e-ISSN:3023-7289

SoC
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Volume: 2 Issue: 3 Year: 2025

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doi: 10.51271/SOC-0041

Evaluation of our post-appendectomy management protocol

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Received: 31/12/2024 ◆ **Accepted**: 18/03/2025 ◆ **Published**: 01/07/2025

Cite this article: İbrahim AA, Rayes TE, Abbas TO, Al Romaihi SA, Ali MA. Evaluation of our post-appendectomy management protocol. *Surg Child*. 2025;2(3):82-86.

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ABSTRACT

Aims: Many post-appendectomy treatment protocols have been created and modified, but none were aspiring to be the best in pediatric surgery. In 2008 Shawn et al, standardized the definition of perforated appendix to be only when you see a hole in the appendix or a fecolith in the peritoneal cavity1; We continued the definition and set a new treatment protocol accordingly.

Methods: It is a retrospective study for the age group 0 to 14 years old. The Treatment protocol was created and applied in 2012. We have created two groups (100 patients in each group). First group from 2010 - 2011 and second group from 2013-2014. The treatment in the first group is clinical and empirical. In the second group the treatment depended on a designed criteria matching our protocol.

Results: There was no statistical significance regarding the post-operative collection (5 patients in the 1st group and 4 patients in the 2nd group), mean length of hospital stays (4.57 in the 1st group and 5.30 in the 2nd group), emergency visits (5 visits in each group), and finally hospital re-admission (1 in the 1st group and none in the 2nd group).

Conclusion: There was a clinical significance but no statistical significance between the two groups. Only 4 patients in the 2nd group and 5 patients in the 1st group developed post-appendectomy collection. The re-admissions in the 2nd group were not related to surgery. The antibiotic cost is almost the same, although the number of perforated appendices is higher in the second group. Furthermore, we recommend our protocol for safe patient discharge, especially after complicated appendicitis operations.

Keywords: Appendicitis, collection, antibiotics

INTRODUCTION

Acute appendicitis being the most common emergency condition in pediatric surgery, there appears to have no consistency on the management protocol.[1] Numerous researchers sought to find the most effective outcome with the least morbidity rate using various diagnostic algorithms and different antibiotics.[2,3] Many protocols had been created and modified, but none were aspiring to be the best in pediatric surgery. In 2008 Shawn et al.[1], standardized the definition of perforated appendix to be only when you see a hole in the appendix or a fecolith in the peritoneal cavity. Their outcome was improved after the application of the definition of perforation but not to a great extent.[1] We decided to extend the definition and set a treatment protocol accordingly. Various surgeons decided on the duration of post-appendectomy antibiotics based purely on the clinical findings, in turn they stopped the antibiotics and discharged patients home when the child was afebrile, able to tolerate feeding and pass a bowel motion. Their decisions led to the flaring up of post-appendectomy infections due to inadequate treatment in some cases.[4] Because there was no agreement about the appendicitis treatment, we tried to hold onto these cases early on before they were discharged to avoid potential future complications. In 2012 Holcomb and Shawn[2] studied optimal antibiotic regimen and the length of antibiotic therapy, in 2015 Farach et al.[5] published a study in which they found that operative findings are a better predictor of resource utilization in pediatric appendicitis, and this aligns with our protocol which employed more precautions in order not to miss early post-appendectomy abscess formation.

Objective

We analyzed the management outcome in two post-appendectomy groups retrospectively before and after the implementation of our treatment protocol according to the new intraoperative appendicitis definitions, regarding primary objectives which are length of hospital stay (LHS,

Days) and the incidence of post-appendectomy abscess formation. Additionally, we aimed to identify other variables like the incidence of wound infections, re-admissions within the 1st month after the initial appendectomy and the in-hospital intravenous antibiotic charges (US$).

METHODS

The study was conducted with the permission of the Medical Research Centre HMC Ethics Committee (Date: 10.08.2021, Decision No: MRC-01-21-446). All procedures were carried out in accordance with the ethical rules and the principles of the Declaration of Helsinki.

We used a retrospective cohort study; our age group is 0 to 14 years old. All cases were done by laparoscopy, and the converted cases are not included in the study. C-reactive protein (CRP) and CBC are being done as a routine blood tests for all the cases with abdominal pain in the emergency department as a baseline before admission to the hospital. We studied the post appendectomy treatment before and after the application of our designed treatment protocol which was applied in 2012. We created 2 groups, one before and one after 2012, we avoided the year 2012 because of the overlap period. The sample size was calculated with the help of our medical research center (MRC).[6] The sample size collection identified the required number would be 89 patients to identify statistical significance. Nevertheless, we collected 100 patients in each group. Our MRC statistician based the sample size calculation on Shawn et al.[1] study. The sample size was determined on the primary outcome variable length of stay (days) reported in the literature: with effect size 1.2, standard deviation of the change in the outcome 3.5, statistical power 90% and level of significance 5%, the required sample size would be n=89 cases. However, to account for multiple secondary outcome measures such as dropouts and non-responses, it was good to increase an additional 30% in the calculated sample size i.e., a total of 200 participants divided equally into two groups will be included in this study. The following sample size equation was used[6]:

The standard normal deviate for α=Zα=1.960,
The standard normal deviate for β=Zβ=1.282,
A=1.000,
B=(Zα+Zβ)2=10.507,
C=(E/S(Δ))2=0.118,
AB/C=89.39,
Group size N: 89,

In the first group, post-appendectomy treatment including antibiotics duration was empirical and it differed between each surgeon. It was dependent on the clinical picture (presence of fever, tolerance of feeding, abdominal pain, and presence of diarrhea).

The protocol of group 2 is guided by the preoperative white cell count (WBC), CRP, and intraoperative classification (**Table 1**). Our antibiotic regimen adheres to our local Institution of Infection Control Guideline standards. The first-line antibiotics are ceftriaxone and metronidazole, the duration of which was established by our intraoperative new classification.

Table 1. Intra-operative classification of the appendix

Appendix type	Description
Normal	No signs of inflammation
Inflamed	Swollen, congested
Severely inflamed	Inflamed with pyogenic membranes or pus in the peritoneal cavity (localized or generalized)
Perforated	When there is a hole in the appendix or a fecolith in the peritoneal cavity

In the second group (**Figure 1**) our new protocol strictly classified appendicitis into 3 categories (**Table 1**); Normal and inflamed, severely inflamed and perforated according to the intraoperative findings.

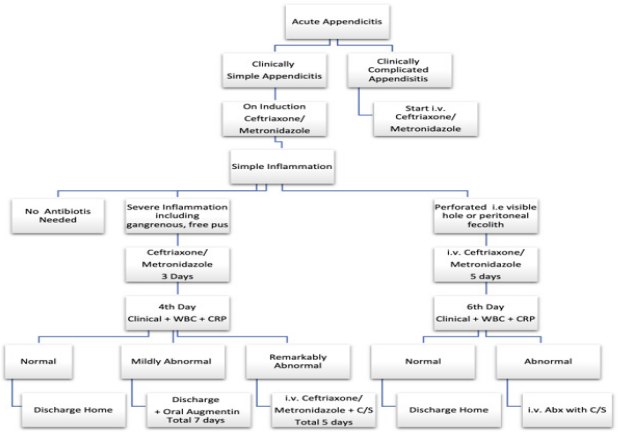

Figure 1. The post-appendectomy antibiotic treatment based on intraoperative classification with the aid of CRP and WBCs
i.v.: Intravenous, WBC: White blood cell, CRP: C-reactive protein, C/S: Culture and sensitivity, Abx: Antibiotics

Normal appendix meant the peritoneal cavity is clean and no local signs of inflammation in the appendix. Just inflamed appendix meant the peritoneal cavity is clean, and the inflammation was confined to the appendix only (**Figure 2A**); these patients received a single dose of antibiotics upon induction of anesthesia. Severely inflamed meant inflamed appendix with the presence of pyogenic membranes (**Figure 2C**) or pus (**Figure 2B**); these patients received 3 days of antibiotics starting upon induction of anesthesia, and before discharge we repeated the CRP and WBCs then we followed up according to the CRP and WBCs results (**Figure 1**). The perforated appendix meant inflamed appendix with the presence of a hole in the appendix (**Figure 2E**) or a fecolith (**Figure 2D**) in the peritoneal cavity; these patients received 5 days of antibiotics starting upon induction of anesthesia, before discharge (Morning of the 6th post-operative day) we repeated the CRP and WBCs then we followed according to the CRP and WBCs results (**Figure 1**).

RESULTS

A total of 200 patients were collected, 100 patients before applying the treatment protocol (group 1) and 100 patients after employing the protocol (group 2) (**Table 2**). The number of males was 140 (66 in the first group and 74 in the second group, total being 70%) while the females were 60 (34 in the first group and 26 in the second group, total being 30%).

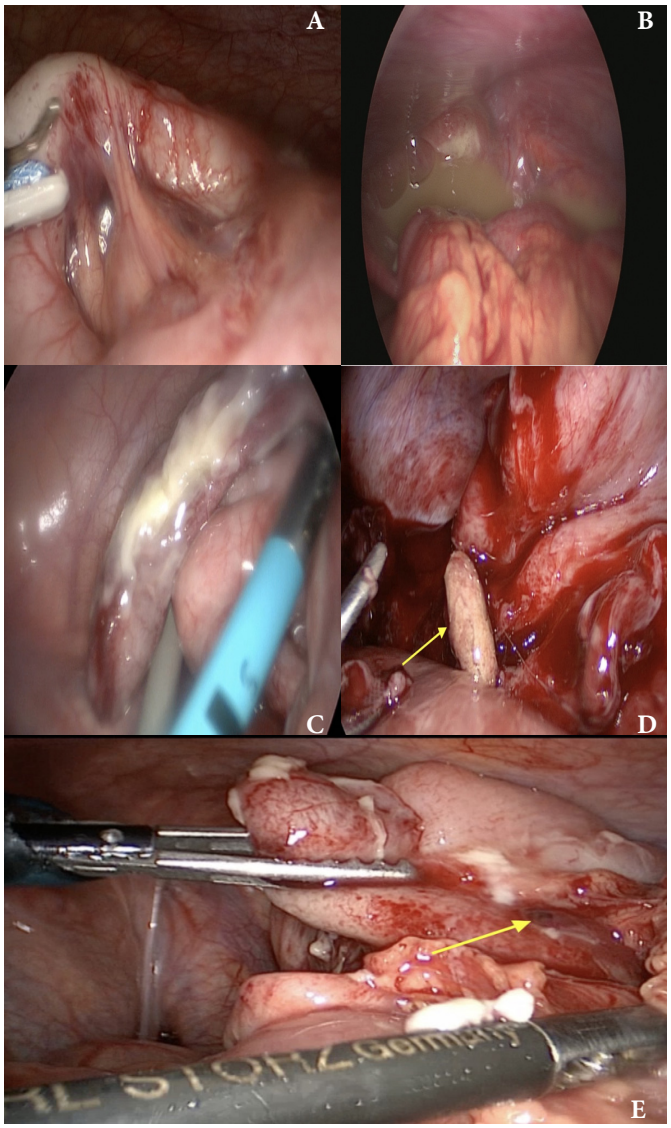

Figure 2. Intraoperative classification of the appendix. **2A.** Inflamed appendix, **2B.** Pus in the pelvis, **2C.** Severely inflamed appendix with pyogenic membranes, **2D.** Fecolith in the peritoneal cavity, **2E.** Perforation in the appendix (arrowed)

Table 2. The frequency of different variables

Variable	First group	Second group	p value
Sex: Male	66	74	0.217
Female	34	26	0.280
Age (mean)	8.99	8.95	0.918
Length of hospital stay (mean)	4.57	5.30	0.050
LOS (median)	4.0	5.0	0.015
Intraabdominal collection	5	4	0.733
Wound infection	1	2	1.000
Re-admission (1st month)	1	0	1.000
Emergency visits	5	4	0.733
Antibiotic charges (US$, mean)	180	228	0.034
Antibiotic charges (US$, median)	144	240	0.001
Histopathology: Normal	8	1	0.000
Inflamed	27	51	0.019
Suppurative	60	36	0.000
Perforated	5	12	0.000

US$: United States Dollar

The total number of post operative intra-abdominal collections (Abscess formation) is 9, where there were 5 in the first group and 4 in the second group (Table 2). There was no significant statistical difference between group 1 and group 2 as regard the risk of developing a post appendectomy intraabdominal collection (p=0.733) OR=0.792 (95% CI 0.206 to 3.039). Regarding the LOS, group 1 had shorter mean length of hospital stay than group 2 (4.57 versus 5.3 days). The one-sided t-test showed a marginally significant statistical difference between the two groups-0.73 days (95% CI-1.6 to 0.14) (p=0.05). Regarding the antibiotic charge, group 1had a lower mean antibiotic charge compared to the group 2 (180.96 versus 228.06 USD). The one-sided t-test showed a marginally significant statistical difference between the two groups -47.10 USD (95%CI-97.74 to3.54). Finally, hospital re-admission was 1 in the 1st group and none in the 2nd group (Table 3).

Table 3. Statistical analysis for the LHS and antibiotic charges

		Age (years)	LHS (days)	Abs (charge US$)
Age (years)	Pearson correlation	1	-.200**	-.218**
	Sig. (2-tailed)		0.005	0.002
	n	200	200	200
Length of hospital stay (days)	Pearson correlation	-.200**	1	.974**
	Sig. (2-tailed)	0.005		0.000
	n	200	200	200
Abs (charge US$)	Pearson correlation	-.218**	.974**	1
	Sig. (2-tailed)	0.002	0.000	
	n	200	200	200

** Correlation is significant at the 0.01 level (2-tailed). US$: United States Dollar, Abs: Antibiotics, LHS: Length of hospital stay

DISCUSSION

Although acute appendicitis is the most frequent emergency condition in pediatric surgery, unfortunately there is no standard protocol for its management. Surgeons around the world classify patients with complicated appendicitis using a variety of criteria and categories. The absence of a standardized definition of perforated appendicitis is responsible for the large variation in reported perforation rates and discrepancies in postoperative abscess rates among individuals with perforated appendicitis. Surgeons use terms such as purulent, perforated, gangrenous, necrotic, ruptured, contained-perforated, and degrees of peritonitis for which there is no evidence, additionally, they represent the same entity. Recent research on this subject revealed a dramatic spectrum for perforation, ranging from 20% to 76%.[7] Furthermore, several surgeons make these diagnoses based on their general clinical impressions, as opposed to the objective criteria. American Pediatric Surgical Association (APSA) members were surveyed in 2000, and the majority reported basing their practice approaches on personal preferences.[8]

Exercising various diagnostic algorithms and antibiotics[2], numerous researchers attempted to discover the optimal

method for achieving a favorable outcome with minimal morbidity. Numerous protocols had been developed and modified, but none stood out as the most effective in pediatric surgery. In 2008, Shawn et al.[1] standardized the definition of a perforated appendix to include only the presence of an appendix perforation or a peritoneal fecolith. Appropriately, we decided to expand the definition and establish a new treatment protocol. Numerous surgeons determine the duration of post-appendectomy antibiotics based solely on clinical findings, so they were discontinuing antibiotics and releasing patients when they were afebrile, and they were able to consume and have bowel motion. In some cases, this led to the worsening of post-appendectomy infections caused by such inadequate treatment. As there was no consensus regarding the treatment of appendicitis, we attempted to identify these cases early to avoid potential complications. In 2012, Holcomb and Shawn[2] investigated the optimal antibiotic regimen and duration of treatment. Operative findings are a better predictor of resource utilization in pediatric appendicitis, according to a 2015 study by Farach et al.[5] This finding aligns with our protocol, which incorporated more precautions so as not to neglect early post-appendectomy abscess formation. Prior to 2012, Nadler et al.[9] treated perforated appendicitis with a triple antibiotic regimen consisting of ceftriaxone, gentamicin, and metronidazole (triple antibiotics course). However, this regimen demands a complicated dosing schedule and blood tests to monitor nephrotoxic gentamicin levels. Recent research indicates that monotherapy with piperacillin/tazobactam for intra-abdominal infections is as effective as traditional triple therapy.[9] However, Individual doses of piperacillin/tazobactam are costly, and patients still require three to four doses per day. In our institution we spare this regimen for the resistant organisms or when it comes to the culture and sensitivity studies for the peritoneal fluid aspirated during the appendectomy.

During our study, although there was no significant statistical difference between the 2 groups there was an important clinical difference which reflected on the patient outcome.

In the first group, 5 cases had post operative abscess formation, 4 of them after complicated appendicitis cases (2 suppurative & 2 perforated) and 1 after simple inflamed appendicitis, where one which was discharged and re-admitted again.

In the second group, 4 cases had post operative abscess formation, only 1 after suppurative appendicitis and 3 cases after simple inflamed appendicitis. All of them were picked up before discharge from the hospital.

In the second group, none of them got re-admitted due to surgical complications; 2 patients were re-admitted in this group, 1 due to pneumonia with pleural effusions and the other with non-specific abdominal pain managed conservatively in general pediatrics after clearance from pediatric surgery. None of the postoperative collections in the second group was discharged home, all were picked up before being discharged and were treated early. Although The percentage of patients with perforated appendix in the second group (12%) is greater than those in the first group (5%), incidence of complications, re-admission and antibiotic cost is near in both groups which gave advantage to the second group.

CONCLUSION

We recommend the application of our management protocol as it has a clinical significance; less postoperative complications guiding the antibiotic duration and we can pick up of post operative collections before sending the patients to their homes. However, we recommend future prospective larger studies or randomized trials with the use of more sensitive inflammatory markers (like procalcitonin) to enhance the study's impact.

ETHICAL DECLARATIONS

Ethics Committee Approval

The study was conducted with the permission of the Medical Research Centre HMC Ethics Committee (Date: 10.08.2021, Decision No: MRC-01-21-446).

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.

Acknowledgment

We appreciate the significant effort and help done by Dr. Prim Chandra, for doing the sample size and statistical analysis for our study as well as Mr. Eyad Abdelhamid for the professional English-editing done as required by the journal editors.

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MHA MEDIHEALTH ACADEMY

Surgery on Children

Original Article

doi: 10.51271/SOC-0042

Surgical and postoperative complications of circumcision performed by unqualified individuals: the need for regulatory oversight

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Received: 13/02/2025 ◆ **Accepted**: 22/03/2025 ◆ **Published**: 01/07/2025

Cite this article: Kuas N, Tekkanat B, Turan H, Göllü Bahadır G. Surgical and postoperative complications of circumcision performed by unqualified individuals: the need for regulatory oversight. *Surg Child*. 2025;2(3):87-91.

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ABSTRACT

Aims: Circumcision is a common surgical procedure performed for many reasons. When conducted by non-medical personnel, the risk of complications increases significantly. This retrospective study analyzes pediatric cases requiring emergency care due to complications from non-medical circumcisions.

Methods: Over three months, data from ten pediatric patients who underwent circumcision by non-medical individuals and later required emergency department visits were analyzed. Age, admission time, symptoms, and clinical findings were recorded.

Results: Patients, aged 3 months to 13 years, presented within 40 minutes to three weeks post-circumcision. Common complications included bleeding, hematoma, improper incision, and wound dehiscence. Two patients required additional surgery. Despite legal restrictions, such procedures persist in multiple regions.

Conclusion: Non-medical circumcision increases complication risks, leading to physical and psychological distress. Strict regulation enforcement and public awareness are essential to ensuring safe, medically supervised procedures.

Keywords: Circumcision, complications, pediatric urology

INTRODUCTION

Circumcision is the surgical removal of the foreskin that covers the glans penis (prepuce) to expose the penil tip.[1] With a history of approximately 5000 years, it is one of the oldest and most commonly performed surgical procedures.[2]

Circumcision is a widely performed surgical procedure with various medical, cultural, and religious indications.[3] While generally considered safe, it is not without risks, as complications such as bleeding, infection, and inadequate or excessive tissue removal may occur.[4,5] The frequency and severity of these complications depend on multiple factors, including the experience of the practitioner and the conditions under which the procedure is performed.[6]

This study aims to retrospectively evaluate ten cases of children circumcised by non-medical practitioners who subsequently presented to the emergency department and were referred to the pediatric urology department.

METHODS

This study was conducted with the permission of the Non-interventional Clinical Researches Ethics Committee of Van Training and Research Hospital (Date: 29.11.2024, Decision No: GOKAEK-2024-01-10). All procedures were carried out in accordance with the ethical rules and the principles of the Declaration of Helsinki.

This study was designed as a retrospective analysis conducted in a tertiary training and research hospital. Patients included in the study were children who had undergone circumcision performed by unqualified individuals and subsequently presented to the emergency department due to post-circumcision complications between June and September 2023. These patients were later referred to the pediatric urology department for further evaluation and management. All patients consulted were reviewed. Data collected included patient age, time of hospital admission, presenting complaints, and clinical examination findings.

Unqualified individuals were defined as those who lacked formal medical education, did not possess any official healthcare certification, and were not trained in sterile surgical techniques. In the present study, patients were evaluated based on this definition, and only those circumcised by unqualified individuals were included. The classification of the practitioner as qualified or unqualified was based on the statements provided by the patients' families at the time of admission.

RESULTS

A total of 10 patients who presented to the emergency department following circumcision procedures performed by unqualified individuals and were subsequently referred to the pediatric urology department were included in the study.

The patients were aged between 3 months and 13 years, with the time to hospital admission ranging from 40 minutes to three weeks (**Figure 1, Table**). The most common presenting complaints included abnormal scabbing on the glans (20%), bleeding (20%), hematoma (10%), improper circumcision incisions and wound dehiscence (40%) (**Figure 2, 3**).

Figure 1. 13 years old, 9th day postoperatively, no additional complaints after wound care and regular dressing

Two patients (20%) required additional surgical intervention. An 8-year-old patient with uncontrolled bleeding despite compression dressings was taken to the operating room, where bleeding sites were cauterized and visible blood vessels were individually ligated (**Figure 4**). The patient was discharged on the third postoperative day without complications. Another

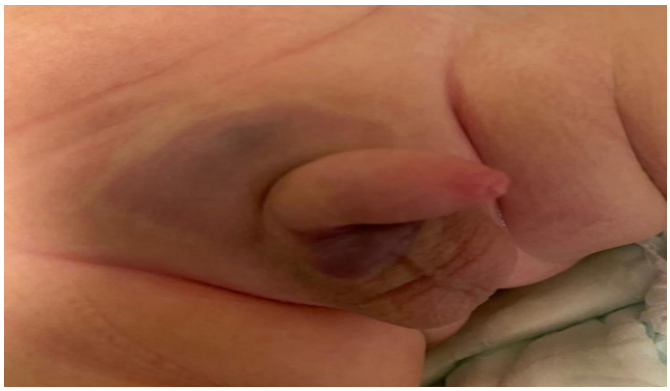

Figure 2. The 3-month-old baby, after local anesthesia, the procedure could not be continued. No information could be obtained about the drug and its dosage. He was evaluated by the pediatrician for poisoning. The patient, whose glans blood flow was normal and urine output was easy, was recommended circumcision under elective conditions.

Figure 3. 5 years old, 10th postoperative day, no additional complaints after local care

8-year-old patient (**Figure 5**), who had been circumcised seven days earlier and developed wound dehiscence following balanitis treatment, underwent surgical intervention where the wound edges were trimmed and re-sutured. No further complications were observed during follow-up.

Patients from three different provinces were identified; however, no information could be obtained from the parents regarding the identity of the practitioner or the location of the procedure. Due to the high incidence of complications in a short period, discussions with local authorities were initiated.

Table. Patients, postoperative time, complication, treatment

	Year	Postoperative day	Complication	Treatment
1	13 y	9th day	Wound dehissance and infection	No additional complaints after wound care and regular dressing
2	6 y	10th day	Wound dehissance and infection	No additional complaints after wound care and regular dressing
3	5 y	7th day	Wound dehissance and infection	No additional complaints after wound care and regular dressing
4	9 y	7th day	Crusting on the glans	All complaints regressed after medical therapy
5	3 m	20 minutes	Ecchymosis at the root of the penis	Recommended circumcision under elective conditions
6	6 y	10th day	Crusting on the glans	Antibiotic local ointment treatment
7	8 y	7th day	Infection	No additional complaints after wound care and regular dressing
8	5 y	10th day	Crusting on the glans	No additional complaints after local care
9	8 y	1st day	Bleeding	Bleeding foci were cauterized. Visible vascular structures were tied one by one
10	8 y	7th day	Wound dehissance, infection and bleeding	Wound edges were trimmed and re-sutured

Figure 4. 8 years old patient, on the first postoperative day, whose bleeding did not stop after pressure dressing and local care, was taken to the operating table. The suture line was opened. Bleeding foci were cauterized. Visible vascular structures were tied one by one. He was discharged three days later.

Figure 5. 8 years old, 7th postoperative day; after balanitis treatment, wound edges were trimmed and re-sutured

DISCUSSION

Although some anthropologists estimate that circumcision dates back to as early as 10.000 BCE, the earliest historical evidence of circumcision comes from the Ankh-Mahor inscriptions in Egypt, dating to approximately 4000 BCE.[7] Modern surgical principles were first defined in 1903 by Sir Frederick Treves[8], and since then, various techniques such as the guillotine method, dorsal slit and excision, sleeve technique, Sheldon method, and various circumcision clamps have been developed.[9]

According to the World Health Organization, approximately 33% of men over the age of 15 worldwide are circumcised.[5] In Turkey, the median age for circumcision is estimated to be six years, with 15% of circumcisions performed in children under one year of age.[5] According to a study by Özdemir et al.[10] 85% of circumcisions are performed by traditional circumcisers, 10% by non-medical healthcare personnel, and only 5% by surgeons. Despite being one of the most frequently performed surgical procedures in Turkiye, the long-standing exclusion of circumcision from social security coverage has significantly contributed to these figures.[11]

The complication rate for circumcision reported in the literature ranges from 0.2% to 2%, but in inappropriate conditions, this rate can increase to 8–17%.[4] In mass circumcision events, the complication rate can reach up to 95%, leading to severe complications that are difficult to manage.[5]

Have documented an incidence of bleeding and infection in medically performed circumcisions, supporting the idea that these complications can occur even under the most favorable conditions.[4,5] However, studies such as Ceylan et al.[6] have also shown that complication rates are significantly higher in procedures performed by unqualified individuals. This comparison highlights the need to ensure that circumcisions are performed in controlled medical environments to minimize adverse outcomes. By aligning our data with the existing literature, our study contributes to the ongoing debate about best practices and safety precautions in circumcision procedures.

Complications of circumcision can arise from anesthesia and/or the surgical procedure itself. Early complications include excessive shortening of the skin or mucosa, insufficient circumcision, injury or excision of the glans, bleeding, hematoma, infection, suture reactions, pain, and psychosocial issues. Late complications include castration anxiety, urethral fistula, meatal stenosis, skin bridges, and dissatisfaction with the outcome.[4] Local anesthesia-related complications such as methemoglobinemia, hematoma, convulsions, penile necrosis, cardiac arrhythmia, and arrest may also occur.[12] General anesthesia-related complications include hypoxia, apnea, laryngeal spasm, aspiration pneumonia, cardiac arrest, convulsions, and malignant hyperthermia.[13]

The most common complications are bleeding and infection, while the most severe is glans amputation.[5] These were the most common complications in this study. The mortality risk associated with circumcision is reported as 2 per million in the literature.[14]

In countries where circumcision is routinely performed, circumcision is frequently performed under inappropriate conditions due to both traditional habits and the inability of health institutions to cope with this workload. A study from Yemen reported distal penile amputation in a 5-month-old patient and death in a 20-day-old patient.[15] Compared to the complications in this study, these two cases are much more catastrophic. However, patients presenting to health institutions may be just the tip of the iceberg.

A study conducted in the United States reported that 7.4% of all pediatric urology consultations at the study hospital were related to neonatal circumcision, and approximately 4.7% of all surgeries performed in the pediatric surgery department were for circumcision complication repairs.[16]

Proper preoperative preparation, including a detailed physical examination, medical history, and laboratory tests, is essential.[11] Thorough preparation can help reduce complication rates and prevent the oversight of coexisting anomalies. If an aseptic environment is not provided, which is not possible in home conditions, blood-borne diseases can be transmitted to both the operator and the patient.

Circumcision in Turkiye was included in the social security system's coverage in 2007. Its prior exclusion led to many

circumcisions being performed by unqualified individuals, and in some regions, non-medical circumcision continues under inadequate conditions and has become normalized. Law enforcement has been insufficient or indifferent in addressing this issue.

In 2007, the American Academy of Pediatrics stated that the health benefits of neonatal male circumcision outweigh the risks. It stated that families should be given a choice about neonatal male circumcision and that the procedure should be reimbursed by third parties. It was written that circumcisors should have adequate training and that both sterile techniques and effective pain management should be used. It was also noted in this study that untrained providers performing circumcisions had more complications than well-trained providers performing the procedure.[17]

A law was passed in Turkiye in 2014 banning circumcision by non-medical practitioners. However, this law allows all physicians, regardless of their surgical training or circumcision expertise, to perform circumcision.[18] We believe that a certification program for physicians who do not have detailed training, especially those who are not trained in circumcision, on issues such as sterilization conditions, suture techniques, pain management, etc., will reduce complications. Families should be repeatedly told that circumcision should not be performed by people who have not received training on this subject or under inappropriate conditions.

Limitations

The limitations of this study are; first of all, the inability to reach the people who performed the circumcision. The way the circumcision was performed, what was done in the preoperative evaluation, the quality of the materials used, how asepsis conditions were provided and how postoperative care was provided could not be clarified. In addition, the patients identified were only those who were brought to the health institution by their families and then consulted with the relevant department. Patients who were not brought to the health institution could not be identified.

To ensure competency in performing circumcision, each country's health authority should establish specific training requirements, including the type of courses required, the number of supervised procedures, and the minimum number of circumcisions needed for certification. Otherwise, what is considered a "simple" procedure can turn into a "major nightmare".[16]

CONCLUSION

To prevent the financial and emotional burden caused by complications arising from procedures performed by unqualified individuals in inappropriate conditions, circumcision should be performed by specialized surgeons under proper medical conditions. As physicians, we are responsible not only for treating patients but also for providing preventive healthcare. Raising public awareness can help prevent irreversible complications by ensuring that circumcision is performed by qualified professionals in appropriate conditions.

ETHICAL DECLARATIONS

Ethics Committee Approval

This study was conducted with the permission of the Non-interventional Clinical Researches Ethics Committee of Van Training and Research Hospital (Date: 29.11.2024, Decision No: GOKAEK-2024-01-10).

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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MHA MEDIHEALTH ACADEMY

Surgery on Children

Original Article

doi: 10.51271/SOC-0043

The unspoken burden: emotional resilience and healing among pediatric urologists in high-stress clinical environments

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Received: 11/03/2025 ◆ **Accepted**: 06/04/2025 ◆ **Published**: 01/07/2025

Cite this article: Viswanathan V. The unspoken burden: emotional resilience and healing among pediatric urologists in high-stress clinical environments. *Surg Child.* 2025;2(3):92-94.

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ABSTRACT

Aims: Pediatric urologists routinely face emotionally charged clinical scenarios, including life-altering diagnoses, patient mortality, and ethical dilemmas. These experiences often lead to unresolved emotional residue, which can manifest as burnout, compassion fatigue, or somatic symptoms. This qualitative study explores how pediatric urologists process defining moments of loss and moral distress, and evaluates mindfulness-based interventions to mitigate their psychological burden.

Methods: Participants filled out a questionnaire attached in Appendix A, which was anonymized and then reviewed to get results. The study included 24 pediatric urologists across tertiary care centers in India. Narratives were analyzed using thematic coding. A pilot mindfulness intervention (5-minute diaphragmatic breathing) was trialed with 12 participants.

Results: Four key themes emerged: (1) anchoring trauma (76% reported recurrent intrusive thoughts about patient deaths), (2) emotional suppression (89% normalized compartmentalization as a coping mechanism), (3) somatic manifestations (63% linked unresolved grief to chronic insomnia or hypertension), and (4) healing through introspection (mindfulness practitioners showed a 40% reduction in burnout scores).

Conclusion: Unaddressed emotional trauma in pediatric urology correlates with diminished professional fulfillment and physical health. Institutional support systems, including mindfulness training, may foster resilience.

Keywords: Pediatric urology, emotional resilience, mindfulness, moral distress, physician burnout

INTRODUCTION

Pediatric urology is uniquely fraught with high-stakes decisions—repairing congenital anomalies, navigating parental despair, or withdrawing care in futile cases.[1] Such moments imprint deeply, often resurfacing as "emotional ghosts" that haunt clinicians years later.[2] While technical competence is prioritized in surgical training, emotional resilience remains undervalued, particularly in resource-limited settings like India, where one pediatric urologist may oversee 500+ annual cases amidst systemic overcrowding.[3]

This study examines how unresolved grief and self-doubt manifest among pediatric urologists and evaluates breath-focused mindfulness as a reparative tool.[4]

METHODS

Ethics

Since the research is conducted according to the survey results, no ethics committee report is required.

- Participants were assured of confidentiality (e.g., anonymized quotes, aggregated data).
- Answers were self-recorded with consent and transcribed verbatim.
- Follow-up questions were tailored to participant comfort level.

Participants: 24 pediatric urologists (14 male, 10 female; mean age 44 years) from 8 institutions.

Design: Mixed-Methods, Comprising

-Data collection: Participants filled out a questionnaire attached in Appendix A, which was anonymized and then reviewed to get results. The questionnaire explored pivotal clinical memories and coping mechanisms (**Table**).

-Mindfulness intervention: Participants practiced diaphragmatic breathing (5 minutes/day for 4 weeks).[4]

-Pre/post burnout was assessed via the Maslach inventory.[5]

Table. Appendix A: questionnaire

Participant demographic information
Age: _____
Gender: _____
Years in pediatric urology practice: _____
Approximate number of high-stakes cases (e.g., life-threatening anomalies, patient deaths) managed annually: _____
Institution type: ☐ Government ☐ Private ☐ Academic
Survey questions
Section 1: Defining moments in clinical practice
Describe a clinical case that has stayed with you long after it ended. What made it memorable?
Probe: How has this experience influenced your approach to subsequent cases?
Can you recall a moment where you questioned your decisions or abilities as a surgeon?
Probe: How did you reconcile these doubts?
Section 2: Emotional suppression and coping mechanisms
How do you typically manage emotions during or after a stressful procedure (e.g., patient death, parental conflict)?
Probe: Have you ever felt pressured to prioritize efficiency over emotional processing?
In your training, were you taught strategies to cope with grief or moral distress? If not, how did you learn to navigate these feelings?
Section 3: Somatic and psychological impact
Have you noticed physical symptoms (e.g., insomnia, headaches) linked to unresolved stress?
Probe: How do these symptoms affect your personal or professional life?
Do you believe your work has changed how you view vulnerability or emotional expression?
Section 4: Healing and institutional support
What practices or rituals, if any, help you process difficult experiences?
Probe: Have you ever sought formal mental health support? If not, what barriers exist?
How does your institution support clinicians' emotional well-being? What changes would you advocate for?
Section 5: Mindfulness intervention reflection
After practicing the 5-minute diaphragmatic breathing exercise:
Did you notice any shifts in your emotional or physical state during/after the practice?
Probe: Would you integrate such techniques into your routine? Why or why not?
Closing question
What advice would you give to a trainee struggling with the emotional weight of pediatric urology?

Statistical Analysis

Thematic analysis using NVivo 12.

RESULTS

Anchoring Trauma

76% (n=18) described vivid recollections of patient deaths in their questionnaire responses, such as a 7-year-old with bladder exstrophy succumbing to sepsis. One participant noted: "Her mother's wail… it's etched into my bones. I still hear it closing my eyes" (Participant 9).

Emotional Suppression

89% (n=21) reported suppressing distress to maintain clinical efficiency. As participant 14 stated in the anonymized questionnaire: "Grieve later, operate now—that's our mantra." This compartmentalization aligns with observed patterns in emergency medicine.[6]

Somatic Manifestations

63% (n=15) reported insomnia, migraines, or hypertension. Participant 3 linked his arrhythmia to "years of swallowing guilt over a preventable post-op complication," reflecting the physical toll documented in surgeon burnout research.[7]

Healing Through Introspection

Mindfulness practitioners (n=12) reported improved emotional regulation in their questionnaire responses. Participant 22 described "a visceral shift—like exhaling a weight I'd forgotten I carried," consistent with neurological changes observed in mindfulness research.[8]

DISCUSSION

The findings from the anonymized questionnaires align with global data on surgical burnout[7], but highlight cultural nuances in India, where stoicism is valorized and mental health stigmatized.[9] Suppression, while pragmatically necessary during emergencies, becomes maladaptive when habitualized, corroding empathy and clinical judgment.[6]

The pilot intervention's success mirrors evidence that breath-awareness reduces amygdala hyperactivity, offering a feasible antidote to chronic stress.[8]

Limitations

Limitations include sample size and self-reporting bias inherent in questionnaire-based studies.

CONCLUSION

Pediatric urologists shoulder a dual burden: healing children while grappling with invisible wounds. Institutions must integrate resilience training into surgical curricula and dismantle stigma around vulnerability. As participant 17 urged in their questionnaire response: "We suture bladders, but who sutures us?"

ETHICAL DECLARATIONS

Ethics Committee Approval

Since the research is conducted according to the survey results, no ethics committee report is required.

Informed Consent

All patients signed and free and informed consent form.

Referee Evaluation Process

Externally peer-reviewed.

Conflict of Interest Statement

The authors have no conflicts of interest to declare.

Financial Disclosure

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

Author Contributions

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

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MHA MEDIHEALTH ACADEMY

Surgery on Children

Original Article

doi:10.51271/SOC-0044

Comparison of transversus abdominus plane block with two different doses of levobupivacaine for postoperative pain management in pediatric patients

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Received: 18/04/2025 ◆ **Accepted**: 20/05/2025 ◆ **Published**: 01/07/2025

Cite this article: Uyar E, Çakar Turhan S, Özçelik M, Alkış N. Comparison of transversus abdominus plane block with two different doses of levobupivacaine for postoperative pain management in pediatric patients. *Surg Child*. 2025;2(3):95-100.

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ABSTRACT

Aims: Transversus abdominus plane (TAP) block is a simple, effective and reliable method in postoperative pain management following lower abdominal procedures. The aim of the current study is to investigate the effects of two different doses of Levobupivacaine in TAP block for postoperative management and to evaluate whether we can achieve effective pain management with lower dose than the recommended dose of Levobupivacaine.

Methods: Fifty patients aged between 2-12, undergoing unilateral inguinal surgery were included to the current randomized, controlled study. The patients were randomized to group C (control) (TAP block with Levobupivacaine 0.25%, 0.5 ml/kg) and group LD (low dose) (TAP block with Levobupivacaine 0.25%, 0.25 ml/kg). Following monitorization and achieving sufficient anaesthesia depth, ultrasound (USG) guided TAP block was performed to all patients. The pain intensity [visual analogue scale, observer pain scale and faces scale], recovery profile (Modified Aldrete Recovery Score) and patient satisfaction were evaluated postoperatively. Peripheral nerve block complications and side effects were recorded during postoperative period.

Results: No significant difference was found between groups demographic data ($p>0.05$). There was no significant difference between groups in terms of VAS, OPS, faces scale, patient satisfaction ($p>0.05$). The number of patients requiring rescue analgesic was 12 patients in control group and 14 in low dose group ($p>0.05$). No complication and side effects were observed.

Conclusion: TAP block with low dose Levobupivacaine may be successful to manage pain with less toxicity, and reduces complications due to local anesthetics.

Keywords: Levobupivacaine, TAP block, pediatric patients

INTRODUCTION

Postoperative pain in the pediatric age group may cause discomfort, hemodynamic status impairment, and organ dysfunction. Postoperative pain control is very important in order for children to have a more peaceful recovery after surgery with fewer complications. In addition, postoperative pain is one of the most significant causes of morbidity in patients undergoing surgery.[1]

The method used to evaluate pain in children should be selected based on important considerations such as age, general status, and level of pain recognition, and should be repeated at certain intervals. Factors such as developmental level, family attitudes, effects of inpatient hospital care, symbolic meaning of pain, and physiological response to pain can all be used to evaluate pain in children. It is difficult to distinguish between hunger, pain, and fear as causes of crying and anxiety in young children. The effectiveness of pain treatment in surgical practice and modern pediatric anesthesia is therefore assessed based on numerous physiological responses to stress.[2-4] It is not always possible to assess pain in infants or in children with cognitive or physical disorders. In these cases, observational evaluation of behaviors and biological methods can be used as alternative tools. QUESTT is the standard approach for the evaluation of pain in children.[2]

Q–Question the child

U–Use pain rating scales

E–Evaluate child's behavior

S–Secure parent's involvement

T–Take cause of pain into account

T–Take earliest action

The transversus abdominis plane (TAP) block is one of the abdominal field blocks used for the treatment of acute postoperative pain after lower abdominal surgical procedures. First described by Rafi[5] TAP block is a procedure in which local anesthetic agents are administered to the neurofascial gap between the internal oblique and the transversus abdominis muscles located in the anterolateral region of the abdominal wall, in order to block the lower thoracic and upper lumbar nerves. The analgesic efficacy of TAP block has been demonstrated with several studies.[6] TAP block is a procedure that local anesthetic agents are given to anatomic neurofascial gap between the internal oblique and transversus abdominis muscles located in the anterolateral region of abdominal wall to block anterior branches of thoracic intercostal (T7-T12) and first lumbar (L1) nerves. While TAP block application is performed with resistance loss technique by using Petit's triangle, a part of the procedure is performed with USG.[7-11] Sharma and co-workers[12] noted that TAP block provides effective long-lasting analgesia for patients undergoing abdominal surgery. Ultrasound-guided (USG) TAP block provides effective analgesia in the postoperative period and allows the high level of analgesic distribution to the nerves under the muscle fasciae.[5]

The innervation of inguinal region (skin, muscles, and parietal peritoneum) is mediated by the anterior branches of the spinal nerves between T7 and L1. The intercostal nerves (T9-T11), subcostal nerves (T12), ilioinguinal (L1) and iliohypogastric nerves (T12-L1) are located in the area between the internal oblique muscle and the transversus abdominis muscles which is called the "TAP". The block is performed to anaesthetize the neurofascial area, which is an anatomically empty space. The needle should be placed slightly directed toward the cranial just above the external part of the iliac crest where the lateral part of the latissimus dorsi muscle inserts, behind the midaxillary line. The needle then passes through the internal oblique muscle and the fascia. A second "click" is felt when passing the fascia. After careful aspiration, the appropriate volume of local anesthetic agent is injected. When the technique is performed with USG, the needle tip can be advanced to the TAP more safely and accurately. USG technique provides visualization of the three abdominal muscles at the lateral point of the rectus abdominal muscle, allowing for different injection points appropriate for varied surgical procedures. Selective blocks of the ilioinguinal and the iliohypogastric nerves are possible with USG, as in a cadaver study, and the three different muscle layers (external, internal oblique, and transversus abdominis) are visible across the aponeurotic area at the lateral abdominal wall. If local anesthetic is administered to the fascial layer, the distribution may be seen. It is defined as the TAP. We conducted a study to evaluate the effectiveness of the USG guided TAP block, using two concentrations of levobupivacaine for postoperative pain control in pediatric patients who undergoing unilateral inguinal surgery.

METHODS

This randomized, controlled, single-center, prospective, phase 4 clinical trial was conducted in the Department of Pediatric Surgery at Ankara University in 2015. A total of 50 pediatric patients between the ages of 2 and 12 years (American Society of Anesthesiologists (ASA) health status 1-2 levels) scheduled for unilateral inguinal hernia surgery were included in this study. The Turkish Health Ministry and Turkey Medical Medicines and Medical Devices Authority approved the study (Date: 30.05.2015, Decision No: 93189304-514-04-01) and consent forms were obtained from all patients before the surgery. Patients with skin infections near the incision site, coagulation problems, allergies to any of the analgesic agents used in the study, and chronic cardiac-renal and hepatic insufficiencies were excluded from the study. All procedures were carried out in accordance with the ethical rules and the principles of the Declaration of Helsinki. Patients were randomly divided into two treatment groups by closed envelope method.

All fifty patients underwent unilateral inguinal surgery. The patients randomized to group C (control) received the TAP block with 0.5 ml/kg Levobupivacaine 0.25%. The patients in group LD (low dose) received the TAP block with 0.25 ml/kg Levobupivacaine 0.25%. The age and weight of patients were recorded.

Routine monitoring (ECG, non-invasive arterial blood pressure, arterial oxygen saturation) of the patients was performed in the operating room, and the data were recorded as preoperative control data.

A peripheral venous line with an appropriately sized intravenous catheter was opened for cooperative patients. 1% propofol 2 mg/kg (propofol-lipuro 1%, B. Braun, Melsungen, Germany) was administered by intravenous line. For non-cooperative patients, induction was performed by first administering 8% sevoflurane (Sevorane® Liquid %100, Queenborough, England) and 50% nitrous oxide and 50% oxygen combination by facial mask, and then 1% propofol 2 mg/kg was administered by intravenous line. Laryngeal mask was placed following induction. Anaesthesia management was provided by 2 % sevoflurane with a combination of 50 % nitrous oxide and 50 % oxygen. Intravenous fluid maintenance was provided with a sodium chloride 0.3% and glucose 3.3% infusion at a rate of 10 ml/kg/h.

Preoperative preparations were performed according to the postoperative regional analgesia technique to be performed. TAP block dosage was calculated for all patients according to assigned study group and weight.

The lateral abdominal wall of the patients (in supine position) was sterilized with povidone-iodine. For all procedures, the practitioner was on the same side as the procedure side. A 10-18 MHz linear USG probe coated with a sterile sheath was placed transversely between the costal margin and the iliac crest on the same side of the abdominal wall. When necessary, the USG probe was directed to the cephalic-caudal, the anterior-posterior direction, or the angle needed to visualize the three lateral abdominal muscles clearly, and the initial image was optimized. Once the external oblique, internal oblique, and transversus abdominis muscles were clearly visualized, a 50 mm, 20-gauge needle (B.Braun Stimuplex, Melsungen, Germany) was inserted and advanced from the anterior surface of the USG probe by in plane technique. After visualization of needle placement in the TAP with

USG, levobupivacaine 0.25% was injected at a concentration of 0.5ml/kg for patients in group C and at a concentration of 0.25 ml/kg for patients in group LD. Once the distribution of the anesthetic agent was observed in the TAP, the needle was removed, and surgery was allowed to proceed. After the surgical procedure, administration of anesthetic gasses was discontinued and patients were ventilated with 100% oxygen until oxygen saturation values reached 97% and above and patients had spontaneous breathing. The type of surgery, duration of surgery, intraoperative hemodynamic parameters were recorded. The time of completion of surgery was recorded as t=0 and patients were followed at specific intervals: 10 min, 20 min, 30 min, 60 min, 6h, and 24h. During this period, all of the patients were evaluated using the modified aldrete recovery scores. Patients with a postoperative score of 7 or above were taken to the recovery room. Pain levels were evaluated using the Observer Pain Scale, Visual Analog Scale (VAS), Faces scala and data were recorded. Observer pain scala and face scala used for all of the patients. VAS used for patients who above 4 years. A designated researchers assessed pain with a four-graded observer scale (no pain, mild pain, moderate pain, severe pain). The child with reported pain by using a six-graded faces scale. In every patient, the result of the four-graded scale was recorded before the evaluation with the faces scale. Children asleep were considered not to be in pain. The first analgesic request time, total analgesic consumption, and any adverse effects such as nausea or vomiting were recorded. Supplementary analgesia was to be provided when the observer considered the child to be in moderate or severe pain or the child recorded pain scores of 3, greater on the faces pain scale or VAS score was 4 or above. Paracetamol 15 mg/kg was administered as supplementary analgesia. After paracetamol administration, ibuprofen 10 mg/kg was given to patients who had still pain.

Statistical Analysis

The data analysis of the data obtained in the study was performed using SPSS for Windows v. 15.0 (IBM Corp, NY). Numerical values were expressed as mean±SD or median (interquartile range). The student's t-test was used to compare the normal distribution of the numerical data with the Shapiro-Wilkes test followed by the normal distribution and the result was evaluated according to the equality of variances. The Mann-Whitney U test was used to compare non-normal numerical data. Categorical data were given as numbers. Pearson's Chi-square test and Fisher's exact test were used for comparison of categorical data. The Wilcoxon test was used for intra-group comparison. $p<0.05$ was considered significant.

RESULTS

For both groups, there was no statistically significant difference in gender, weight, age, or duration of surgery.

When intraoperative heart rate was evaluated (at 1, 3, 5, 10, 15, 20, 30 and 40 min), there was no statistically significant difference between groups. Heart rate values at 5, 10, 15 and 20 min. were significantly lower in the C group than at 1 min ($p<0.05$). The heart rate value at 15 min. was found to be significantly lower in the LD group than at 1 min ($p<0.05$) (**Figure 1**). There was no significant difference between groups in terms of average arterial blood pressure (**Figure 2**). There was no significant difference between groups in terms of recovery score (**Figure 3**). There was no statistical difference between groups in terms of the Observer Pain Scale (**Figure 4**). There was no statistical difference between groups in terms of the Faces Scale.

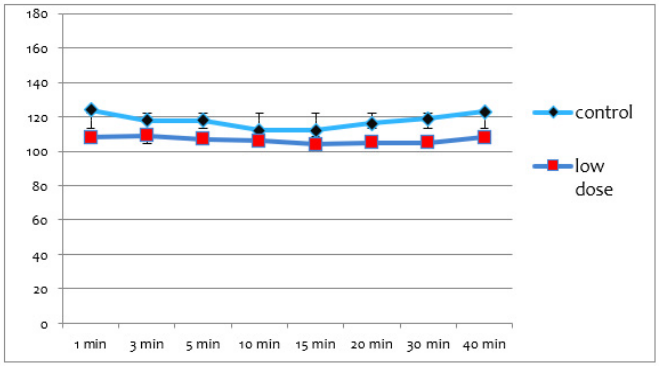

Figure 1. Heart rates

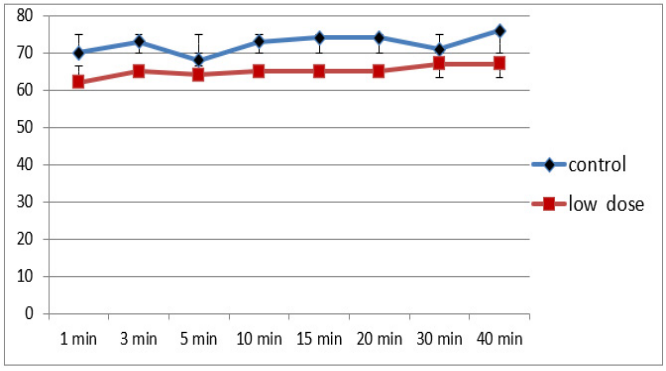

Figure 2. Average arterial blood pressure

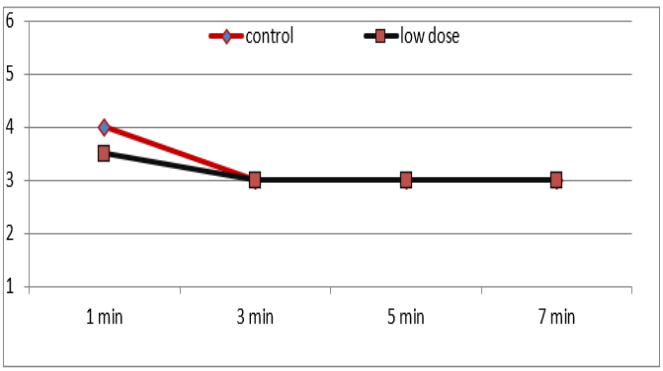

Figure 3. Modified Aldrete Recovery Score

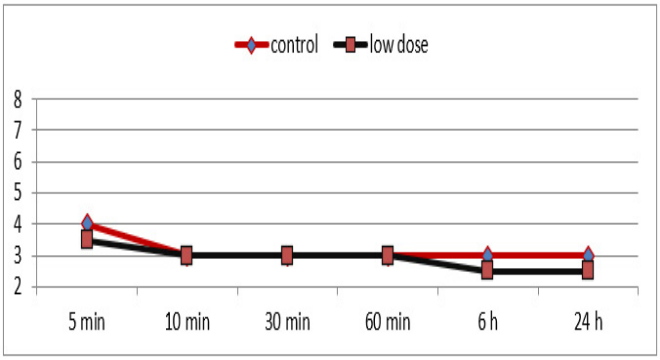

Figure 4. Observer Pain Scale

Fifty patients were included in our study (C group: 25, LD group: 25). No postoperative analgesic was required for 13 patients in the C group and 11 patients in LD group. At t=30 min post-surgery. There were 12 patients requiring postoperative analgesic in the C group, compared to 14 in the LD group. There was no statistically significant difference between groups in terms of analgesic requirement or patient satisfaction. There were no complications reported related to the procedure.

DISCUSSION

Recently, the number of block applications performed in pediatric patients has increased, because the use of USG increases the possibility of success and reduces the risk of complications. Based on our results, we think that TAP block with low dose levobupivacaine may be successful for managing pain with less toxicity, and may reduce complications due to local anesthetic. We aimed to control the postoperative pain in pediatric patients with TAP block application with low dose levobupivacaine. According to Taddio and co-workers[13] neonates having pain experience had different behaviors according to their pediatric ages, so they found that the main purpose of pain control was to prevent pain instead of reducing the pain. Therefore, pain management in pediatric patients is very important. The negative effects and complications of pain include decreased respiratory movements and coughing limitations. This situation may cause atelectasis and pulmonary complications. Severe pain may cause cardiac arrhythmia, hypertension, and myocardial ischemia. Immobility and prolongation of hospital stay may increase the likelihood of thromboembolic complications.[14-16] Findlow and co-workers[17] found that ilioinguinal/iliohypogastric nerve block with bupivacaine was more effective than caudal block. We preferred levobupivacaine because of lower cardiotoxicity. In a randomized controlled trial, Carley and co- workers[18] cited that TAP block reduced postoperative opioid consumption, and also that VAS scores was found to be lower in the TAP group (both at rest and motion). We thought that TAP block with USG guidance was safer based on a previous study so we conducted our study with USG guidance. The local anesthetic dose should be adjusted to the child's age, weight, physical condition, and operation area.[19]

Takashi et al.[20] used TAP block for 40 patients undergoing gynecologic procedures and found that local anesthetics might reach toxic blood concentration levels and sometimes cause systemic toxicity. Therefore, they recommended reducing local anesthetic doses for TAP block to minimize the risk of toxicity. But this study was conducted in adult patients. Therefore, we also reduced the dose for pediatric patients who were more vulnerable to toxicity. For blockage, Jacobs and co-workers[21] used different levobupivacaine doses for 5 neonates and 5 babies.

They administered 0.25% levobupivacaine 1 ml/kg for unilateral blockage and 0.25% levobupivacaine 0.5 ml/kg for bilateral blockage, and found that the TAP block with levobupivacaine was effective in all patients. Also, they cited that a low dose of levobupivacaine might be used, and suggested further dose studies.[20,21] In our study, we used two different levobupivacaine doses and found that low dose efficacy was the same as normal dose efficacy.

Desgrandes and co-workers placed a subcostal TAP catheter in a 4-year-old patient undergoing upper abdominal surgery and provided successful pain management. Through this catheter, 0.25% levobupivacaine 0.5 ml/kg of was administered and then 0.25% levobupivacaine 1.5 ml/kg was given continuously by pump.[22] We preferred levobupivacaine as a local anesthetic agent and compared the success of TAP block application in two different doses in pediatric patients at the same concentration for postoperative pain control.

Sahin and co-workers[23] cited that TAP block provided long-lasting postoperative analgesia for unilateral inguinal hernia operations and reduced analgesia requirements to 0.5 ml/kg of 0.25% levobupivacaine diluted with 1/200000 adrenaline when compared to infiltration anaesthesia. Willschke and co-workers[24] cited that abdominal wall blocks with USG was a successful regional anaesthesia technique for providing postoperative analgesia. In our study, we used TAP block with USG to reduce complications and increase the success rate.

Suresh and co-workers compared two different levobupivacaine concentrations in pediatric patients undergoing inguinal hernia operations and found analgesia difference between the two doses, and less analgesia requirement in the high dose group 24 hours after the surgery. In our study, this is one of the important outcomings of our work.

Frederikson and co-workers[25] showed that the analgesic activity of USG TAP block using a 50:50 combination of 1% lidocaine 0.3 ml/kg and 1% ropivacaine diluted with 1/200000 epinephrine in 8 children. In another study of authors, they compared the analgesic activity of TAP block with USG to ilioinguinal block with USG[26] and found that recovery room pain scores, morphine consumption, ibuprofen usage, and satisfaction score were similar in both groups, but that clinical pain scores and ibuprofen usage were higher in the TAP group. We found that 0.5 ml/kg of 0.25 ml/kg doses of 0.25% levobupivacaine were effective for postoperative analgesia. Therefore, we think that low dose levobupivacaine might be suitable for postoperative pain management in pediatric patients at risk for toxicity with TAP block.

TAP block complications include bowel hematoma, liver laceration, and temporary nerve blockage.[27,28] The spleen and kidneys are other organs that may be injured during the procedure. A case report of a liver laceration in an adult patient has been reported due to TAP block. Farooq and Carey[27] reported liver lacerations in TAP blocks with the landmark technique. Lancaster and Chadwick[29] also reported liver laceration in TAP block with USG. We did not observe any complications due to TAP block.

Multiple analgesia methods may lead to more than one specific problem in the neonatal group of patients. With TAP block with USG, less local anesthetic agent was required and toxicity risk was reduced compared to infiltration method.[30,31] Fredrickson and co-workers[32] used TAP block in 4 newborns

undergoing abdominal surgery to provide pain management for the intraoperative and early postoperative period.

Vincent[33] reported that the optimal dose of levobupivacaine is not clearly established and is still empirical. At 0.4 mg/kg, none of the 1- to 5-year-old children presented toxicity signs, and there is evidence that this dose could be increased to obtain a better efficacy. And children's weight must be considered to evaluate their clearance and anticipate any risk of toxicity.

In children, quality of postoperative pain control provided by TAP block using levobupivacaine 0.4 mg·kg–1 administered as either HVLC or LVHC did not differ and was associated with a very low risk of local anesthetic systemic toxicity.[34]

CONCLUSION

We conclude that it is very important to use low-dose local anaesthesia in the pediatric age group, which is more at risk for local anesthetic toxicity. TAP block provides effective blockage with low dose analgesia. Ultrasound-guided TAP block with 0.25% levobupivacaine 0.25 ml/kg was found to be effective and reliable in pediatric patients. In conclusion, because toxicity is a significant consideration for the pediatric age group, we recommend TAP block for pediatric patients.

ETHICAL DECLARATIONS

Ethics Committee Approval

The Turkish Health Ministry and Turkey Medical Medicines and Medical Devices Authority approved the study (Date: 30.05.2015, Decision No: 93189304-514-04-01).

Informed Consent

Parents of all patients signed a free and informed consent form.

Referee Evaluation Process

Externally peer-reviewed.

Conflict of Interest Statement

The authors have no conflicts of interest to declare.

Financial Disclosure

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

Author Contributions

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

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MHA MEDIHEALTH ACADEMY
Surgery on Children
Review
doi: 10.51271/SOC-0045

The essential principles of evaluation and management pathways in prenatally detected fetal anomalies

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Received: 08/02/2025 ◆ **Accepted**: 25/03/2025 ◆ **Published**: 01/07/2025

Cite this article: Sarma VP. The essential principles of evaluation and management pathways in prenatally detected fetal anomalies. *Surg Child.* 2025;2(3): 101-108.

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ABSTRACT

The evaluation and management of prenatally detected fetal anomalies constitute a challenging problem. The optimal outcome for the fetus and the mother depends on the accurate diagnosis, appropriate evaluation, and informed, scientific decision-making regarding prenatal, perinatal and postnatal management. The article aims to analyze the complexities in evaluation and management of prenatally detected fetal anomalies and highlight the challenges in therapy. The current literature was reviewed to understand the issues with regard to treatment of prenatally detected fetal anomalies in the light of clinical experience and practical considerations with regard to management of these variegated group of disorders. The observations were summarized to present a broad outline of the common and challenging set of conditions. The recent advances in prenatal diagnosis and widespread adoption of fetal imaging have increased the detection of fetal anomalies. The availability of maternal screening investigations and newer methods of genetic testing have further added to the diagnostic spectrum. The abnormalities can range from minor alterations of uncertain significance to major defects with grave prognosis. The establishment of definite clinical practice guidelines for screening, diagnosis and therapy are mandatory to bring clarity to management. A deep understanding of the natural history of these disorders, their evolution during gestation and the effect on the mother, fetus and the pregnancy are critical to the optimal management of these conditions.

Keywords: Fetal diagnosis, prenatal diagnosis, maternal screening, fetal genetic testing, fetal diagnostic testing, fetal therapy, perinatal management, termination of pregnancy for fetal anomaly

INTRODUCTION

The evaluation and management of prenatally detected fetal anomalies constitute a challenging problem. The optimal outcome for the fetus and the mother depends on the accurate diagnosis, appropriate evaluation, and informed, scientific decision-making regarding prenatal, perinatal and postnatal management. The recent advances in prenatal diagnosis and widespread adoption of fetal imaging have increased the detection of fetal anomalies.

The availability of maternal screening investigations and newer methods of genetic testing have further added to the diagnostic spectrum. The abnormalities can range from minor alterations of uncertain significance to major defects with grave prognosis. The inputs of multiple specialists are required to ascertain the severity and prognosis of an anomaly, and the need for fetal diagnostic and therapeutic modalities. The decision regarding termination of pregnancy for fetal anomaly and need for advanced perinatal therapy or changes in obstetric management are also vital in this regard.

AIM

The article is designed as a narrative review on the broad topic of prenatally detected fetal anomalies, with the aim of summarizing and synthesizing clinically relevant information from literature. The aim is to review the common prenatally detected fetal anomalies with regard to their diagnosis, evaluation and management principles. The article aims to analyze the complexities in evaluation and management of prenatally detected fetal anomalies and highlight the challenges in therapy, while formulating and elaborating the scientific approach to treatment.

MAIN TEXT

The evaluation and management of prenatally detected fetal anomalies constitute a challenging problem. The optimal outcome for the fetus and the mother depends on the accurate diagnosis, appropriate evaluation, and informed, scientific decision-making regarding prenatal, perinatal and postnatal management. The effect of the fetal anomaly on the physiological interaction between the fetus and mother

is complex and variable, with a significant bearing on the outcome of pregnancy. These factors should also be accounted for in the management of such cases.[1-3]

Prenatally detected fetal anomalies constitute a challenging problem because of the following reasons[4-6]:

- The wide spectrum of the disorders (hydronephrosis, heart disorders, hydrocephalus etc.)
- Differentiation of transient physiological changes from structural anomalies (renal pelvic dilatation, ventricular dilatation, bowel dilatation etc.)
- Variable significance and severity of the defect that require detailed evaluation like grading/ detailed organ measurements (renal anomalies, lung anomalies, cardiac anomalies)
- Possibility of evolution of the problem during the gestation (hydronephrosis, lung lesions, teratoma etc.)
- Occurrence as a part of a syndrome/ genetic disorder/ association (Trisomy 21; Potter's syndrome, VACTERL association of vertebral, anorectal, cardiac, tracheoesophageal, renal and limb anomalies etc.) where multiple anomalies can co-exist
- Findings like oligihydramnios, polyhydramnios, fetal hydrops etc. could be secondary manifestations of an underlying anomaly (fetal obstructive uropathy, fetal intestinal obstruction and fetal teratoma/ major lung lesion, respectively)
- Decision on the need for further evaluation/fetal imaging (fetal MRI)/invasive fetal diagnosis (amniocentesis/ chorionic villus sampling-CVS)
- Decision on the need for antenatal intervention/ex-utero intrapartum treatment (EXIT) procedure
- Decision on the need for termination of pregnancy, in cases incompatible with life
- Determination of the prognosis and parental counselling

METHODS

This narrative review analyses current literature with emphasis on relevant guidelines and recommendations, with reference to the evaluation and management of prenatally detected fetal anomalies. The literature was reviewed, in the light of the clinical experience and observations made with regard to the management of these cases. The databases searched for literature this narrative review included EMBASE, PubMed, Cochrane, and Web of Science. The studies analysed were English-language, peer-reviewed journal articles, particularly consensus statements and commonly followed guidelines.[1-5] Institutional review board, as well as the ethical committee approval were not required. An effort has been made to break down the problem into its various aspects including clinical challenges, investigative modalities, therapeutic options and their appropriate use. The generally accepted principles in the treatment of usually encountered anomalies were reviewed thoroughly to simplify and tabulate management principles. An overview of the various clinical scenarios and a broad outline of the management strategies are presented here.

RESULTS

Prenatal Genetic Testing

The availability and widespread application of maternal screening and prenatal genetic testing modalities have revolutionized the diagnosis of genetic, metabolic and syndromic disorders. But the clinical application of these new techniques has presented challenges regarding patient selection, communication with patents, factual interpretation of results, analysis of benefit as well as moral and ethical concerns. The methods employed for prenatal genetic testing, maternal screening and fetal imaging are to be performed, interpreted and communicated with diligence and caution.[2-6] A brief summary of the applications of the modalities of fetal genetic testing, maternal screening, fetal diagnostic tests and fetal imaging techniques is presented here (**Table 1**).

Prenatal Sonological Diagnosis

Ultrasound scan (USG) in the first trimester is helpful in accurate dating, diagnosis of twin chorionicity, and early diagnosis of major structural anomalies like Anencephaly. Measurement of nuchal translucency (NT) on USG is a marker for fetal genetic and chromosomal disorders, neural tube defects (NTD), and cardiac anomalies.[1,2,4] Second trimester USG is helpful to assess major defects of the heart, brain and spine, kidneys, abdomen, craniofacial region, limbs etc. It is to be done between 18 weeks and 22 weeks of pregnancy. Detailed fetal imaging using 3D/4D USG, fetal echocardiogram, fetal MRI and serial USG are indicated in complex anomalies. Fetal structural defects can occur with or without fetal aneuploidy. Therefore, all patients should be offered a second-trimester USG between 18 and 22 weeks of gestation (+/- second-trimester maternal serum alfa fetoprotein-AFP).[3,5,6]

"Soft Markers" on Antenatal Sonology-What are They, and When Should They be Considered Significant?

With regard to the prenatally detected structural anomalies in the fetus on USG, it is imperative to recognize some of the abnormalities, that are generally referred to as sonological "soft markers". These findings should be reassessed diligently and interpreted carefully as they can be markers or features of very significant pathology, in spite of some of them being benign findings on many occasions. These findings generally mandate a detailed anatomical assessment and systemic survey to confirm findings and evaluate other organs.[7-9]

Many of these findings require systematic follow up based on the initial assessment of grade/ severity, possibility of associated pathologies and likelihood of complications. Some of these mandates additional testing like TORCH (Toxoplasma, Rubella, Cytomegalovirus, Herpes simplex) screening and fetal imaging like fetal ECHO/fetal MRI, while others mandate invasive fetal diagnostic modalities. Many of these could serve as markers of genetic disorders like Trisomy 21 or other causes of aneuploidy. They can also help to predict the occurrence of prenatal/postnatal complications like Intrauterine growth retardation (IUGR), Uteroplacental insufficiency (UPI), Oligohydramnios, preterm birth, preeclampsia, need for caesarean and still birth.[10-12]

Table 1. Fetal diagnostic modalities and their applications

Fetal screening and diagnostic modalities	Details	Timing	Advantages	Comments
Prenatal genetic screening tests				
Carrier screening	Couples undergoing IVF with increased risk of genetic/ chromosomal disorder; Blood or buccal smear.	On those planning pregnancy/ during pregnancy.	Can detect gene for inherited disorders like spinal muscular atrophy, cystic fibrosis, hemoglobinopathies, fragile X syndrome, and Tay-Sachs disease.	Can only assess the chance of aneuploidy/ other disorders.
Preimplantation genetic diagnosis		Before pregnancy. Only embryos that do not test positive for the disorders are transferred.		
Cell-free fetal DNA	Isolation of fetal cells in the maternal circulation by advanced sorting techniques permit genetic testing from a maternal blood sample. Accurate test for the common fetal aneuploidies.	Can be done from 10 weeks of pregnancy and beyond.	Can be used for the screening of Trisomy 21, 18, 13 and sex-chromosome aneuploidy. A positive cell-free DNA test result should be followed by a diagnostic test with amniocentesis or CVS.	It is not recommended for the screening of microdeletion. There is a low positive predictive value of this test in low-risk group and the possibility of 'no call' from inadequate sample.
Maternal screening				
First trimester screening (maternal serum biomarkers and US screening)	Pregnancy-associated plasma protein A (PAPP-A) and free beta-human chorionic gonadotropin (HCG); Nuchal Translucency (NT) on US.	At 10-13 weeks of pregnancy; the testing for NT should be done at 11-13 weeks, not earlier or later.	USG aids in precise dating, detection of twin chorionicity, and early detection of major structural abnormalities. A high NT measurement (> 3.5 mm) is a marker for fetal chromosomal disorders, structural anomalies (cardiac) and genetic disorders such as RASopathies.	The combined first trimester screening test can identify a pregnancy with increased chance of Down syndrome (trisomy 21) and Edward syndrome (trisomy 18).
Fetal diagnostic test: Chorionic Villus Sampling (CVS)	Transvaginal/trans abdominal; USG guided procedure. Sample taken from the placenta for the diagnosis of genetic and chromosomal disorders.	Between 10 to 13 weeks of pregnancy.	Detect conditions such as Down syndrome, Edwards' syndrome, or Patau's syndrome	Complications are pregnancy loss, bleeding, infection, rupture of membranes, and indeterminate results. Limb reduction defects and oromandibular hypogenesis are reported with early CVS.
Second trimester screening	The quad screen test is a maternal blood screening test for four markers: alfa fetoprotein (AFP), hCG, estriol, and inhibin-A.	Between the 16th-18th week of pregnancy	It screens for Down syndrome, Edwards syndrome (trisomy 18), and NTDs; The penta screen also measures hyperglycosylated hCG; MSAFP can be performed between 16 and 18 weeks for NTD screening.	An elevated AFP level in maternal serum/ amniotic fluid is seen in NTD, Abdominal wall defects and tumors like Sacrococcygeal Teratoma. Other markers like Estriol and HCG are useful in Aneuploidy screening
Second trimester USG	Assess major defects in the CVS, CNS, kidneys, abdomen, craniofacial region, Limbs etc.	Between 18 weeks and 22 weeks of pregnancy.	The second-trimester uterine arteries' pulsatility index (PI) can detect the presence of issues with the placenta that could lead to stillbirth.	Detailed USG for structural defects and feta ECHO are to be done in the second trimester for follow-up.
Fetal diagnostic test: amniocentesis	For diagnostic evaluation in the form of chromosomal, biochemical, histopathological, and microbial assessments.	Second trimester; can be performed from 15 weeks of gestation.	Common indications: advanced maternal age, known carrier state and history of genetic disorder in previous child.	When performed as a therapeutic procedure, it is done to reduce the volume of amniotic fluid in patients with polyhydramnios (Amniodrainage). Complications are fetal loss, bleeding, infection, rupture of membranes, amniotic fluid leak and indeterminate results.
Fetal blood sampling or cordocentesis	For haematological, immunological, and biochemical analysis. Used in diagnosis of fetal anemia, genetic disorders, fetal infections etc.	Considered safer to perform at 20 to 28 weeks of gestation	Can be used with therapeutic intent in fetal anemia, for drug administration and Rh disease.	The benefits include conversion to fetal transfusion.
Fetal urine sampling	Used to assess status of renal function in bilateral renal disease associated with oligohydramnios. Assessment of urine electrolytes, beta 2 microglobulin etc.	Second trimester.	Commonly indicated in complicated posterior urethral valves.	Abnormal urine biochemistry indicates poor prognosis for renal outcome and less benefit of fetal therapy.
Fetal imaging modalities				
3D/4D USG	Detailed assessment of fetal anomalies. Serial USG can help to define the natural history and progression of fetal disease like hydronephrosis, ventriculomegaly, lung anomalies etc.	After 10 weeks.	Can facilitate the performance of USG-guided intervention procedures; USG measurement of organ volume/ volume ratio/ size measurements that imply severity and prognostic significance (lung-to-head ratio, left ventricular wall thickness etc.)	Real-time USG gives information about fetal well-being like fetal movement and fetal vital functions like fetal heart rate variability and fetal breathing movements.
Fetal MRI	Defects detected on US that require detailed analysis can undergo ultrafast fetal MRI.MRI can image the fetus in any plane, providing a large field of view of the fetus and placenta with excellent soft tissue resolution of the brain, airway, lungs, and abdomen.	After 20 weeks.	Enable detailed study of fetal CNS malformations, lung malformations, cystic hygroma, teratoma etc.	MRI is not limited by fetal lie, oligohydramnios, overlying bone, or obesity. Advanced techniques provide volumetric data, spectroscopy, and functional images.

The most commonly observed soft markers on prenatal sonology and their details are listed below:[13-18]

1. **Fetal pyelectasis/ renal pelvic dilatation:**

- Definition**:** Renal pelvic anteroposterior diameter in transverse plane (APD) of 4 mm or more in second trimester and/or 7 mm or more in the third trimester.
- Pelvic APD of 10 mm or more is the criteria for definition of hydronephrosis.
- Fetal renal pelvic dilatation can be a transient physiological finding, but it can also be the sign of a major urinary tract disorder/marker of aneuploidy.
- A follow-up USG at 32 weeks of gestation should be performed. If the APD measures >7 mm at 30-week USG, postnatal follow-up is suggested.

- A significant proportion of prenatally detected pelvic dilatation, especially in the second trimester, will resolve either prenatally or during postnatal life.
- A significant progression of dilatation during prenatal follow up is indicative of a pathological cause.

2. **Single umbilical artery (SUA):**

- Definition: The absence of one of the umbilical arteries, which occurs in 0.5 to 5% of pregnancies. This is an isolated finding in 60–80% of cases.
- Isolated SUA is associated with small for gestational age (SGA) babies, higher rate of placenta or umbilical cord abnormalities and a higher rate of caesarean due fetal heart rate abnormalities.
- It is associated with a higher risk of SGA, preterm delivery, pregnancy-induced hypertension, requirement of intensive care and perinatal mortality.
- The finding should prompt targeted anatomical survey and growth assessment.

3. **Thickened nuchal fold (NF):**

- Definition: The thickening of the skin and the subcutaneous tissues on the posterior aspect of the fetal neck measuring 6 mm or greater before 20+6 weeks' gestation.
- In fetuses with normal USG screening for aneuploidy in the first trimester, thickened nuchal fold in second trimester is the most important soft marker in the detection of Down syndrome.
- Karyotyping should hence be offered when thickened nuchal fold is detected.
- Even with normal karyotyping, targeted USG and fetal ECHO are indicated when a thickened nuchal fold is identified, due to the association with structural defects, like congenital heart defects.

4. **Intra-cardiac echogenic focus (IEF):**

- Definition; A small echogenic spot inside the heart, most commonly seen in the left ventricle, having brightness equivalent to that of the bone.
- In about 90% of cases, it resolves by the third trimester.
- Isolated IEF are associated with an increased risk of Down syndrome.
- The presence of an IEF is not an indication for invasive procedures in low-risk populations for aneuploidy. In the presence of negative first trimester screening (FTS)/non-invasive prenatal testing (NIPT), IEF can be described as a normal variant.

5. **Isolated ventriculomegaly (VM):**

- Fetal cerebral ventricular dilatation: It is a dilatation of the lateral ventricle atrium to a width of 10 mm or more.
- Mild VM: 10–12 mm; Moderate VM: 12–15 mm; Severe VM: >15 mm. Isolated mild and moderate VM is more likely to regress or become stable, unlike severe VM.
- It can be a normal variant or be associated with aneuploidy, genetic syndromes, primary brain abnormalities, congenital infection and intracranial haemorrhage.
- MRI can be used for detailed evaluation of cases of VM and TORCH screening is also indicated.
- The prognosis of VM depends on the severity, the presence of other abnormalities and also an asymmetric pattern of VM.

6. **Choroid plexus cysts (CPC):**

- Definition**:** CPC is a small fluid-filled space ≥5 mm within the choroid plexus, which is sonologically discrete, usually seen at 16 to 24 weeks of gestation.
- Isolated CPC is usually an isolated finding in an otherwise normal low-risk pregnancy and not considered as a structural or functional abnormality.
- CPC typically regresses by 23 weeks regardless of the underlying karyotype.
- Amniocentesis is indicated only if other anomalies, like those of Trisomy 18 are present.

7. **Echogenic bowel:**

- Definition: Fetal bowel of similar or greater echogenicity than the surrounding bone or fetal liver. Two-third are detected during the first and the second trimesters.
- Echogenic bowel resolves spontaneously in ~20% of cases.
- Echogenic bowel can be a normal variant, and may also be associated with congenital viral infections like cytomegalovirus, aneuploidy (Down syndrome), intra-amniotic bleeding, severe UPI, meconium peritonitis, cystic fibrosis, anemia, and fetal growth restriction.
- It mandates detailed evaluation including evaluation for cystic fibrosis, TORCH screening and USG at 32 weeks to assess bowel obstruction.
- The likelihood of postnatal surgical intervention for intestinal anomalies is significantly increased if the finding is seen during the third trimester.

8. **Shortened humerus and femur:**

- Definition: Bone length below the 5th percentile for gestational age.
- Shortening of the fetal long bones is seen associated with aneuploidy, skeletal dysplasia, fetal structural anomalies, preeclampsia, stillbirth and IUGR.
- Shortened humerus and femur length may be an early sign of placental dysfunction. It mandates prenatal surveillance with serial USG for growth assessment and blood pressure monitoring.
- USG at 32 weeks is done to rule out skeletal dysplasia and to assess growth.

9. **Absent or hypoplastic nasal bone:**

- Definition: A nasal bone that is not visible in first trimester/ nasal bone with a length of <2.5 mm in the second trimester.
- It is described as one of the phenotypic features of Down syndrome, but also has racial differences.
- The finding mandates a detailed evaluation of fetal anatomy. Microarray studies should be done in addition to fetal karyotype when the finding occurs with other anomalies on USG.
- The prognosis is good if there are no other anomalies and karyotype is normal.

Soft marker screening is a tool in screening for non-aneuploidy-related conditions such as, structural anomalies and adverse pregnancy outcomes that requires follow-up during marker in the setting of a negative non-invasive prenatal testing (NIPT) result.[16-18]

DISCUSSION

The Common Management Pathways of Prenatally Detected Fetal Abnormalities

The broad, general pathways in management only apply for the specific abnormality, in isolation. The decision-making in complicated, multisystemic and genetic disorders are to be taken in consideration of the major, life-threatening anomaly as the key parameter, on an individualized basis.[19-24]

1. **Fetal abnormalities that usually need prenatal follow up and postnatal therapy:**

- Renal anomalies like hydronephrosis (unilateral/bilateral-without complications like oligohydramnios and Potter's syndrome), unilateral multicystic dysplastic kidney/ unilateral renal agenesis
- Isolated congenital diaphragmatic hernia and oesophageal atresia.
- Gastrointestinal anomalies like intestinal atresias, anorectal malformation, meconium ileus, enteric cysts and duplications, small and intact exomphalos/gastroschisis
- Tumors/cystic lesions like small sacrococcygeal teratoma, small lymphovascular malformations/cystic hygroma, benign cysts such as adnexal (ovarian) cysts and choledochal cyst
- Uncomplicated craniofacial/limb/chest wall abnormalities

2. **Fetal abnormalities that may require induction of preterm delivery:**

- Ruptured exomphalos/gastroschisis
- Progressive hydrops fetalis
- Progressive hydrocephalus
- Progressive hydrothorax
- Arrhythmias like supraventricular tachycardia
- Severe IUGR

3. **Fetal abnormalities that need planned caesarian section:**

- Large sacrococcygeal teratoma
- Severe hydrocephalus
- Large myelomeningocele
- Conjoined twins
- Large omphalocele/gastroschisis
- Large cervical lympho-vascular malformation/cystic hygroma
- Malformations that require preterm delivery in the presence of inadequate labor/fetal distress

4. **Fetal abnormalities that may require an ex-utero intrapartum treatment (EXIT) procedure:**

- Conditions that require emergency upper airway access like congenital high airway obstruction syndrome (CHAOS), large cervical tumor like teratoma and mass lesions causing obstruction to the oral cavity/trachea (lymphatic or vascular malformations)
- Conditions requiring immediate Extracorporeal membrane oxygenation (ECMO) cannulation
- Thoracic/mediastinal mass lesion that prevents lung expansion

5. **Fetal abnormalities that are usually managed by termination of pregnancy for fetal anomaly (TOPFA):**

- Severe neurological defects like anencephaly
- Severe chromosomal anomalies like Trisomy 13
- Bilateral renal agenesis and infantile polycystic kidney disease
- Severe metabolic disorders like Tay-Sach's disease
- Severe and lethal bone dysplasias like recessive osteogenesis imperfecta

The essential principles regarding the evaluation and management of some of the common prenatally detected fetal anomalies is summarized here (**Table 2**).

Major congenital malformations are abnormalities that are severe enough to reduce life expectancy or compromise normal function. They cause neonatal death in more than 20% of cases. They are considered to be lethal if they cause stillbirth or infant death in 50% of cases. If newborn infant with major malformation cannot survive without medical intervention, then they are considered severe.[22-27]

The Antenatal Diagnosis of A Fetal Anomaly can Lead to One of the Following Outcomes

- Continuation of pregnancy, without the need for any further evaluation other than the routine follow up scans (applies when the anomaly detected is deemed to be physiological/ mild with no perceived risk to the fetus or mother/no risk of worsening or complications)
- Continuation of pregnancy, with targeted serial follow up scans/detailed USG assessment or grading (to assess the evolution of the problem/ involvement of other organ systems/complications like organ damage)*
- Continuation of pregnancy with the need for further imaging like CT/MRI or fetal sampling (when major congenital disorders are suspected, with the findings of evaluation having a significant bearing on the outcome)*
- Continuation of pregnancy with the need for maternal/fetal intervention (in occasional cases of treatable major disorders where the intervention is expected to reduce morbidity and mortality of the fetus)*
- Continuation of pregnancy with changes in the timing and mode of delivery to improve the outcome of the fetus (option of early induction of delivery/caesarian section) *
- Termination of pregnancy for fetal anomaly (TOPFA) after counselling when a serious disorder that is incompatible with life is diagnosed sufficiently early in gestation

*Delivery should be conducted only at an appropriate center, with maternal transport if required

The apt use of the techniques of maternal screening, genetic testing, first trimester USG screening and diagnostic fetal testing would help in timely diagnosis and early detection of major fetal disorders and major structural anomalies, thus offering an opportunity for early termination of pregnancy. Similarly, the use of advanced fetal imaging and fetal sampling studies can help to assess the severity of the disorder in doubtful situations, and in also assist their follow up. Though fetal therapy is not yet widely practiced, the

Table 2. Common fetal anomalies and the general principles of evaluation and management in specific situations

Fetal anomaly	Associated issues	Prognostic factors	Complications (Prenatal and Postnatal)	Fetal diagnosis/ fetal therapy (In select cases)	Perinatal management: general guidelines
Fetal cardiac defects	20-40% incidence of extracardiac anomalies; high incidence of chromosomal anomalies; occur in association with syndromes, CDH, OA, genetic disorders etc.	Single/multiple defects; associated conditions; type of lesion (duct dependent)	Fetal Arrhythmias; IUGR; Cardiac failure; Hydrops fetalis; Preterm labour; Chorioamnionitis	Fetal ECHO (18-22 weeks); Fetal genetic testing; Fetal therapy includes medical therapy for arrhythmias, FETENDO	Delivery at specialised center with access to cardiology services; Term gestation ~39 weeks; Vaginal delivery; Caesarian for obstetric indications.
Neural tube defects (usually associated with Hydrocephalus/ myelomeningocele-MMC)	Associated with other CNS anomalies, non-CNS anomalies and genetic disorders.	Severity of defect; type of defect (Chiari type II); associated conditions.	Progressive hydrocephalus; neurological damage; complications due to large MMC	Fetal Neurosonogram; Fetal MRI; Fetal genetic testing; Seral USG; Fetal surgery for MMC and FETENDO for Hydrocephalus.	Delivery at center with access to Paediatric Neurosurgery services; Term gestation; Vaginal delivery; Caesarian delivery for possible dystocia due to MMC/ large Hydrocephalus.
Congenital diaphragmatic hernia	Associated anomalies (cardiac, NTD, genetic disorders); syndromic association.	Side of the defect; fetal lung-to-head ratio (LHR); Liver herniation; associated anomalies; left ventricular wall thickness	Polyhydramnios; Lung immaturity; Pulmonary arterial hypertension; pulmonary hypoplasia; Persistent fetal circulation.	Fetal MRI; Fetal ECHO; Fetal genetic testing; Serial USG; Fetal therapy includes Antenatal steroids and FETO. EXIT for reversal of FETO.	Delivery at center advanced neonatal ventilation, ECMO and Paediatric Surgical services; Term gestation (38-39 weeks)-planned induction; Vaginal delivery; Caesarian for obstetric indications.
Oesophageal atresia-tracheoesophageal fistula	Associated anomalies (cardiac, ARM, renal, skeletal); Chromosomal disorders like Trisomy 18; syndromic association (CHARGE).	Type of the anomaly; associated conditions/ syndromes (VACTERL); growth retardation	Polyhydramnios; Growth retardation; Cardiac anomaly related issues; Respiratory distress; Aspiration pneumonia.	Fetal MRI; Fetal ECHO; Fetal genetic testing. Amniodrainage in severe polyhydramnios.	Delivery at center with advanced neonatal ventilation and paediatric surgical services; Term gestation-38 weeks; planned induction; Vaginal delivery; Caesarian for obstetric indications.
Obstructive airway lesions of the fetus, congenital high airway obstruction syndrome (CHAOS)	Lymphovascular lesions like cystic hygroma are associated with genetic syndromes/ chromosomal disorders in 40-50% cases; cervical teratomas are usually isolated lesions.	Type of lesion; size of the lesion; hydrothorax; associated conditions.	Fetal anemia, thrombocytopenia; Fetal hyperdynamic circulation with placentomegaly, hydrops fetalis and heart failure; Polyhydramnios; Maternal Mirror syndrome (preeclampsia); Dystocia	Fetal MRI; Fetal ECHO; Fetal genetic testing in Cystic Hygroma; EXIT Procedure in case of airway compromise.	Delivery at center with advanced neonatal ventilation and paediatric surgical services; Term gestation-38 weeks; planned induction for Vaginal delivery; Caesarian for dystocia/ obstetric indications.
Abdominal wall defects: exomphalos, gastroschisis	Associated genetic disorders, syndromes (BWS, OEIS), cardiac defects in exomphalos; associated gastrointestinal anomalies in gastroschisis-other disorders are rare.	Type of defect; size of defect; presence or absence of cover; herniated viscera; associated conditions/ syndromes	Oligohydramnios in Gastroschisis; Rupture of sac in Exomphalos; Bowel related complications in Gastroschisis; IUGR; Fetal death; Preterm delivery; Term stillbirth	Fetal ECHO, Fetal genetic testing in Exomphalos; Serial USG in Gastroschisis.	Delivery at center with advanced neonatal and paediatric surgical services Term gestation (37-38 weeks); planned induction for Vaginal delivery; Caesarian if large Exomphalos/ Gastroschisis with extracorporeal Liver.
Sacrococcygeal teratoma	Genetic disorders are not associated; commonest type is external; benign lesion with potential for malignant change; highly vascular.	Type of lesion; size of lesion; vascularity.	High output cardiac failure; Fetal anemia, thrombocytopenia; Hydrops fetalis; Polyhydramnios; Maternal Mirror syndrome (preeclampsia); Preterm delivery; Dystocia; Malignant transformation in infancy	Fetal ECHO; Serial USG; Fetal therapy includes fetal blood transfusion, Amniodrainage, FETENDO for LASER vascular occlusion, and fetal surgery.	Delivery at center with neonatal and paediatric surgical services; Term gestation~ 38 weeks; Vaginal delivery; Preterm Caesarian for large lesions and high-risk cases.
Congenital lung malformations (CPAM, BPS, CLE)	CPAM can be microcystic/ macrocystic; BPS can be intralobar/extralobar and has systemic blood supply. No genetic disorders.	Size of the lesion; extent of involvement of lung; vascular supply	High output cardiac failure; Polyhydramnios; Hydrops fetalis; Chance of spontaneous involution; may become apparent again postnatally; Respiratory distress in newborn (CLE)	Fetal ECHO; Serial USG; Fetal therapy includes Thoracoamniotic shunt, maternal steroids and fetal surgery.	Delivery at center with advanced neonatal ventilation and paediatric surgical services; Term gestation ~38 weeks; Planned induction for Vaginal delivery; Caesarian for high-risk cases.
Adnexal cysts (Ovarian cysts)	Commonly sporadic, association with maternal DM and Rh isoimmunization.	Size of the lesion; associated syndromes (Mc Kusick -Kaufman); associated gastrointestinal and genitourinary defects.	Torion; Hemorrhage; Rupture and ascites in large cysts (>6cm) Polyhydramnios in large cyst;	Serial USG (Monitor size and to be differentiated from other intraabdominal cysts- mesenteric/ omental/ duplication); USG aspiration of large cysts.	Delivery at center with paediatric surgical services; Term gestation ~38 weeks; Vaginal delivery.

Continues

Table 2. Common fetal anomalies and the general principles of evaluation and management in specific situations (Continues)

Congenital urinary tract obstruction (CAKUT)	Especially significant for severe bilateral hydronephrosis with renal damage, as in PUV, bilateral PUJO etc. Genetic disorders are seen in multicystic kidneys, renal agenesis etc.	Inherent renal dysplasia; Bladder outlet obstruction.	Progressive renal damage due to pressure changes/ reflux; Oligohydramnios; Pulmonary hypoplasia;	Serial USG and grading of HN; Fetal Urine sampling for electrolytes and beta 2 microglobulin; Fetal genetic testing in select cases.	Delivery at center with paediatric surgical services; Term gestation ~38 weeks; Vaginal delivery.
Intestinal atresias	Commonly duodenal and jejunoileal atresias. High incidence of chromosomal anomalies and other defects in duodenal atresia and other intestinal issues in jejunoileal atresias.	Type of atresia; associated anomalies.	Polyhydramnios; Growth retardation; Complications due to associated anomalies; bowel related issues like perforation, meconium peritonitis and meconium pseudocysts.	Serial USG; Fetal ECHO; Fetal genetic study in select cases with suspicion of genetic disorders. Amniodrainage in severe polyhydramnios.	Delivery at center with paediatric surgical services; Term gestation ~38 weeks; Vaginal delivery.
Anorectal malformations	Commonly rectourethral fistula in male and rectovestibular fistula in female; Association with syndromes (VACTERL) and genetic disorders.	Type of anomaly; Associated anomalies (OA/ cardiac, renal)	Complications due to associated malformations.	Serial USG; Fetal ECHO; Fetal genetic study.	(Dictated also by major associated anomalies-OA, cardiac). Delivery at center with paediatric surgical services; Term gestation ~38 weeks; Vaginal delivery.

detailed diagnosis and prognostication can aid in instituting the appropriate perinatal management.[28-30]

CONCLUSION

The evaluation and management of fetal anomalies poses unique challenges, due to the variegated nature of the disorders and the wide spectrum of presentation. The distinction of minor, physiological alterations from actual structural defects is crucial in this regard. The establishment of definite clinical practice management pathways for screening, diagnosis and therapy are mandatory to bring clarity to management. Greater understanding of the natural history of these disorders, their evolution during gestation and the effect on the mother, fetus and the pregnancy are critical to the optimal management of these conditions.

ETHICAL DECLARATIONS

Referee Evaluation Process

Externally peer-reviewed.

Conflict of Interest Statement

The authors have no conflicts of interest to declare.

Financial Disclosure

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

Author Contributions

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

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MHA MEDIHEALTH ACADEMY

Surgery on Children

Review

doi:10.51271/SOC-0046

An approach to identify high-risk neonatal surgical cases to optimize postoperative results

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Received: 24/02/2025 ◆ **Accepted**: 09/03/2025 ◆ **Published**: 01/07/2025

Cite this article: Purbey OP, Rahul SK, Keshri R, Chaubey D. An approach to identify high-risk neonatal surgical cases to optimize postoperative results. *Surg Child.* 2025;2(3):109-113.

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ABSTRACT

Postoperative recovery in pediatric surgery can vary greatly, even among patients with identical anomalies. While some cases meet expectations, others may result in disappointing outcomes. To underscore the importance of identifying high-risk surgical neonates early and understanding the critical factors influencing postoperative results, a thorough literature review was undertaken to pinpoint the key elements contributing to poor outcomes in surgical neonates. It is important to recognize that each surgical neonate requires a unique approach to management, influenced by factors such as family background, preoperative condition, and the involvement of a multidisciplinary team. The elevated mortality rates observed in emergencies can be attributed to various factors, including delays in hospital arrival, the timing of surgical intervention, and the operating surgeon's level of expertise. Ultimately, the early identification of high-risk surgical neonates is essential for improving clinical outcomes. A comprehensive strategy that incorporates individualized management and fosters collaboration among multidisciplinary teams is crucial for achieving the best possible postoperative recovery.

Keywords: Determinants, emergencies, mortality, patient care, postoperative care, surgical neonates

INTRODUCTION

A surgical neonate is defined as a child who is either (i) born at >37 weeks of gestation (term neonate) and younger than 29 days at the time of surgery or (ii) born at <37 weeks of gestation (preterm neonate) and younger than 50 weeks postconception at the time of surgery.[1] The World Health Organization estimates that approximately 240.000 newborns die annually within the first 28 days of life because of congenital disorders, with 90% of affected children residing in low- and middle-income countries.[2] In developed nations, the primary contributors to mortality among surgical neonates include irreparable abnormalities of the heart, lungs, kidneys, and central nervous system; metabolic disorders; extreme prematurity; and low birth weight.[3]

Despite advances in neonatal surgery, high-risk surgical neonates continue to experience significant morbidity and mortality. Several critical gaps in knowledge must be addressed to improve clinical outcomes. First, standardized criteria are needed to identify high-risk surgical neonates, because the current lack of standardization hinders timely intervention and effective management. Second, the optimal timing of surgery for different surgical conditions in neonates remains unclear, highlighting the need for further research in this area. Third, the effectiveness of multidisciplinary team approaches in improving outcomes in high-risk surgical neonates requires further investigation. Furthermore, studies on the role of prenatal factors, such as maternal health and fetal monitoring, in predicting postoperative outcomes in surgical neonates are limited. Moreover, there is a significant knowledge gap in low- and middle-income countries, where the challenges and opportunities for improving outcomes in high-risk surgical neonates are unclear. Other critical knowledge gaps include the impact of hospital volume and surgeon experience on outcomes and the potential benefits and challenges of using technology, such as telemedicine, to improve outcomes for high-risk surgical neonates.

Although the overall infant mortality rate has decreased over time, birth defects, which affect an estimated 3–5% of pregnancies, continue to be the leading cause of infant mortality in the United States.[4] Nearly 10% of all neonatal fatalities are attributed to congenital malformations that necessitate surgical intervention.[5] Survival rates vary according to the type of defect, owing to differences in functional impact and organ systems involved. Reported neonatal surgical mortality rates exhibit geographical

disparities, with figures of 4% in the United States, 6–7% in Japan, 35–45% in India, 52.7% in Uganda, and 62.2% in Nigeria.[1,6-9] The margin of error in neonatal surgery is very low, as evidenced by a significant global variation in neonatal surgical mortality rates, which range between 4% and 80%.[10] This highlights the need for early recognition of high-risk surgical neonates to improve clinical outcomes. Furthermore, studies have shown that delayed recognition of high-risk neonates can lead to increased morbidity and mortality.[11] Therefore, it is essential to identify high-risk surgical neonates early to provide timely and appropriate interventions. Developing predictive models and scoring systems can aid in the early recognition of high-risk surgical neonates. However, further studies are required to improve the accuracy and reliability of these models (**Table 1**).

Table 1. Knowledge gaps

1. Lack of standardized criteria for identifying high-risk surgical neonates.
2. Limited understanding of the impact of surgical timing on outcomes.
3. Insufficient data on the effectiveness of multidisciplinary team approaches.
4. Limited studies on the role of prenatal factors in predicting postoperative outcomes.
5. Knowledge gap in low- and middle-income countries.
6. Limited understanding of the impact of hospital volume and surgeon experience on outcomes.
7. Insufficient data on the role of technology such as telemedicine in improving outcomes.

REVIEW

Familial Factors

Familial factors, particularly those associated with socioeconomic conditions, have been correlated with reduced access to pediatric subspecialty care and increased early mortality rates among neonates with low birth weight.[12] These factors include the family's overall literacy, economic status, preference for institutional or home deliveries, planned or unplanned pregnancies, consanguineous marriages, hereditary diseases, and maternal age. Family assessments can provide valuable insights into these factors. Families with low literacy levels and limited socioeconomic resources may lack awareness of critical health issues, including family planning, personal hygiene, the advantages of planned pregnancies, dietary adjustments during pregnancy, and the importance of regular antenatal visits. Consequently, surgical neonates from these families have lower survival rates.[13-18] One review indicated that maternal alcohol consumption is associated with an increased risk of conditions such as d-transposition of the great arteries, neural tube defects, and multiple cleft palates in infants.[19] Additionally, maternal smoking, whether before or during pregnancy, significantly increases the risk of various congenital anomalies, even with minimal exposure to 1–5 cigarettes daily.[20] Congenital anomalies linked to chromosomal defects may be more prevalent in populations where women have children after the age of 35 years and in pregnancies resulting from consanguineous unions.[21,22] Furthermore, studies have shown that maternal nutrition and micronutrient deficiencies can affect fetal development and increase the risk of congenital anomalies.[23,24] Additionally, exposure to environmental pollutants and toxins during pregnancy is associated with an increased risk of birth defects.[25,26] High-risk pregnancies and a lack of awareness of health and health policies can transform a normal newborn into a high-risk one. Therefore, the fundamentals of health and awareness should be extended to remote populations. This effort can yield some success in decreasing infant mortality in the neonatal period due to surgery for anatomical defects by facilitating either early arrival at a well-equipped center or planned delivery at a tertiary care center.

Administrative Factors

Multiple factors beyond familial considerations, including early diagnosis, prompt initiation of supportive treatment, and an effective referral policy, can contribute to successful outcomes. A robust referral system is a vital element of the healthcare framework and is essential for enhancing outcomes for mothers and newborns during childbirth.[27] Reducing maternal and neonatal mortality and morbidity rates can be achieved through prompt recognition of conditions requiring specialized treatment, which may not be available at every medical facility, coupled with an effective referral process.[28,29] **Table 2** classifies patients according to their referral urgency, while **Figure 1** outlines the suggested surgical referral protocol for critically ill newborns in need of surgical intervention.

Table 2. Category of referral with examples

1. Acute surgical referrals	Postnatally diagnosed congenital malformations like Oesophageal atresia +/- trachea-oesophageal fistula-congenital diaphragmatic hernia Suspected Hirschsprung's disease Anorectal malformations Abdominal wall defects (gastroschisis and exomphalos) sacrococcygeal teratoma Gross abdominal distension and/or bowel obstruction including duodenal, jejunoileal and colonic atresia; meconium ileus and malrotation+/- volvulus Bowel perforation associated with necrotising enterocolitis or spontaneous intestinal perforation Other visceral injury/perforation (oesophageal/gastric) Severe NEC unresponsive to medical management Testicular torsion Incarcerated inguinal hernia Emergency requirement for surgical central venous access Swallowed foreign body Pneumothorax/empyema
2. Elective surgical referrals	Inguinal hernia repair Stoma closure Removal of a tunnelled central venous catheter Vascular malformation in stable patients Tumors in stable patients Congenital hypertrophic pyloric stenosis Ruptured meningo-myelocoele Rachischisis Biliary atresia Choledochal malformation and other surgical cholestatic conditions Abscesses Posterior urethral valves/ ureteropelvic junction obstruction Bladder exstrophy
3. Outpatient transfers	Undescended testes Umbilical granuloma Ranula Haemangioma/lymphangioma Cleft lip/palate Hydrocephalus Other neural tube defects Hydrocephalus Pectus excavatum Umbilical hernia

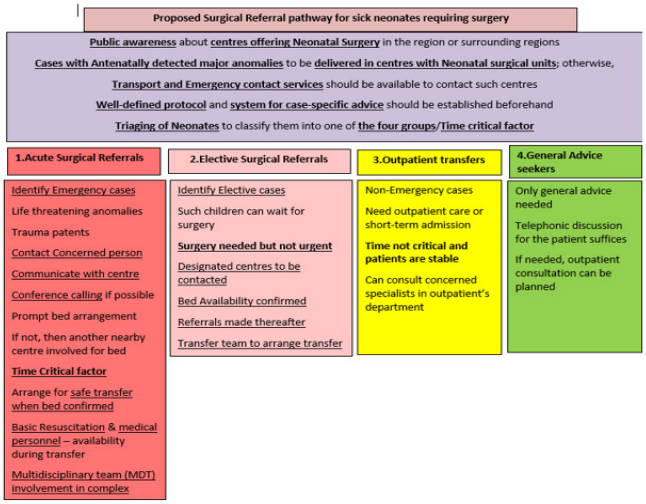

Figure 1. Proposed surgical referral pathway for sick neonates requiring surgery

The prognosis of surgical neonates may be adversely affected by uncoordinated arrival at tertiary care centers. Infants with congenital anomalies require continuous monitoring and multiple surgical procedures. The transportation of surgical neonates presents a considerable challenge that can influence their survival rates. Insufficient transportation resources can result in treatment delays, increasing the risk of morbidity and mortality.[30] Furthermore, families may face significant financial burdens that affect their economic well-being and morale. In low-income nations lacking comprehensive government health policies, delays in pursuing surgical interventions or the decision to forgo surgery can result in unfavourable outcomes. A comprehensive policy package for managing small and sick newborns underscores the importance of specialized care policies during the antepartum and immediate postpartum periods to ensure neonatal survival.[31] Studies have shown that the availability of pediatric surgical services, including emergency surgical care, can significantly affect neonatal mortality rates.[32-35] The development of pediatric surgery as a speciality has been associated with improved outcomes in neonates requiring surgery.[36] Moreover, the implementation of quality improvement initiatives, such as the American College of Surgeons National Surgical Quality Improvement Program Pediatric, can help reduce morbidity and mortality rates in surgical neonates.[37]

Patient-Related Factors

Patient-related factors were term or preterm delivery, single or twin pregnancy, birth weight, isolated anomalies or syndromic associations, timing and location of diagnosis, and general condition of the neonate at diagnosis.

Successful postoperative recovery of surgical neonates primarily depends on patient-specific factors, with other previously mentioned factors aiding in thorough assessment and preparation for surgery. Neonatal surgery requires a profound understanding of neonatal physiology/anatomy, advanced surgical expertise, and comprehensive neonatal care before and after the operation.[38] A preliminary study by Puri et al.[39] in 2019 highlighted numerous variables that must be considered when dealing with surgical neonates, including preoperative, intraoperative, and postoperative variables. Based on these variables, a severity score was generated (normal values were allocated a score of 0, and a score of 1 or 2 was allocated values moving towards abnormal) to categorize the surgical neonate as low-, moderate-, or high-risk. These variables include 20 parameters, as summarized in **Table 3**.

Table 3. Numerical grading of preoperative, intraoperative and postoperative variables

Grade	0	1	2
Preoperative variables			
Admission weight (kg)	≥3	2-3	≤2
Place of delivery (level)	III	II or III	Home
Gestational age (weeks)	≥36	32-36	≤32
Delay in seeking treatment (h)	<24	24-48	>48
Associated anomalies	None	Minor	Major
Illness severity score (ISS)	0-4	5-8	9-12
Temperature (°C)	34-36	32-34	<32
Heart rate (per minute)	140-160	160-180	<100 or >180
Respiratory rate (per minute)	30-40	40-60	>60
Urine output (mL/kg/h)	≥2	1-2	<1
Platelet counts (cells/μL)	>100.000	60.000-100.000	<60.000
Blood pH	7.3-7.45	7.2-7.3	<7.2
Intraoperative variables			
Site of surgery	Superficial	Intraabdominal	Intrathoracic
Duration of surgery (min)	<60	60-120	>120
Amount of blood loss (% blood volume)	<5	5-10	10-15
Temperature after surgery (°C)	34-36	32-34	<32
Postoperative variables			
Extubation after surgery (h)	On table	<24	≥24, reintubation
Need for vasopressor	No	D1 or D2*	D1+D2±A**
Infection	Nil	Local	Systemic
Complications	Nil	Minor	Major#
Time till resuming enteral feeding (days)	<7	7-14	>14

D1: Dopamine, D2: Dobutamine, *Dose of D1 or D2-@5 mcg/kg/min, A: Adreline (dose of 0.05-0.3 mcg/kg/min), ** Dose of D1 and D2 @ >10 mcg/kg/min, # major: Complication requiring reoperations

In their study, numerically graded scores were assigned to preoperative (0–12), intraoperative (0–8), and postoperative variables (0–10).

However, the study highlighted that preoperative variables did not significantly affect the postoperative outcome; however, intraoperative and postoperative variables were major determinants of survival.

Newborns born prematurely have an increased risk of developing sepsis before surgery, as reported by Mpody et al.[40] They highlighted the synergistic effects of sepsis and premature birth on postoperative neonatal mortality. Additionally, they found that premature newborns with preoperative sepsis have a significantly higher risk of death and complications after surgery. Hasan et al.[41] investigated neonatal surgical morbidity and mortality in a tertiary

center in low-and middle-income countries. They found that emergency surgery was a major cause of death in newborns, with approximately 67.6% of newborns who died after surgery having undergone emergency operations. Numerous other factors, such as the surgeon's and anesthetist's experience, timing of surgery (same-day vs. delayed), and timely involvement of a multidisciplinary team are also significant determinants of satisfactory postoperative outcomes, in addition to those listed by Puri et al.[39] Surgical stress to the neonate undergoing surgery varies considerably with the type of surgery, its duration, whether performed by open or minimally invasive techniques, whether done in an emergency or electively, whether adequate anaesthesia and pain control was taken care of and related to various patient-related factors like age, body weight, overall health, and psychological state. The more complex the surgery, the more profound its stress level. For example, while major cardiac surgery alters physiology due to its complexity, major abdominal surgery with prolonged exposure of the bowel leads to dehydration and fluid imbalances which need to be taken care of. An open abdominal/thoracic surgery is more stressful compared to the same surgery performed via laparoscopy or thoracoscopy. Similarly, elective surgeries done in stable and optimized neonates lead to less surgical stress compared to emergency surgeries in sick children. The longer the duration of surgery, the more stress it induces.

According to the 2020 Consensus guidelines for perioperative care in neonatal intestinal surgery: enhanced recovery after surgery society recommendations, a standardized perioperative team communication system with a structured process and protocol can lower the risk of negative patient outcomes, ensure continuity of care, and enhance communication among staff.[43]

Caring for a newborn undergoing surgery is always a team effort that includes neonatal surgeons, neonatologists, and neonatal anesthesiologists. By working together, a team can provide a higher level of care, thereby improving the overall survival rate. **Figure 2** provides a comprehensive overview of the factors to consider when managing a surgical neonate in an emergency setting. This enables the identification of high-risk cases, thereby facilitating the implementation of preventive measures to avert complications.

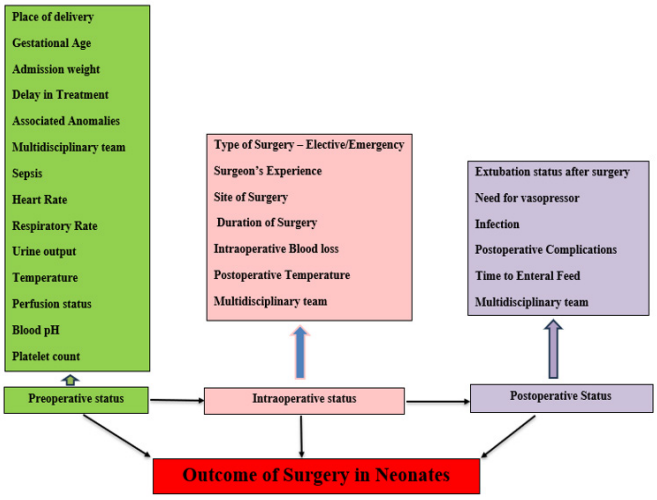

Figure 2. How to approach a surgical neonate

CONCLUSION

Timely evaluation and intervention are crucial for improving postoperative outcomes in neonates undergoing surgery. The high mortality rate in emergency cases can be attributed to various factors, including delayed hospital arrival, surgical timing, and expertise of the operating surgeon and anaesthetist. A comprehensive approach that considers the unique needs of each premature newborn is essential for achieving favourable postoperative outcomes. The growing focus on reducing infant mortality rates worldwide underscores the importance of addressing the mortality rates of surgical neonates. Early identification of high-risk surgical neonates, particularly those with multiple congenital anomalies and other risk factors, is critical for improving their chances of survival. A multidisciplinary approach incorporating the expertise of neonatologists, pediatric surgeons, and anesthesiologists is essential for providing specialized care to these vulnerable patients.

Future Scope

Future studies should focus on developing and implementing effective strategies for the early identification and management of high-risk surgical neonates. This may include the development of predictive models and scoring systems to identify neonates with the highest risk of poor outcomes. Additionally, studies examining the impact of standardized care protocols and multidisciplinary team approaches on the outcomes of neonates undergoing surgery are warranted. Furthermore, efforts to improve access to specialized care for surgical neonates in low- and middle-income countries are critical for reducing global disparities in infant mortality rates.

ETHICAL DECLARATIONS

Referee Evaluation Process

Externally peer-reviewed.

Conflict of Interest Statement

The authors have no conflicts of interest to declare.

Financial Disclosure

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

Author Contributions

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

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MHA MEDIHEALTH ACADEMY

Surgery on Children

Case Report

doi: 10.51271/SOC-0047

Nutritional and dietary approach in the post-surgical follow-up of Hirschsprung disease: a case report

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Received: 13/11/2024 ◆ **Accepted**: 27/12/2024 ◆ **Published**: 01/07/2025

Cite this article: Taşdan K. Nutritional and dietary approach in the post-surgical follow-up of Hirschsprung disease: a case report. *Surg Child.* 2025;2(3): 114-117.

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ABSTRACT

Hirschsprung disease (HD) is a congenital condition caused by abnormal development of the enteric nervous system during the embryonic period, characterized by colonic aganglionosis. Total colonic aganglionosis (TCA) refers to the involvement of the entire colon extending to the terminal ileum, accounting for 3–12% of all HD cases. A 45-month-old male patient presented to the clinic with complaints of frequent vomiting and more than 10 watery diarrhea episodes per day. The patient's medical history revealed the development of short bowel syndrome (SBS) following TCA. Physical examination indicated a poor general condition, decreased skin turgor and tonus, along with growth and developmental delay. Laboratory findings demonstrated low sodium (Na) and potassium (K) levels with electrolyte imbalances. A low-carbohydrate, high-protein diet was planned alongside hydration therapy. Additionally, the family was educated on nutritional management and avoiding harmful foods. Following 17 days of treatment and dietary interventions, the patient's weight increased by 1.5 kg, and complaints of diarrhea and vomiting subsided. During a two-year follow-up period, a reduction in hospitalization frequency and improvements in the patient's overall clinical condition were observed. The individualized treatment and nutritional plan significantly enhanced the patient's quality of life. The management of SBS, which may develop after HD surgery, requires individualized nutritional plans, a multidisciplinary approach, and continuity in patient-family education, all of which are critically important.

Keywords: Hirschsprung disease, pediatric surgery, nutrition and diet, short bowel syndrome

INTRODUCTION

Hirschsprung disease (HD) is a congenital condition caused by abnormal development of the enteric nervous system during embryonic development, characterized by colonic aganglionosis.[1] It occurs approximately in 1 out of every 5000 live births and is one of the most common congenital disorders of the lower gastrointestinal tract.[2] Children with HD typically present with symptoms related to bowel obstruction, such as delayed passage of meconium, abdominal distension, and bilious vomiting within the first 24-48 hours of life. Diagnosis is usually made within the first six months of life. The congenital defect underlying HD involves the complete absence of ganglion cells in the most distal part of the gastrointestinal system.[3] Due to this aganglionosis, there is a lack of peristaltic movements and the inability of the smooth muscles to relax, leading to functional narrowing of the affected bowel segment. Approximately 80% of patients have aganglionic segments localized to the rectosigmoid region; however, in rare cases, the condition may involve the entire colon or even a portion of the small intestine.[2] The involvement of the entire colon extending to the terminal ileum is referred to as total colonic aganglionosis (TCA), accounting for 3–12% of all HD cases.[4] Postoperative complications frequently observed in patients with TCA include stoma prolapse, chronic constipation, fecal incontinence, anastomotic leakage, and Hirschsprung-associated enterocolitis (HAEC).[5,6] Furthermore, the development of short bowel syndrome (SBS) is a potential postoperative concern in cases of TCA.[7]

SBS is a condition characterized by malabsorption resulting from the resection of a portion of the small intestine or due to congenital malformations.[8,9] The degree of malabsorption varies depending on the extent of resection or the remaining intestinal segment.[9] Shortened bowel length leads to accelerated intestinal transit and insufficient absorption of nutrients.[10] Factors such as gastric acid hypersecretion, bacterial overgrowth in the intestine, impaired absorption of fats and bile salts, and reduced fluid absorption collectively contribute to the development of high-osmolarity diarrhea.[7,11] In addition to refractory diarrhea, symptoms such as steatorrhea, weight loss, dehydration, and electrolyte imbalances are commonly observed.[11,12] Managing levels

of magnesium, calcium, and potassium can be particularly challenging in individuals with SBS.[11] Furthermore, these patients often experience nutritional deficiencies and growth and developmental delays.[4]

In the postoperative recovery period of SBS, parenteral nutrition therapy should be initiated to maintain fluid and electrolyte balance and optimize carbohydrate, protein, and fat intake. Following a period of adaptation after resection, moderate enteral nutrition should be introduced.[13] During this phase, semi-elemental or elemental formulas may be utilized. Subsequently, the transition to age-appropriate solid foods with a low-carbohydrate and high-protein composition is recommended.[14]

Surgical intervention forms the cornerstone of treatment for HD; however, postoperative follow-up and care are crucial for the patient's long-term health and quality of life.[1] Issues such as bowel dysfunction and surgical complications that affect quality of life necessitate long-term monitoring. A multidisciplinary approach and individualized treatment plans are essential for improving the quality of life in these patients.[4] This article discusses a case aimed at evaluating the nutritional and dietary approach in the postoperative period of a patient operated on for HD.

CASE

A 45-month-old male patient presented to the pediatric surgery outpatient clinic with complaints of vomiting and more than 10 episodes of profuse diarrhea daily. The patient's medical history revealed an absence of meconium passage within the first 24–48 hours during the neonatal period. Following a preliminary diagnosis of HD based on findings from a colon radiograph and biopsy, a colostomy was performed. During surgery, frozen biopsy results identified TCA. Subsequently, an ileoanal pouch was constructed using the Duhamel–Martin procedure, and the patient was monitored with an ileostomy for six months before its closure. It was noted that the patient continued to experience frequent enterocolitis episodes, leading to SBS symptoms characterized by impaired nutrient absorption due to persistent diarrhea.

Due to persistent diarrhea and SBS, the patient exhibited growth and developmental delays, weight loss, and impaired absorption of vitamins, sodium, and potassium. On physical examination at the time of admission to our clinic, the patient was in poor general condition, with decreased skin turgor and tonus. Abdominal examination revealed old scar tissue from previous surgeries, and bowel sounds were hyperactive. On palpation, no tenderness, guarding, or rebound was noted. Laboratory findings showed decreased sodium (Na) and potassium (K+) levels, while the rotavirus and adenovirus antigens were negative (Na: 135 mEq/L, K+: 3.62 mEq/L, Cl: 96.5 mEq/L, Mg: ALT: 65.5 U/L, AST: 50.1 U/L, CRP: 56.09 mg/dl, albumin: 47.8 g/L, Hgb: 14.0 g/dl).

Dietary habits revealed frequent and excessive consumption of foods such as tea, biscuits, chips, olives, raw meatballs, and raw minced meat. Anthropometric measurements indicated a body weight of 11.5 kg (3rd percentile) and a height of 90.8 cm (3rd percentile).

The patient's treatment plan included intravenous (IV) hydration, metronidazole, ondansetron, and pantoprazole. Due to insufficient oral intake, a nasogastric (NG) tube was inserted, and nutritional support was initiated with an isocaloric enteral formula (**Table 1**).

Table 1. Nutritional plan

Days 1-2:	1300 kcal/24 h (enteral product 1.0 kcal).
Days 3-6:	1300 kcal/24 h enteral product+continued oral intake (small amounts of water/light tea, table salt intake).
Day 7:	960 kcal/24 h enteral product+supported with oral intake (soup, meatballs).
Days 8-10:	720 kcal/24 h enteral product+improved oral intake but still insufficient based on hospitalization.
Days 11-12:	540 kcal/24 h enteral product+increased oral intake (able to consume 2 eggs, meatballs, and ½ bowl of soup comfortably).
Day 13:	270 kcal/24 h enteral product.
Days 14-17:	Various enteral products were trialed (strawberry, vanilla, and unflavored). Oral intake was good.

During the initial days of hospitalization, the patient experienced at least 10 episodes of watery, voluminous diarrhea and 4 episodes of vomiting daily. By the second day of admission, the patient's general condition began to improve, vomiting subsided, and the frequency and consistency of diarrhea improved progressively, decreasing to 4–6 semi-formed stools per day. With the implementation of an adjusted oral feeding and nutrition plan, the patient showed improvement in blood values, oral intake, clinical findings, and physical activity levels. Over the 17-day hospitalization, the patient gained 1.5 kg in body weight and was subsequently discharged.

Approximately 2 months after discharge, the patient presented to the clinic again with more severe complaints of diarrhea, nausea, and vomiting, and was readmitted. A significant decrease in sodium levels (Na: 121 mEq/L) was observed, and IV hydration along with NG tube-enteral feeding therapy was initiated. Alongside enteral nutrition, a healthy eating plan was developed and implemented. Upon assessing the patient's daily diet, it was determined that the patient paid little attention to their nutrition, and there was poor adherence to dietary recommendations. The family's socio-economic level contributed to a high level of non-compliance, as they permitted the consumption of unprocessed foods like raw minced meat and encouraged direct consumption of salt, potentially affecting sodium levels. Dietitian collaboration was reinforced with the family, offering nutritional and dietary education both face-to-face and through communication tools (e.g., phone), with frequent monitoring intervals.

Over the course of approximately 2 years, the patient visited the clinic 14 times, with 7 admissions spaced two months apart due to poor clinical condition and low sodium and potassium levels. It was observed that admission durations decreased progressively with each visit (**Figure 1**). At the

most recent visit, the patient's sodium level was 123 mEq/L and potassium level was 3.02 mEq/L. Although the patient's overall condition remained poor, there was a noticeable increase in growth and weight gain, with body weight reaching 18 kg. Additionally, the patient began to comfortably consume previously avoided foods (e.g., eggs). With the latest follow-ups, the foods the patient desired to consume were prioritized, and a personalized nutrition plan was redesigned to maintain fluid-electrolyte balance and prevent diarrhea (Table 2).

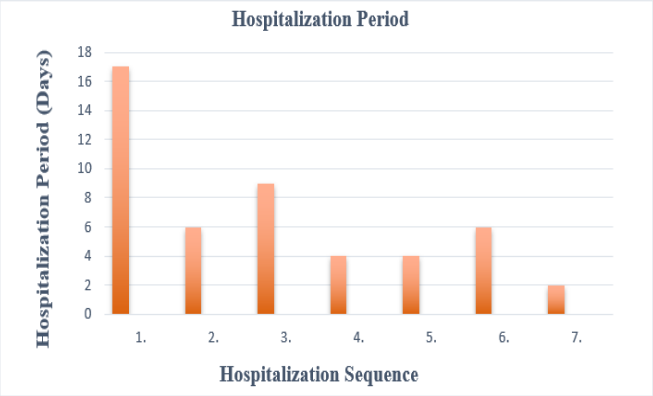

Figure 1. Graph of patient's hospitalization duration

Tablo 2. Sample nutritional plan

Morning
1 cup of herbal tea (linden tea)
1 boiled egg (50-60 g)
1 slice of white cheese (40 g)
4-5 olives
2 thin slices of white bread (50 g)
Mid-morning snack
½ carton of enteral product/plus fiber (1 ml=1.5 kcal) (100 ml)
Lunch
½ bowl of soup (100 ml-½ ladle)
2 tablespoons of yogurt or tzatziki
1 small banana/apple/peach
2 thin slices of white bread (50 g)
Afternoon snack
½ carton of enteral product/plus fiber (1 ml=1.5 kcal) (100 ml)
Dinner
2 portion of meat/chicken/ground beef (60 g) (e.g., boiled chicken, meatball, homemade hamburger)
4 tablespoons of vegetable dish
3 tablespoons of rice pilaf
1 bowl of tzatziki (made with 4 tablespoons of yogurt)
Night snack (2-3 hours before bed)
1 carton of enteral product/plus fiber (1 ml=1.5 kcal) (200 ml)

Upon analyzing the patient's anthropometric measurements, initial admission (at 45 months) showed a body weight of 11.0 kg (3rd percentile) and height of 90.8 cm (3rd percentile). At the latest admission (at 66 months), the body weight had increased to 18.0 kg (25-50th percentile), and height was 106 cm (10-15th percentile) (Figure 2).

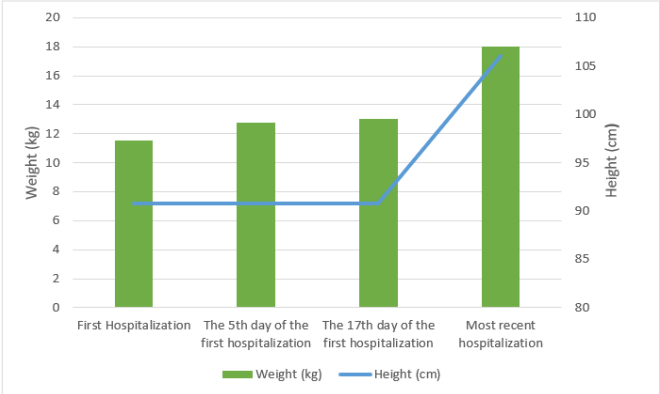

Figure 2. Graph of patient's body weight and height

DISCUSSION

This study aims to comprehensively examine the nutrition and diet approach in cases of TCA secondary to Hirschsprung's disease and associated SBS. TCA is a rare form of Hirschsprung's disease that leads to significant clinical and nutritional challenges early in life.[4] In the literature, TCA cases are frequently complicated by conditions such as enterocolitis, malnutrition, growth retardation, and electrolyte imbalances.[15] This highlights the need for a multidisciplinary approach beyond surgical management, involving close monitoring by pediatric surgeons, gastroenterologists, pediatricians, dietitians, psychologists, and nurses. In cases of Hirschsprung's disease with total colectomy and ileostomy, the importance of medical treatment, as well as care and nutritional support, is emphasized. Multidisciplinary collaboration in such cases is crucial in reducing hospital stays and minimizing the need for additional surgeries.[4]

In Hirschsprung's disease, surgical treatment methods such as Soave, Swenson, and Duhamel-Martin procedures are commonly used, each with distinct advantages and associated risks of complications.[16] The Duhamel-Martin procedure, in particular, has been associated with frequent postoperative complications such as enterocolitis, constipation, and vomiting, as reported in the literature.[17] In our case, similar findings were observed, with postoperative enterocolitis attacks and a progressive course of SBS being documented.

The outcomes of SBS include malabsorption, diarrhea, dehydration, electrolyte imbalances, nutrient deficiencies, and inadequate weight gain or loss, necessitating parental support along with both enteral and solid nutritional support.[8,18] Non-cellulose forms of fiber in grains contribute to an increase in stool volume, while pectin and guar found in fruits and vegetables provide a diarrhea-preventive effect by slowing intestinal transit time.[18] In our case, efforts were made to include these foods in the individualized nutrition plan.

A significant issue in this case was the family's lack of adherence to nutritional treatment, exacerbated by a low socio-economic level, which contributed to this noncompliance. This situation negatively affected the patient's clinical course. Continuous support through a structured follow-up mechanism and collaboration with dietitians helped implement an individualized nutrition plan. This approach is believed to have played a significant role in the patient's progress in growth and development. Literature supports that a multidisciplinary approach positively influences the health outcomes of individuals.[19]

CONCLUSION

As a result, the management of SBS following HD surgery requires individualized nutrition plans, a multidisciplinary approach, and continuous patient-family education. Maintaining direct contact with the patient and family, utilizing communication channels, and ensuring timely hospital admissions when necessary are essential. Early evaluation of blood electrolyte levels and appropriate dietary interventions are crucial for facilitating a smoother recovery and improving the patient's quality of life during this period.

ETHICAL DECLARATIONS

Informed Consent

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

Referee Evaluation Process

Externally peer-reviewed.

Conflict of Interest Statement

The authors have no conflicts of interest to declare.

Financial Disclosure

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

Author Contributions

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

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MHA MEDIHEALTH ACADEMY

Surgery on Children

Case Report

doi:10.51271/SOC-0048

Para meatal urethral cyst: a rare condition

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Received: 07/01/2025 ◆ **Accepted**: 08/02/2025 ◆ **Published**: 01/07/2025

Cite this article: Gupta A, Bose V, Singh AK. Para meatal urethral cyst: a rare condition. *Surg Child*. 2025;2(3):118-119.

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ABSTRACT

There are very few documented occurrences of parameatal urethral cyst in the literature, making it an extremely uncommon benign disorder. These cysts may go away on their own and are typically asymptomatic. For patients who are exhibiting symptoms, surgical removal is required. Here, we describe the effective management of a child's parameatal urethral cyst with careful excision.

Keywords: Parameatal cyst, splaying, smegma pearl

CASE

A six-year-old child was referred from the pediatrics outdoor department for a cystic-like lesion on the urethral meatus tip. Upon local assessment, he exhibited a spherical cystic mass measuring 75x75 mm^2 near the lateral edge of the external urethral meatus (**Figure 1**). The cyst appeared to be filled with a serous to somewhat milky fluid, which made it easy to identify from a smegma pearl. The cystic mass was partially covered by preputial skin, which was incapable of being separated even with mild tearing. The penis appeared typical and was not hypospadias. His urinalysis showed no evidence of infection, and his urine stream was good with no splaying. Physical examination results were within normal limits. The kidneys and bladder appeared normal on an abdominal ultrasound. Excision of the cyst with meatoplasty was performed under general anaesthesia (**Figure 2**) and the tissue was sent for a histopathological examination. Histopathological examination showed (**Figure 3**) a cyst lined by stratified columnar cells and sub-epithelium had fibro collagenous tissues infiltrated with chronic inflammatory cells. The post-operative period was uneventful. He has been in follow-up for the last one year with no sign of recurrence.

Figure 1. Parameatal urethral cyst

Figure 2. Excised cyst

DISCUSSION

The parameatal urethral cyst is an uncommon disorder. Thompson and Lantin[2] in 1956 first reported the case of parameatal urethral cyst. Currently, the number of cases described in the literature is less than 100.[1,2] The majority of parameatal urethral cysts are asymptomatic. The cysts may form at any point in childhood or adulthood, or they

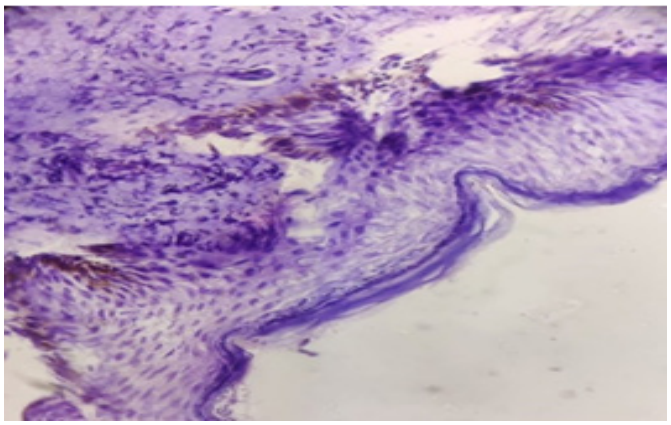

Figure 3. Histopathological examination

may be present from birth[3], and they can occur in females[4] and males, despite the fact that they often appear in males throughout their 1st year of life. The parameatal cyst's etiology is not known. One probable explanation is that cystic spaces continue to exist throughout preputial delamination[2], development of cysts due to abnormal urethral fusion[1], or blockage of the paraurethral duct.[5] Patients exhibit symptoms such as urine splaying, difficulties in voiding, and dysuria.[1,4] Physical examinations are sufficient to diagnose the majority of parameatal urethral cysts. Between 91.3% and 100% of cysts have a diameter of less than 1 cm[1,4] and are situated either lateral or ventral to the meatus.[6] It's critical to distinguish these cysts from skin tags, dermoid cysts, epidermal inclusion cysts, and cysts that are not found in the urethral meatus.[4] The uncommon nature of parameatal urethral cysts means that there are no established treatment standards. In 25% of cases, a parameatal urethral cyst resolves on its own.[1,4] Oka et al.[7] explained parameatal urethral cyst's spontaneous resolution in a neonate at four months. Surgical excision should be taken into consideration when cysts present symptoms. Surgical excision is an additional option for those whose cysts do not go away on their own after 24 months or who are worried about their appearance.[4] After these cysts are surgically completely removed, there have been no instances of recurrence. However, simple aspiration or marsupialization of the cyst should be avoided as it may result in a recurrence or inadequate cosmesis.[8] We removed the cyst with good cosmesis in our case report.

CONCLUSION

Infants and newborns tend to have spontaneous clearance of parametal urethral cysts. Therefore, conservative care with routine follow-up is indicated; however, total surgical excision is the preferred course of treatment for symptomatic individuals seeking cosmesis.

ETHICAL DECLARATIONS

Informed Consent

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

Referee Evaluation Process

Externally peer-reviewed.

Conflict of Interest Statement

The authors have no conflicts of interest to declare.

Financial Disclosure

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

Author Contributions

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

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