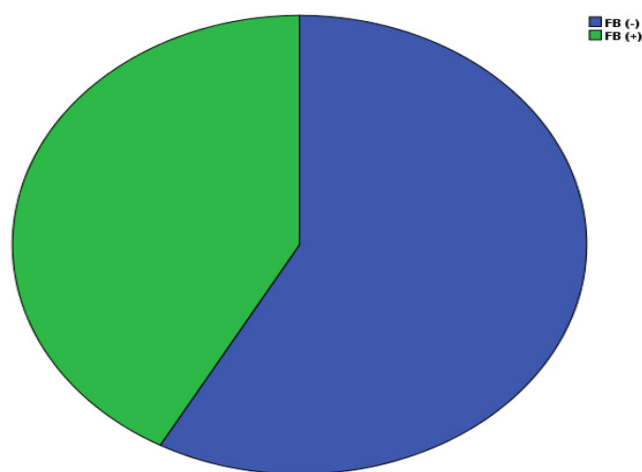
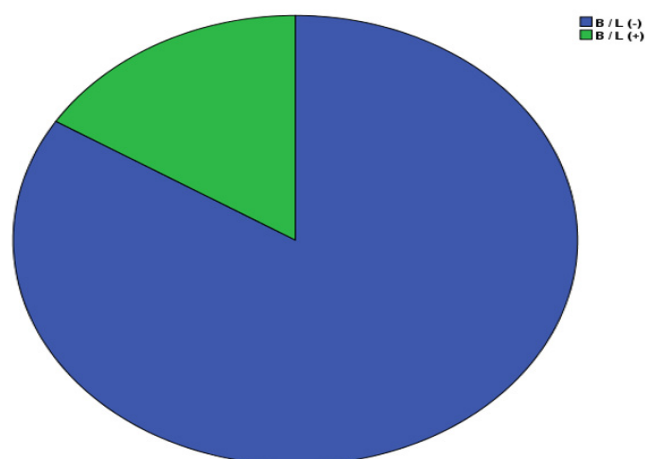
In our study, the prolonged duration of rigid bronchoscopy was found to increase the likelihood of developing laryngospasm/bronchospasm. Additionally, it was noted that longer bronchoscopy durations led to increased hospital stay.

The need for bronchoscopy due to foreign body aspiration is common in children aged 1-3 years.¹⁶ Massie et al.¹⁷ identified that the most prevalent age group was between 1 and 3 years. Studies conducted in our country have also shown that cases of foreign body aspiration predominantly occur in male children under the age of three.¹⁸ Consistent with the literature, our study revealed that the majority of patients undergoing

Table 2. Evaluation of airway complications

	Laryngospasm/bronchospasm								
	L/B(-)				L/B(+)				p
	Mean	SD	Min.	Max.	Mean	SD	Min.	Max.	
Age (months)	33.04	29.27	6.00	132.00	47.80	67.42	11.00	168.00	.893
Weight (kg)	15.15	7.20	8.00	40.00	19.60	17.13	10.00	50.00	.914
Procedure duration (minutes)	32.12	12.74	15.00	60.00	53.00	23.35	20.00	80.00	.049
Length of stay (days)	1.62	0.80	1.00	3.00	5.20	4.38	2.00	10.00	.017
Max: Maximum, Min: Minimum, SD: Standard deviation, p<0.05 indicates statistical significance.									

Max: Maximum, Min: Minimum, SD: Standard deviation, p<0.05 indicates statistical significance

**Figure 1.** Airway control rates
LMA: Laryngeal mask airway, R: Rigid bronchoscopy**Figure 2.** Results of foreign body detection in bronchoscopy
FB (+): Airway foreign body was found; FB (-): No airway foreign body was found**Figure 3.** Incidence of bronchospasm/laryngospasm in bronchoscopy
B/L (+): Bronchospasm/laryngospasm was developed; B/L (-): Bronchospasm/laryngospasm was not developed

bronchoscopy for foreign body aspiration were male children under three years of age.

Anesthesia in bronchoscopy can be categorized into three areas: the choice of anesthesia method, ventilation during bronchoscopy, and maintenance of anesthesia.¹⁹ However, there are different opinions in the literature regarding the anesthetic techniques used in pediatric bronchoscopy,^{20,21} with reports of applications using either sedation or general anesthesia. The choice of anesthetic technique and agents is determined based on the characteristics of each patient and the planned ventilation technique (spontaneous/controlled). For complex procedures involving both rigid and flexible bronchoscopy, the gold standard remains general anesthesia.²² Spontaneous or controlled ventilation techniques may be preferred for patients with foreign body aspiration, with both having their respective advantages and disadvantages discussed in the literature.²³ Several meta-analyses and reviews have reported lower frequencies of respiratory complications when controlled ventilation is applied.²⁴⁻²⁶ In our clinic, controlled ventilation is preferred for bronchoscopy procedures. Patients undergoing controlled ventilation typically require a deeper level of anesthesia. Some studies have reported the use of propofol combined with remifentanyl in anesthesia for flexible bronchoscopy in children.^{27,28} In our clinic, propofol and fentanyl are used for flexible bronchoscopy procedures, while intravenous general anesthesia is applied with the addition of muscle relaxants for rigid bronchoscopies. Anesthesia practices and ventilation methods should aim to minimize the risk of complications for the patient.

In children, the most common cause of acute asphyxia is tracheobronchial foreign body aspiration.¹⁸ Another danger associated with foreign body aspiration is the partial obstruction or mucosal edema that may occur due to the foreign body moving into the lower airways with strong respiratory efforts; this can significantly reduce lung ventilation and oxygenation.^{29,30} Particularly in small children aged 0-3 years, anesthesia management can become challenging due to the shared airway with the bronchoscopist. This situation can lead to a higher incidence of serious postoperative airway adverse events, such as laryngospasm and bronchospasm, and can quickly result in severe hypoxemia.^{31,32} In our study, the incidence of laryngospasm and bronchospasm was found to be 16.1%, which is comparable to the values of 17.6% and 17.4% reported in two retrospective studies evaluating complications of foreign body aspiration in China.^{31,33}

During bronchoscopy, it can be quite difficult to achieve and maintain an appropriate depth of anesthesia to prevent reflexes such as laryngospasm and bronchospasm while also sustaining spontaneous respiration.¹⁹ Some studies indicate that prolonged procedure times in bronchoscopy can lead to perioperative complications in children.^{23,33} Procedure time



has been reported as an independent risk factor for predicting intraoperative desaturation, laryngospasm, and bronchospasm during the removal of foreign bodies in pediatric patients.³⁴ Similarly, in a study by Zaytoun et al.²⁹ a significant increase in complication incidence was observed in patients whose bronchoscopy procedures lasted 30 minutes or longer. In our study, patients with longer bronchoscopy durations had a higher frequency of laryngospasm and bronchospasm, which aligns with the findings of Yu Cui et al.³³ and Maddali et al.³⁵

Limitations

Our study has several limitations. Firstly, being a single-center and retrospective study, the diversity of patients and sample size could not be expanded. Secondly, due to the involvement of different bronchoscopists during the evaluation period, procedure durations varied according to the level of experience. Lastly, while intraoperative desaturation occurred, there is uncertainty regarding the rational documentation of these events.

CONCLUSION

In conclusion, the majority of foreign body aspiration cases occur in male children under three years of age. The length of the procedure increases the likelihood of bronchospasm, independent of age and sex, and prolongs hospital stay. This study may contribute to the examination of pediatric bronchoscopy practices in larger sample groups through multicenter research and the comparison of the effectiveness of different anesthesia techniques, as well as the reassessment of clinical protocols for anesthesia management and procedure durations.

ETHICAL DECLARATIONS

Ethics Committee Approval

This study was approved by the Ethics Committee of Bozok University (Date: 06.03.2024, Decision No: GOKAEK-245_2024.06.05_76).

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.

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Evaluation of the effect of preoperative caudal epidural block on early complications of hypospadias in distal hypospadias surgery

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ABSTRACT

Aims: Hypospadias is an anomaly of the urogenital system with an incidence of approximately 0.5% in children. Caudal epidural block (CEB) is one of the most preferred analgesia methods in hypospadias surgery. Recently, there have been reports in the literature about an increase in postoperative complications (especially fistula) due to CEB after hypospadias surgery. In this study, we aimed to retrospectively evaluate the effect of CEB analgesia on surgical outcomes in distal hypospadias surgery performed in our center.

Methods: The study included 63 pediatric patients operated for distal hypospadias between 2020 and 2023. These patients were divided into two groups according to whether CEB was applied during surgery. Patients were evaluated in terms of surgical technique, age, and complications.

Results: Sixty-three pediatric surgeries with a mean age of 45.12 ± 26.76 months ranging from 1 to 10 years were analyzed. Sixty-three cases with subcoronal hypospadias were included in the study. Twenty-two cases (41.3%) underwent CEB, while 37 cases (58.7%) received intravenous analgesia (IA). The surgical complication rate in our study was 30.1% (n=19). There was no significant difference in postoperative complications between CEB and IA groups ($p=0.197$).

Conclusion: In this study, there was no statistically significant difference in the incidence of postoperative complications between CEB and IA in children operated for distal hypospadias. CEB can be used as a safe pain management method in hypospadias repair. However, we think prospectively designed studies with more patients will be more helpful in clarifying the issue.

Keywords: Anesthesia, epidural, caudal, child, hypospadias, postoperative complications

INTRODUCTION

Hypospadias is one of the most common congenital urogenital anomalies, with a prevalence of approximately 0.5%.¹ There is still controversy about the analgesic methods used for hypospadias surgery in the perioperative period.

The complication rate of distal hypospadias surgery varies between 5% and 10%. Each procedure performed after the complication develops increases the complication rate even more.^{2,3} The most common long-term complications are fistula, meatal stenosis, tissue or glans separation, chordee, and diverticulum.^{2,4}

In hypospadias surgery, it is essential to reduce postoperative pain in the early postoperative period until routine oral analgesia is initiated. There are many methods for early postoperative pain relief; penile block, caudal epidural block (CEB), and intravenous/ oral analgesics are frequently used for this purpose. CEB and intravenous analgesia (IA) are the most used methods among these methods. However, publications indicate that CEB, which has recently been widely used in hypospadias surgery, causes more postoperative complications.⁵⁻⁸ Our study aimed to compare these two methods regarding postoperative surgical complications.



METHODS

The study was conducted with the permission of the Clinical Researches Ethics Committee of University of Helath Sciences Gülhane Training and Research Hospital (Date: 27.09.2023, Decision No: 2023/209), the records of 103 hypospadias cases performed between 2020 and 2023 were retrospectively analyzed. All procedures were carried out in accordance with the ethical rules and the principles of the Declaration of Helsinki. Data such as operative procedures, patients' ages, or outcomes were obtained from patient digital archives. Patients who underwent their first operation in a different center and operated in our center due to a fistula or another complication were excluded from the study. A total of 63 patients who were operated on for distal hypospadias, who had not undergone previous surgery, and who underwent tubular incised plate urethroplasty (TIPU) technique were included in the study. The pressure dressing was opened on the 3rd day, the urethral catheter was withdrawn on the 7th day postoperatively, and the patients were discharged. Patients were followed up at 1 week, 1 month, 3 months, 6 months, 1 year, and then annually after discharge. Surgical complications (according to the Clavien-Dindo grading system, Grade III complications), fistula development, and meatal stenosis were evaluated.

Patients were divided into two groups: CEB and IA groups. In the IA group, paracetamol IV at 10 mg/kg and pethidine HCL at 0.5 mg/kg/dose were administered at the end of surgery (before extubation). In the CEB group, CEB (0.2 ml/kg 0.25% bupivacaine) was performed using a Tuohy needle under preoperative ultrasound (USG) guidance. In both groups, IA was given at the 4th hour postoperatively, and oral analgesia was given afterward.

Statistical Analysis

The Kolmogorov-Smirnov and Mann-Whitney U tests were used to compare the data, and the Student t-test was applied after normal distribution. Jamovi open-source software package version 2.3.26 was used for all statistical procedures.

RESULTS

The study included 63 patients, divided into the CEB group (n=26) and the IA group (n=37). The mean age was 45.12±26.76 months (12-108 months) in the CEB group and 47.28±25.56 months (12-120 months) in the IA group. The difference between the ages was not statistically significant (p=0.197).

Our study's surgical complication rate (Clavien-Dindo III grading system) was 30.1% (n=19). When the patients were evaluated in terms of complications (urethral fistula, meatal stenosis), 4 (15.3%) of a total of 14 (22.2%) urethral fistula cases were observed in the CEB group and 10 (27.0%) in the IA group. The sum of the complication rate for meatal stenosis was 5 (7.9%) patients, such as 3 (11.5%) in the CEB group and 2 (5.4%) in the IA group. Patients in both groups responded thoroughly to one-time dilatation. There was no statistically (Chi-Square) significant difference between IA and CEB groups in terms of development of urethral fistula (p=0.639) and meatal stenosis (p=0.375) (Table).

The mean age of patients who developed complications was 51.24±25.44 months (12-120 months), and the mean age of patients who did not develop complications was 43.8±25.32 months (12-108 months); this difference was not statistically significant (p=0.407).

Table. Development of fistula and meatal stenosis after CEB and IA in pediatric patients with distal hypospadias and age correlation

	CEB	IA	Total
n	26 (41.3%)	37 (58.7%)	63 (100%)
Mean age (months)	45.12	47.28	46.38
SD	26.76	25.56	25.56
Min. (months)	12	12	12
Max. (months)	108	120	120
Complications	7	12	19
Meatal stenosis	3 (15.6%)	2 (10.4%)	5 (26%)
Fistula	4 (21.5%)	10 (52.5 %)	14 (74%)

CEB: Caudal epidural block, IA: Intravenous analgesia, SD: Standard deviation, Min: Minimum, Max: Maximum

DISCUSSION

Hypospadias surgery and related complications are still a challenging problem for pediatric urologists. Numerous surgical techniques to cope with this issue prove this requirement. Another issue that challenges physicians in hypospadias surgery is the fact that different results are obtained in cases with hypospadias of almost similar severity degree despite performing as standardized as possible surgical procedures.⁵⁻⁹ This issue has necessitated a detailed examination of the dressing, analgesia, and surgery materials. Recent studies have raised questions about the effect of perioperative analgesia methods on the success rates of hypospadias surgery. Some of the studies, which are pretty limited in number, report the negative impact of CEB on surgical outcomes, while others claim that it does not affect postoperative outcomes.⁹⁻¹³

In a study by Kim et al.¹⁴ it was reported that caudal block caused pelvic congestion and associated penile edema during hypospadias repair and could potentially cause postoperative complications, especially urethral fistula. The primary physiopathological mechanism is vasodilatation in the penile sinuses due to sympathetic block and consequent pelvic venous stasis.¹¹ In our series, significant penile edema was observed in both groups, and it is often not possible to avoid penile edema in surgery of this region. Therefore, since no significant penile edema difference was observed in both groups, we think it is impossible to differentiate postoperative complications related to the analgesic method from surgical penile edema. Our opinion is consistent with the data in a multicenter study by Hu et al.¹⁵

Development of urethral fistula is one of the most common complications after hypospadias surgery. Fistula development after distal hypospadias was reported to be between 0-65% in a meta-analysis, including 17 studies.¹⁶ In our study, the rate of the urethral fistula was 15.3% in the CEB group, 27% in the IA group, and 22.2% in total. No statistically significant difference was found between the groups (p=0.274). In a study by Kim et al.¹⁴ it was reported that the rate was 2.1 times higher in patients who underwent caudal penile block compared to those who did not. Kundra et al.⁵ also associated CAB with a high surgical complication rate (especially in terms of urethral fistula development) due to pelvic congestion in hypospadias patients. We think that the high rate of urethral fistula in our series is because we are a pediatric surgery training hospital.



In the same study, the rate of meatal stenosis developing after hypospadias repair was reported between 0-25%. The rate of development of meatal stenosis was calculated as 5%.¹⁶ In our study, the rate of meatal stenosis was 7.9%, slightly higher than the general average. We attribute this slightly higher rate to the limited number of cases. There were no statistically significant differences between the analgesia methods and the development of meatal stenosis between the groups in our study ($p=0.375$).

Regarding the rate of meatal stenosis after hypospadias repair, the mean age of the patients in our study was 3.76 ± 2.23 years (1-10 years). The total complication rate of meatal stenosis was 17.46%. When the mean ages of patients with and without complications were analyzed, a statistically significant difference was observed between age and the risk of surgical complications ($p=0.045$).

Caudal block is a preferred regional analgesia technique widely used in sub-umbilical procedures in children due to its safety and cost-effectiveness.¹⁷⁻¹⁹ CEB in children provides a good balance between protection and risks in the perioperative period. Data from a large European multicenter study conducted by Ecoffey and colleagues²⁰ show that regional anesthesia is safe and has a meager overall complication rate of 0.12% in children. However, CEB has some complications, even quite rare, such as epidural hematoma, dural penetration, intravascular injections, overdose, and micturition disorder. In our study, no complications related to CEB were developed.

Penile block is one of the most commonly used postoperative analgesia methods in hypospadias surgery. When the literature was reviewed, it was seen that most of the studies compared the effects of CEB and penile block. It has been reported that the analgesic substance given to the area in the penile block may partially decrease the nutrition of the tissues in the surgical area with both volume and vasoconstriction effects.⁵⁻⁸ Therefore, we think it would be more appropriate to compare CEB with intravascular analgesics to evaluate the surgical complications.

Limitations

The main limitations of this study are that it was not prospective, the number of patients was limited, and the evaluations were not performed using visual analog scale (VAS). Since our study was not prospective, the selection of patients for the IA or CEB group was based on the selection at that time and was not standardized. It has also been disregarded by potential confounding factors such as the meatus's position, the chordee's level, and the surgery duration.

CONCLUSION

In our study, CEB did not increase complications after hypospadias. Therefore, we think CEB is a safe method for postoperative analgesia in hypospadias surgery. However, prospectively designed studies in larger groups would more effectively clarify the issue.

ETHICAL DECLARATIONS

Ethics Committee Approval

The study was conducted with the permission of the Clinical Researches Ethics Committee of University of Helath Sciences

Güllhane Training and Research Hospital (Date: 27.09.2023, Decision No: 2023/209).

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.

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Management of spontaneous pneumothorax in children

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ABSTRACT

Primary spontaneous pneumothorax (PSP) is defined as air or gas accumulation in the pleural cavity. It can occur spontaneously or secondary to an underlying pulmonary disease or after a trauma. Spontaneous PSP has been traditionally categorized as primary or secondary. Primary spontaneous PSP affects commonly young healthy people especially tall and thin adolescent males. Secondary cases may occur in patients with underlying pulmonary disease. The main aim in the treatment of PSP is to empty the air in the pleural cavity and allow the lung to re-expand. There are two main approaches for the treatment of spontaneous PSP; non-operative management options include bed rest, oxygen supplementation with watch-and-wait option, needle aspiration and surgical management options ranges from chest tube insertion to more invasive intervention such as lung resection, pleurodesis or bullae excision via thoracoscopy or thoracotomy. There is still a lack of consensus in the diagnostic approach and treatment strategies in PSP in children. Therefore, we aimed to review the clinical findings, radiological imaging, treatment options and surgical strategies in children with PSP.

Keywords: Spontaneous pneumothorax, diagnose, surgery, children

INTRODUCTION

Primary spontaneous pneumothorax (PSP) is collection of air in the pleural cavity, between the lung and the chest wall and it was firstly described in 1891 by Laennec.¹ At that time, most of the cases with PSP were secondary to tuberculosis and some others occur in healthy patients with no underlying disease. The first description of PSP in healthy people was described by Kjaergaard² in 1932.

The mechanism of air collection in the pleural space may occur due to communication between the alveolar spaces and the pleura, direct or indirect communication between the atmosphere and the pleural space and presence of gas-producing organisms in the pleural space.³

It can be categorized as spontaneous, iatrogenic, traumatic or tension PSP. Spontaneous PSP has been traditionally classified as primer or seconder. Primer spontaneous PSP is defined as PSP occurring in patients without lung disorders, whereas secondary spontaneous PSP occurs in the setting of an underlying pulmonary disease.

The incidence of PSP is reported to be 7 to 18 per 100 000 population per year in males and 1 to 6 per year in females.⁴ Spontaneous PSP is a relatively rare condition in children. The peak age of occurrence in this age group is bi-modal

and most of the cases occur either in the neonatal period or in late adolescence.⁵ It tends to occur in adolescence with a predominant male occurrence.⁶

The most important risk factor for PSP is smoking.⁷ Cannabis also increases the risk of PSP and multiple large peripheral bullae at the lung apex are often seen in young patients with a history of cannabis smoking without any significant parenchymal damage.⁷

In addition to smoking, levels of air pollution, pleural porosity, changes in atmospheric pressure or exposure to loud music are also involved in the etiology of PSP.^{8,9}

PSP usually occurs at rest; it presents with acute onset of local pleuritic chest pain accompanied by shortness of breath and may also be occur due to increased intrathoracic pressure. Procedures for the treatment of spontaneous PSP include observation, needle aspiration, insertion of intercostal catheter or invasive surgery procedures such as thoracotomy or thoracoscopy. Patients with prolonged chest tube drainage may require surgical intervention to remove the bullae or blebs and to create a pleural adhesion to avoid recurrences. There is no consensus on the timing and type of surgical intervention and best method for pleurodesis to prevent recurrence. Clinical



guidelines and consensus reports have been developed for adult patients but controversy still exists for the diagnosis and treatment of PSP in children. Herein, it is aimed to review the existing multidisciplinary approach to PSP including the clinical evaluation, radiological imaging, treatment options and surgical strategies.

PATHOPHYSIOLOGY

The pathogenesis of PSP is not completely understood. For many years the accepted theory for PSP is that the subpleural bleb or bulla opens spontaneously and begins to leak air.¹⁰ However, this theory has been challenged recently because blebs and bullae do not always the cause of PSP. Bullae are air filled spaces larger than 1 cm in diameter which located distal to terminal bronchioles and are associated with pulmonary tissue damage.¹¹ Blebs are localized collections of air between the lamina elastica interna and externa of the visceral pleura. Both of these parenchymal abnormalities can be described the term 'emphysema-like changes' (ELCs).¹¹ There are areas of weakness of the visceral pleura which are prone to rupture allowing air to leak into the pleural space.

PSP occurs in patients with no associated lung disease, which does not mean that there is no underlying pathological process. Abnormal pleura is very common in PSP patients.³ The macroscopic appearance of the visceral pleura has been shown to be abnormal in patients with PSP. It has been reported that in 77% of patients with PSP having blebs or bullae seen at thoracoscopy at the first episode.¹² Chronic distal airway inflammation with lymphocyte and macrophage infiltration alongside fibrotic changes are shown in the microscopic appearance of areas of lung tissue resected from patients with PSP.¹³

PSP usually occurs in tall, thin males, between 10 to 30 years-of-age. The gradient of negative pleural pressure increases from the lung base to the apex, so that alveoli at the lung apex is significantly greater distending pressure than those at the base of the lung so this theory can explain the development of apical subpleural blebs in tall individuals.¹⁴

Some inherited disorders may predispose to PSP, such as Marfan's syndrome, Birt-Hogg Dube syndrome, other mutations of the folliculin gene (FLCN), alfa1-antitrypsin deficiency and homocystinuria and approximately 10% of patients with PSP have a positive family history.¹⁵ Female patients with these syndromes, pregnancy and childbirth pose a risk for PSP and women should be particularly informed for the risk of PSP development.¹⁶

Any patient presenting with first PSP had familial syndrome should have CT scan of the chest and a referral genetic test should be considered.¹⁶

The accumulation of neutrophils and macrophages leads to an inflammation by disrupting elastin fibers in smoking patients so that smoking is a major risk factor for PSP. Smoking cessation is the only reversible risk factor known to reduce the likelihood of recurrence.⁷ The cause of the inflammation seen without a history of smoking is still unexplained in PSP patients.

In addition to enviromental factors, structural alterations may play role in the development of PSP. It has been reported that

patients with PSP have emphysematous-like histology and may develop bullae and blebs.⁶ Specific microRNAs (miRNA) may play critical role in inflammation and regulation of eosinophil activation. Previous reports showed that up-regulation of miRNA-24 expression may relate with fibrosis and chronic alterations in the airways of children with PSP.¹⁷ The miRNA-24 expression in the exhaled breath condensate (EBC) of children with PSP significantly down-regulated and suggested that patients especially with bullous disease, should be closely monitored in the long-term period.¹⁸

SSP is commonly associated with chronic obstructive pulmonary disease but also with other underlying important lung disease, including cystic fibrosis, tuberculosis, diffuse parenchymal lung disease.¹⁹

Tension PSP is more common with traumatic reasons or with ventilated patients and extremely rare in spontaneous PSP. It occurs when the intra-pleural pressure becomes greater than atmospheric pressure and is caused by one-way valve effect whereby air passes into the pleural space on inspiration. When the pressure becomes negative, air in the airway unable to escape on expiration due to closing of the valve. It is an emergency situation when compared to other pneumothoraces because the intrathoracic pressure gradually increases and requires rapid intervention.

CLINICAL EVALUATION AND IMAGING

Sudden onset of chest pain, difficulty breathing and tachycardia are the predominant symptoms of PSP in children as in adults. The typical symptoms of chest pain and dyspnea may be relatively minor or even absent, so that a high index of suspicion is required. In general, the clinical symptoms associated with SSP are more severe than those associated with PSP.

The physical signs of a PSP include reduced lung expansion and breath sounds on the side of the PSP. In addition to these signs, cyanosis, sweating, severe tachypnoea, tachycardia and hypotension can be seen presence of tension PSP.

Posterior-anterior chest radiography is the standard examination in patients with suspected PSP. It has been reported that inspiratory and expiratory radiographs are equally sensitive in detecting PSP and therefore expiratory radiographs do not need to be taken routinely.²⁰ Besides these techniques, computed tomography (CT) is more sensitive than chest radiographs and it can be regarded as the gold standard in the detection of small pneumothoraces and size estimation (Figure 1).²¹ CT is also useful in the presence of surgical emphysema and bullous lung disease and for identifying additional lung pathology.²² However, it is not required in the majority of cases, as chest radiograph is usually sufficient to make a diagnosis, thus young patient population is avoided from excessive radiation dose. However, prolonged chest tube drainage and continuous air leak from the chest tube is considered as clear indication to obtain a CT scan.

There is an artificial intelligence (AI) report on chest ultrasound, which is considered to be more sensitive than chest radiography and this is a promising area of research.²³ AI models are only useful as a screening tool to determine the presence or absence of PSP, but future studies are needed as they do not incorporate the individual patient's circumstances and other medical conditions.²⁴

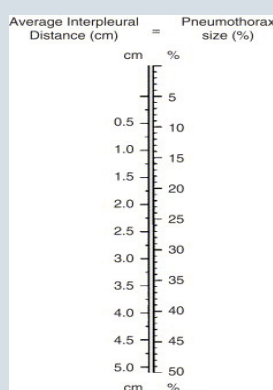


Figure 1. CT image of PSP
CT: Computed tomography, PSP: Primary spontaneous pneumothorax

Moreover, the plain PA chest X-ray has been used to quantify the size of the PSP. However, it tends to estimate the size because it is a two-dimensional image while the pleural cavity is a three-dimensional structure. There are three methods can be used to measure size of the PSP; Light method,²⁵ Collins method,²⁶ Rhea method²⁷ (Table). Although treatment and follow-up of recommendations are based on size of the PSP in different guidelines, there is no consensus on the definition of 'large PSP' and best method to estimate the size of PSP.

Table 1. Formulas used for determining of the size of the pneumothorax

Light	$\% \text{ of pneumothorax} = 100 - (D_L^3 / D_H^3 \times 100)$ D_L is the diameter of the collapsed lung D_H is the diameter of the hemithorax on the collapsed side
Collins	$\% \text{ of pneumothorax} = 4.2(4.7(A+B+C))$ A: distance between partially collapse lung apex to the apex of thoracic cavity B: mid points of upper half of collapsed lung C: mid points of lower half of collapsed lung
Rhea	Measuring average interpleural distance (AID) and using the normogram $AID: A \times B \times C / 3$ A: distance between partially collapse lung apex to the apex of thoracic cavity B: mid points of upper half of collapsed lung C: mid points of lower half of collapsed lung



The British Thoracic Society (BTS) Guideline uses interpleural distance at the level of the hilum, accept that a 2 cm radiographic PSP approximates to a 50% PSP by volume, it is called large PSP.²⁸ The American College of Chest Physicians (ACCP) uses the distance between apex to cupola for calculation of size of PSP and accept that ≥ 3 cm radiographic PSP is large PSP (Figure 2).²⁹ The Belgian Guideline uses Light index and pleural gap along the entire length of the lateral chest wall which is definition of large PSP.³⁰ Comparison between the techniques

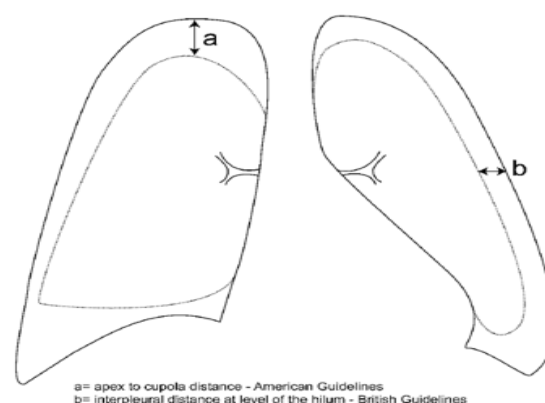


Figure 2. Schematic appearance of the measurement method for depth of the pneumothorax

in different guidelines have shown poor agreement.³¹ In a multivariate analysis, Choi et al.³² found that the presence of blebs on high resolution CT scans and chest X-rays were independent risk factors for ipsilateral recurrence. The European Respiratory Society (ERS) task force statement indicates that cross sectional imaging can be useful in complicated cases, where underlying disease is suspected and in patients requiring surgery.⁷ The ACCP consensus panel did not recommend the routine use of CT imaging for the patients with first episode of PSP.²⁸ In addition, no consensus has been achieved by the experts, regarding the utility of CT scans for evaluating the patients with recurrent PSP, persistent air leaks and as a preoperative imaging modality. In a survey by the European Pediatric Surgeons' Association (EUPSA), it has been found that the use of CT scans in PSP mostly depends on the surgeon's personal preference instead of evidence-based recommendations.³³

Compared the use of ultrasound instead of chest X-rays for follow-up of patients evacuated for PSP and found that ultrasound was superior to chest X-rays for follow-up.³⁴ It requires a trained team and access to ultrasound so that it can be used as an alternative to radiography.

In conclusion, when a child presents with sudden onset of chest pain or difficulty breathing, a detailed anamnesis should be taken and PSP should be suspected especially if there is a difference in breath sounds on physical examination. A chest radiograph should be ordered. A chest X-ray scan may support the diagnosis by showing air in the intrapleural space.

TREATMENT OPTIONS FOR PNEUMOTHORAX

The main aim in the treatment of PSP is to drain the air from the pleural cavity to allow the lung to re-expand and prevent recurrence. The treatment of PSP is lacking a definitive consensus to date and it is still controversial.

There are five main therapeutic approaches for the treatment of spontaneous PSP:³⁵

1. Conservative approach
2. Ambulatory device such as a flutter valve
3. Needle aspiration using a small-bore cannula or aspiration kit

4. Intercostal drain attached to a drainage system
5. Surgery for either acute or preventative treatment.

NON-SURGICAL MANAGEMENT OF PSP

The distinction between PSP and SSP is based on data showing differences in treatment outcomes, duration of air leakage and suitability for surgical resection.

The initial management of PSP depends on the size of the PSP and the clinical condition of the patient. Non-operative treatment options include bed rest, oxygen supplementation with watch-and-wait option or needle aspiration. The air in the pleural cavity is absorbed four times faster with oxygen supplementation, therefore, nasal oxygen therapy is used in conservative follow-up.³⁶ In addition, there is a group that does not recommend the systematic use of oxygen therapy in patients treated for PSP because of poor methodological quality of data from previous studies and the many potential disadvantages, such as additional costs and hospitalization.³⁷

While there are no clear recommendations for choosing a treatment option, previous adult guidelines have been predominantly based on clinical symptoms, PSP size and number of episodes. ACCP consensus reports recommend a more invasive approach considering the size of the PSP, while BTS guidelines recommend a conservative approach based on clinical symptoms.^{28,29}

According to BTS guidelines, observation is the treatment of choice for small PSP (size <2 cm) without significant breathlessness. Asymptomatic patients with large PSP can also be managed with observation alone. Needle aspiration (NA) is first choice for patients with large PSP (size >2 cm) with breathlessness. If not successful, chest drain is recommended.²⁸

According to ACCP guidelines, clinically stable patients with small PSP (size <3 cm apex to cupola distance) should be observed in the emergency department for 3 to 6 hours. Clinically stable patients with large PSP (size ≥3 cm apex to cupola distance) should undergo a procedure to re-expand the lung and should be hospitalized. The lung should be re-expanded by using a small-bore catheter or chest tube. Clinically unstable patients with large PSP should undergo hospitalization with insertion of a chest catheter.²⁹

According to BTS guidelines, breathlessness in an adult with PSP is an indication for active intervention.²⁸ The BTS guidelines highlight the use of NA as an initial intervention in large and/or symptomatic patients because of NA is as effective as chest drains and is less painful than chest drains.²⁸ However, Garipey et al.³⁸ showed that in contrast to the adult series, there was no advantage of the use of needle aspiration in children. But NA is failure in approximately one-third of patients will require a second procedure²⁸ so the ACCP recommend chest drain insertion.²⁹

Complications are rarely reported and the risk is lower with NA than with chest drain insertion but chest drain insertion shows a higher success rate than NA.³⁹ Pediatric surgeons responding to a survey of members of the European Pediatric Surgeons' Association showed that they mostly preferred chest tube and pigtail placement over simple aspiration (Figure 3).³³

Suction is another option for re-expansion of the lung. Suction is not recommended routinely, because it may cause

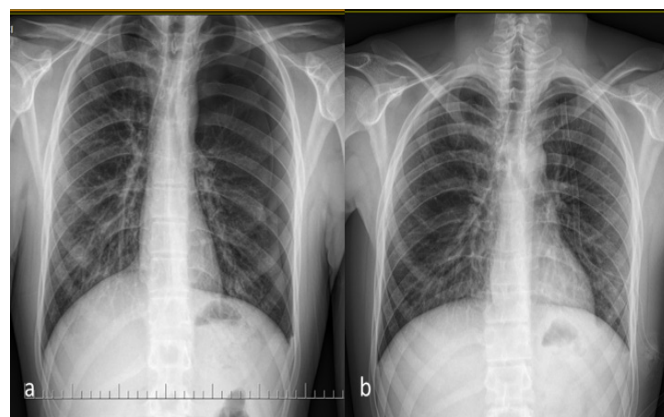


Figure 3. a) Chest radiograph of pneumothorax b) Chest radiograph of primary spontaneous pneumothorax after chest tube, left hemithorax is re-expanded

of re-expansion pulmonary oedema (RPO).²⁸ RPO may occur in some PSP patients followed by chest tube insertion and drainage procedure. It is rare complication but life-threatening, caused by rapid pulmonary re-expansion in especially young patient with large and persistent PSP.

When ensure that the air leak into the pleural space has resolved, chest tube can be removed. Several studies have evaluated attempted clamping before tube removal and one retrospective study found more frequent recurrence after chest tube drainage removal compared with no attempted clamping.⁴⁰ In a consensus statement, 53% of panel members reported that they would never clamp a chest tube to detect the presence of an air leak.²⁹

In a survey, surgeons performing thoracoscopic surgery preferred shorter duration chest tube drainage compared to those performing open surgery.³³

The current survey revealed that the majority of The Turkish Association of Pediatric Surgeons (TAPS) members were influenced the size of the PSP for choosing treatment options. They prefer tube thoracostomy as the first treatment, the second preferred option was oxygen administration. Needle aspiration option was rare and none of them preferred surgical management as the initial.⁴¹

All patients with SSP should be admitted to hospital and most patients will require chest drain. Because SSP is less likely to be tolerated by patients than PSP due to associated lung disease.^{28,29} Aspiration is less successful in SSP but can be considered in symptomatic patients with small pneumothoraces to avoid chest drain.²⁸

Medical chemical pleurodesis can be used for difficult or recurrent pneumothoraces but surgical options are now more effective. Several agents have been used for chemical pleurodesis including talc, tetracycline, autologous blood, fibrin glue. Large particle talc does not lead to the acute respiratory distress syndrome, therefore it is safe in use and most effective sclerosing agent.⁴² So talc is the most widely used agent for chemical pleurodesis in Europe.⁷ It can be used if a patient is unwilling or unable to surgery. Inflammation of the pleura causes dense adhesions so that the pleura adheres to the thoracic wall. This method may prevent recurrence but makes it difficult to perform thoracoscopy or thoracotomy in the future. In addition, chemical pleurodesis is very painful compared to thoracotomy.¹¹

Conservative treatment was superior to non-conservative treatment, even if eight weeks were required for radiographic



resolution. This strategy reduced the number of procedures, length of hospital stay, adverse event rate and days off work.⁴³

SURGICAL MANAGEMENT OF PSP

Seventy percent of patients having their first PSP never relapse, so performing surgery at first episode PSP seems like too invasive.⁴⁴ Therefore the other guidelines for the management of patients with PSP recommend air removal from intrapleural space via NA or chest tube insertion firstly in patients with PSP, surgery should not be performed as first line procedure.³⁷

Surgical management of PSP ranges from chest tube insertion to more invasive intervention such as lung resection, pleurodesis or bullae excision via thoracoscopy or thoracotomy.

The objectives of surgical treatment are surgical repair of persistent air leaks and prevention of recurrence. The first objective is to identify any visible bullae or blebs on visceral pleura and resect this part, the second objective is to create adhesion between pleural surfaces. There is no evidence on when is the ideal time for thoracic surgery in cases of persistent air leak. Although 5 days was previously considered the cut-off point, some studies have shown that persistent air leakage lasting longer than 7 days can be controlled without surgery.⁴⁵

There is also study showing that prolonged delays more than 5 days reduces the effect of thoracoscopy.⁴⁶

In online survey of EUPSA, they found that presence of bullae >2 cm (77%) and recurrence (76%) were accepted as the most common indications for surgical treatment.³³

Although there is a controversy still exists about the timing surgery, some indications for surgery is well accepted by many surgeons;²⁸

1. Second ipsilateral PSP
2. First contralateral PSP
3. Synchronous bilateral spontaneous PSP
4. Persistent air leak (despite 5-7 days of chest tube drainage) or failure of lung re-expansion.
5. Spontaneous hemothorax
6. Professions at risk (i.e, pilots, divers)
7. Pregnancy

Open thoracotomy with pleural abrasion was the surgical treatment for PSP, described by Tyson and Candall⁴⁷ in 1941. Previous experience of surgeons performing open thoracotomy with parietal pleurectomy has shown that the method is highly effective and has low PSP recurrence rates.⁴⁸

The traditional open approach has gradually been replaced by minimal invasive surgery. At the moment, video-assisted thoracoscopic surgery (VATS) has become the “gold standard” of PSP treatment, as evidenced by numerous medical reports from around the world.⁴⁹

Re-operation following VATS is more often required than open thoracotomy with a high rate of both late recurrent pneumothoraces, prolonged early postoperative air-leakage.⁵⁰ Some clinical trials including PSP patients showed that thoracoscopy offered no superiority over limited thoracotomy.⁵¹ However, VATS is significantly superior to thoracotomy in terms of less postoperative pain, better wound cosmetics,

shorter hospital stay, better functional recovery and patient satisfaction.⁴⁹

During surgery may undergo bullectomy with patients with PSP, electrocoagulation, laser ablation, stapling, Ligasure, endo-loop or manual suturing can be used. In addition to bullectomy, adhesion should be created between the two opposing pleura to prevent recurrence.²⁹ In the past, pleural abrasion was widely used for surgical pleurodesis. Some studies have shown that partial pleurectomy is more advantageous than pleural abrasion and that the combination of both techniques leads to better results.⁵²

In one study the surgical treatment of PSP with mesh coating was found to be significantly advantageous compared to thoracoscopic bullectomy, allowing for a lower recurrence rate, hospital stay and chest tube drainage time.⁵³

Medical thoracoscopy with talc poudrage has been shown to be a safe and effective technique for recurrence prevention of both PSP and SSP. Talc poudrage with medical thoracoscopy has similar recurrence rates to VATS, so it is most commonly used approach in current practice.¹⁹

In patients where urgent intervention is not required, management options largely depend on the patient's priorities and available local services. For example, a patient may need to fly in the near future. The current advice is that passengers should not fly until 7 days after resolution on the chest radiograph and that there is a high risk of recurrent PSP.⁵⁴

After thoracic surgery, patients should be informed that they should delay their flight 4 weeks for non-essential air travel and 2 weeks for essential air travel.⁵⁴

Our study represents a review of the available literature and is important in demonstrating the differences in approach in various guidelines. However, the weakness of our study is that most of the studies included adult patients and the guidelines were aimed at adults. We would like to emphasize that it is important to conduct more studies on pediatric patients and to follow the current literature with the developing technological developments.

CONCLUSION

The precise incidence of PSP is unknown but it has become clear that PSP is a disease associated with abnormalities within the pleura. The management PSP shows variability not only in conservative management, but also in surgical treatment. Clinical outcome and underlying pathologies variable, it is difficult to recommend a treatment algorithm for PSP cases. Most of current treatment recommendations are based on adult studies, there is no standard care and treatment for PSP in children. With the development of new technologies, more large-scale randomized studies are needed to better classify PSP and to establish new approaches in the diagnosis, treatment, follow-up of patients and the lack of a guideline for pediatric age groups is one of the most important reasons for the need for new studies.

ETHICS COMMITTEE APPROVAL

Referee Evaluation Process

Externally peer-reviewed.



Conflict of Interest Statement

The authors have no conflicts of interest to declare.

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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.

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Suspecting the unsuspected: spontaneous colonic perforation in healthy children

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ABSTRACT

Colonic perforation is a notorious entity in surgical domain attributing to high morbidity and mortality. Luckily enough this is a rare and sporadic event in pediatric patients, specifically when it comes to spontaneous idiopathic type. The neonates are known to have colonic perforations due to certain known conditions like Hirschsprung's disease, necrotizing enterocolitis, anorectal malformation, meconium ileus and atresia. Beyond infancy the incidence of spontaneous colonic perforations has a steep decline owing to diminution of congenital and developmental causes. In this case report we bring forth a case that we encountered as an acute abdomen secondary to idiopathic colonic perforation.

Keywords: Colonic, child, spontaneous, perforation

INTRODUCTION

Gastrointestinal tract is prone to perforation in events of distension, ischemia, volvulus, or a disease setting that may render the circumferential layers susceptible for thinning out. The cause and effect relationship is constant and sporadic events of perforation probe the investigator to look deeper into underlying reason. These causes include previous enterocolitis, Hirschsprung disease, swallowed foreign bodies, focal intestinal perforations, and distension tears due to atresia, especially in preterm infants.¹ Most cases are clearly related to necrotizing enterocolitis, but some have no symptoms or clinical findings. Interestingly, the regions where most idiopathic perforations are located are the flexures.¹ Biopsy examinations are important in these patients because Hirschsprung's disease may be one of the causes of perforation.

The case here needs mention as there was no disease process or trauma that may have attributed to development of pneumoperitoneum. The diarrhea was the effect not the cause. It wasn't severe or copious and was indirect depiction of peritoneal irritability with mild collection progressing gradually. The child developed a concerning condition without any background setting of disease or trauma that is rather rare in the given age group.

CASE

A 2-years-old female patient presented with 4 days' history of diarrhea, low grade fever, non-bilious vomiting. According

to mother patient had no previously known condition. The patient was born at term, and was doing fine till she developed episodes of diarrhea which was mild in form of loose stool, subsequently she experienced vomiting, 2-3 in frequency and non-bilious. In mean time she developed a continuous low grade fever and she had been taking medication for fever when she presented in hospital. On day of presentation to hospital child had developed abdominal pain and fullness without obvious distension. On arrival child was hemodynamically stable but in agony with grunting. There was moderate dehydration owing to diarrhea and decreased intake. Abdominal review demonstrated guarding and tenderness in all quadrants, abdomen was minimally distended but no evidence of any bruising or collection. Inguinal regions were also normal.

On arrival her blood labs revealed normal hemoglobin of 13.3 g/dl. Platelets were normal and WBC was minimally deranged to 3.65. Serum biochemistry and liver function tests were normal with creatinine deranged due to dehydration. Urgent X-ray sought that revealed pneumoperitoneum on lateral X-ray that was supplemented with computed tomography (CT) scan. Alongside patient was resuscitated with intravenous fluids with initial bolus and later on with maintenance and antibiotics. CT scan findings revealed pneumoperitoneum with minimal collection around ascending colon and diffuse small bowel thickening. Urgent laparotomy planned and parents briefed about the possible outcomes of celiotomy; stoma formation, resection anastomosis, long term morbidity etc.



Blood was arranged and patient was shifted to operating theater. On review whole of the gut from duodenojejunal junction till rectum was intact and normal with no erythema except for a solitary punched out perforation encountered in ascending colon adjacent to hepatic flexure. The diffuse bowel thickening mentioned in CT report couldn't be appreciated and correlated per operatively. No evidence of any local chronic disease, no enlarged lymph nodes. The margins were refreshed and primarily repaired in two layers. Thorough lavage was done to attenuate contamination. Patient was kept in PICU and antibiotics were upgraded postoperatively from ceftriaxone and metronidazole to vancomycin, ceftriaxone and metronidazole. She was rendered non per oral till bowel movements established on bowel auscultation. The culture report came back as positive for pseudomonas for which antibiotics were changed to meropenem that showed promising sensitivity. On regular course of recovery patient was discharged in 3 days with no active complaint. The patient was examined in outpatient clinics and on follow-up evaluation; the patients had a healthy healing wound and soft non tender abdomen.

Pathology examination revealed margins resected from the perforation and the examined sections reveal fibromuscular tissue with heavy infiltration by inflammatory cells rich in neutrophils. The lining mucosa is totally ulcerated with neutrophils and fibrin deposition.

DISCUSSION

Neonates are more prone to perforation due to developmental conditions that may lead to aberrant dynamics of gut transit, commonly encountered neonatal conditions are Hirschsprung's disease and necrotizing enterocolitis.² Older children also pose a risk but are less frequent as compared to aforementioned.² Children without preceding history of any disease or trauma are rarely encountered with perforation more so in colonic region that is more capacious and adaptable with slow transit and formed and lessened bulk as compared to small bowel. In cohort studies, it has been determined that the problem causes perforation regardless of the part of the gastrointestinal tract for three basic reasons: birth under 1,000 grams, premature birth, and use of mechanical ventilation.¹ Additionally, Hirschsprung disease was detected in all patients with colon perforation.

The colonic perforation may be of two types; stercoral being strongly associated with constipation, is even rarer in juvenile as compared to idiopathic type.³ The classification doesn't give much insight into disease process that remains a mystery. No pathophysiology can be associated with idiopathic perforation though some studies have showed an indirect link of over usage of NSAIDs in such patients because of fever or pain. The excess usage of NSAIDs has a more pronounced link with elderly patients having diverticula but the association has been suspected in Pediatric age group as well.⁴ The enteric coated or slow release forms are riskier as they bypass stomach increasing contact time with colon but their use isn't in pediatric age group. Other reason could be frequent dosage, and the inhibition of prostaglandin mediators leading to susceptibility of gut mucosa.⁵ Having said that it remains one of the postulations in idiopathic colonic perforations and not taken as concrete evidence.

Histopathological assessment of such patients is more significantly suggesting acute inflammation and neutrophilic

infiltration instead of ischemic changes or chronic conditions. Similar condition was found in our patient with marked neutrophilic infiltration.

The delay may lead to gross fecal contamination and sepsis that can increase morbidity and mortality many folds. The key is to be vigilant in this subset of patients where there is no obvious differential diagnosis presenting with minimal abdominal findings. As it may be earlier presentation in emergency due to pain and not having developed full blown abdominal distention characteristic of perforation. The learning curve for minimally invasive exploration is high and with gross contamination and hemodynamic instability seen in such patients it remains an inferior approach though a subset of surgeons belong to other school of thought preferring minimally invasive surgery.⁶

The rarity of this condition mandates mention so correlation with different centers and experiences may yield a better understanding of the condition and contribute to evidence based practice.

In our case, the reason of intestinal perforation is unknown. One known cause that could support the reason of perforation could be severe diarrhea or obstruction leading to substantial gaseous distension impinging on anchoring sides of colon. Studies about the effect of *Pseudomonas* on bowel perforations especially for necrotizing enterocolitis have not been proved yet.⁷ Another disease condition common to children in this age group are inflammatory bowel disease but here there wasn't any evidence of any complaint prior to this and that too if present has other sequel like stricture, fistula, abscess or granuloma formation instead of perforation except in instances of toxic megacolon that may sometimes accompany advanced stages of ulcerative colitis. A Meckel's diverticulum may also lead to kink and obstruction but usually with a noticeably sized Meckel's diverticula there is association of ectopic mucosa with history of occult or gross hematochezia. In this case the cause remained truly idiopathic.

CONCLUSION

Intestinal perforations in infancy mainly occur on occasions that the patients had suffered previously. Some of perforations were predisposed by reasons like premature or very low birth weight. Idiopathic spontaneous perforations must not be underestimated. Diagnosis of a spontaneous perforation needs careful examination and evaluation protocols.

ETHICAL DECLARATIONS

Informed Consent

Written informed consent was obtained from the patient's parents.

Referee Evaluation Process

Externally peer-reviewed.

Conflict of Interest Statement

The authors have no conflicts of interest to declare.

Financial Disclosure

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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.

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