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Original Article

Is there a role for caudal anesthesia on postoperative urethrocutaneous fistula in children undergoing hypospadias surgery?

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ABSTRACT

Objective: Hypospadias is defined as incomplete closure of the penile structures during embryogenesis. Surgical correction induces various complications, including urethrocutaneous fistula (UCF). The aim of this study was to determine the risk factors for the occurrence of UCF in children undergoing hypospadias repair under caudal anesthesia.

Method: The medical records of children undergoing hypospadias repair between January 2013 and July 2018 were included. Data on patients' age, body weight, height, type of repair procedure, type of hypospadias, duration of surgery, and hospitalization, and postoperative complications were analyzed.

Results: The mean age of the 122 patients was 4.8±3.7 years. The type of surgery performed was tubularized incised plate urethroplasty (Snodgrass) in 90 (73.8%) and meatal advancement and glanuloplasty (MAGPI) in 32 (26.2%) children. Sixteen (13.1%) children had postoperative complications, all of which were UCF. No statistical association was found between postoperative UCF and patient variables.

The most common complication of hypospadias repair is UCF, which occurs mostly in the immediate postoperative period.

Conclusion: Hypospadias repair can result in complications. UCF remains a significant problem in the postoperative period. All patients underwent caudal block and despite the previous literature, we experienced lower rates of penile engorgement and postoperative UCF. These results showed that there was no cause-and-effect relationship between the caudal block and UCF. We think that the development of a urethrocutaneous fistula is mostly related to surgical causes and well-designed, prospective, and controlled studies are required to elucidate this issue.

Keywords: Caudal block, hypospadias, penile engorgement, tissue edema, urethrocutaneous fistula

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INTRODUCTION

Hypospadias is defined as a urethral opening along the ventral side of the penis, resulting from incomplete closure of the penile structures during embryogenesis. It is one of the most common pediatric urological conditions, with a multifactorial etiology and a prevalence of approximately 18.6 per 10,000 births.[1] Hypospadias varies considerably in terms of presentation and severity. The aim of repair is to normalize functions and cosmetic appearance. The postoperative complication rate for distal hypospadias has been reported to be approximately 10%; however, this rate may be higher when repairing proximal defects.[2] The incidence of postoperative urethrocutaneous fistula (UCF) has been reported to range from 4% to 28%.[3] Previous studies have mostly focused on the severity of the congenital defect (the location of the urethral meatus) or the type of surgical technique used in the analysis of operative success.[4]

Caudal block (CB) is one of the most popular types of regional anesthesia used as an adjunct to general anesthesia in children undergoing inguinal or genital surgery. It provides reliable postoperative analgesia in children, with a reasonable safety profile and few complications. It has been suggested that caudal anesthesia increases the risk of surgical complications.[5] It is therefore an ongoing debate in pediatric anesthesia whether CB should be abandoned for hypospadias correction. The repair of hypospadias helps to correct both cosmetic and functional abnormalities. Spraying of the urinary stream, inability to urinate in a standing position, potential difficulties in sexual intercourse and fertility issues, and decreased satisfaction with genital appearance can be corrected surgically.[6] However, this highly technical surgery can result in significant complications, such as meatal stenosis, urethrocutaneous fistula, and urethral stricture. Although UCF is common after hypospadias repair, pre- and intraoperative risk factors for its development are not yet well-known. Whether CB increases the risk of UCF development has been a recent topic of discussion. The aim of this study was to determine the risk factors for UCF in children undergoing hypospadias repair under caudal anesthesia.

MATERIAL AND METHODS

After obtaining the approval of the Local Ethics Committee (Protocol# 2018/1456), we conducted a retrospective review of the medical records of children who had undergone a hypospadias repair between January 2013 and July 2018 in Aydın Adnan Menderes University, Medical Faculty, Training and Research Hospital. Only the cases that had undergone surgery more than six months previously were included in the study, since fistula formation may be delayed for up to six months. Patients with incomplete medical records, patients who had undergone a multi-step hypospadias repair or a revision surgery, patients with disorders of sexual development, connective tissue and bleeding disorders, and patients with a follow-up period of less than six months were excluded from the study.

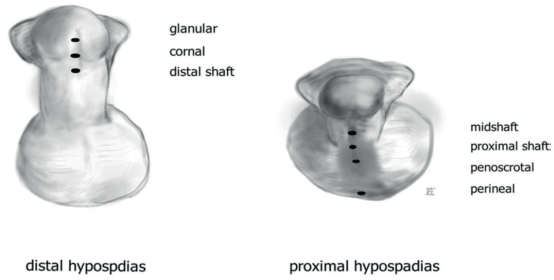

Figure 1. The position of the urethral meatus in hypospadias [1]

Figure 1. The position of the urethal meatus in hypospadias.[1]

Pediatric surgeons and pediatric anesthetists reviewed the medical records. The data recorded for the study include patients' age, body weight, height, type of hypospadias, type of repair procedure, duration of surgery and hospitalization, and postoperative complications (UCF, urethral stricture, tissue edema, hematoma in scar tissue, penile engorgement, delayed tissue repair, and inflammation).

The Snodgrass method used in the study is tubularized incised plate urethroplasty for distal hypospadias. Urethral tubularization was performed using 7.0 polydioxanone (PDS) sutures. Meatal advancement and glanuloplasty (MAGPI) started with the liberalization of the posterior part of the urethra. The urethra was attached to the tip of the glans using 7.0 PDS sutures. Afterwards, glanuloplasty was performed with 5.0 PDS sutures. Instead of cystostomy, urethral stents were used to ensure urine flow. Meatal location was defined as mid-penile, penoscrotal, glanular, coronal, perineal, or sub-coronal (Figure 1). UCF was defined as an abnormal interaction between the reconstructed urethra and the skin, causing urine leakage.

Statistical analysis

Quantitative data were expressed as mean ± standard deviation (SD), and qualitative data as number (%). The compatibility of all measurable variables with normal distribution was evaluated using the Kolmogorov-Smirnov test. Univariate analyses were performed using the Chi-square test to investigate the relationships between UCF formation and the characteristics of

the patient and operation, as well as the other postoperative complications. All analyses were conducted on SPSS® version 22 software, with a p value of <0.05 considered as statistically significant.

RESULTS

The medical records of 122 patients were complete and, therefore, included in the study. The mean age of the patients was 4.8±3.7 years. The mean body weight and height were 20.3±12.7 kg, and 92.3±26.5 cm, respectively. The type of procedure performed for hypospadias surgery was tubularized incised plate urethroplasty (Snodgrass) in 90 (73.8%) children and meatal advancement and glanuloplasty (MAGPI) in 32 (26.2%) children, all of which were performed by an expert pediatric surgeon specialized in pediatric urology. The mean duration was 2.1±0.69 hours for surgery and 6.4±1.9 days for hospitalization.

No patient received preoperative testosterone stimulation. All cases received preoperative caudal anesthesia (0.25% bupivacaine ± epinephrine) for postoperative pain management in addition to general anesthesia.

The types of hypospadias determined in our patients are shown in Table 1. The mid-penile type was the most frequently encountered complication (n=54, 44.1%). The presence of a urethrocutaneous fistula was identified in 16 (13.1%) children. The second most frequently encountered complication was the development of tissue edema, which occurred in eight (6.5%) patients. The complications determined in our patients are presented in Table 2. When the postoperative complications were analyzed, it was found that all of the other complications had occurred only in patients who had developed a urethrocutaneous fistula. Table 3 shows the coexistence of the complications determined in our study.

Table 1. The types of hypospadias in our patients (n=122)

Type of hypospadias	n (%)
Mid-penile	54 (44.3)
Penoscrotal	27 (22.1)
Glanular	21 (17.2)
Coronal	10 (8.2)
Perineal	6 (4.9)
Sub-coronal	4 (3.3)
Total	122 (100.0)

Table 2. The types of complications in our patients (n=16)

Complication	n (%)
Urethrocutaneous fistula	16 (13.1%)
Tissue edema	8 (6.5%)
Urethral stricture	5 (4%)
Penile engorgement	3 (2.4%)
Hematoma	2 (1.6%)
Inflammation	1 (0.8)
Infection	1 (0.8)
Delay in tissue repair	1 (0.8)

Univariate analysis revealed no statistically significant association between the development of postoperative UCF and the variables such as age, body weight and height, duration of surgery and hospitalization, the type of surgical technique, the development of penile engorgement, urethral stricture and hematoma, and other complications in the surgical site.

DISCUSSION

The repair of hypospadias ensures that the phallus gains strength, and a more normal-looking urethral opening is created on the glans. In this study, we investigated the rate of postoperative complications, especially urethrocutaneous fistula, in patients who had undergone hypospadias repair and found that the rate of urethrocutaneous fistula was 13.1%, which is higher than recent studies.[6,7] However, this rate can be considered within the typical range since the incidence of UCB has been reported to range between 4 and 28%.[3] The most common complication of hypospadias repair is UCF, which mostly occurs in the immediate postoperative period. It leads to secondary corrective surgery, resulting in additional cost and discomfort to the patients and their families. The reasons for the development of UCF are not fully understood. In addition to the local deficit in growth factors, the surgical technique is thought to be a key factor.[7] The results of experimental treatments, such as local or systemic, adjuvant or neoadjuvant hormonal therapies, are controversial. In addition, postoperative care, including different types of dressing and surgical material, wound status, and antibiotics, may play a role.[8]

We also investigated the associations between the development of postoperative UCF and patient variables. However, no association was found to be present. Zaidi et al.[9] found no causal relationship between the use of caudal regional anesthesia and UCF formation. Similarly, Kreysing et al.[10] observed that caudal

Table 3. The status of coexistence of complications in our patients with complications (n=16)								
Patient	**Urethro-cutaneous fistula**	**Infection**	**Delay in tissue repair**	**Tissue edema**	**Penile engorgement**	**Inflammation**	**Urethral stricture**	**Hematoma**
1	+	-	-	+	-	-	+	-
2	+	-	-	+	-	-	-	+
3	+	-	-	-	+	-	+	-
4	+	-	-	-	-	-	-	-
5	+	-	-	-	-	-	+	-
6	+	-	-	-	-	-	-	-
7	+	-	-	+	-	-	-	-
8	+	+	+	+	+	+	+	+
9	+	-	-	+	-	-	-	-
10	+	-	-	+	+	-	-	-
11	+	-	-	+	-	-	-	-
12	+	-	-	-	-	-	-	-
13	+	-	-	-	-	-	-	-
14	+	-	-	+	-	-	+	-
15	+	-	-	-	-	-	-	-
16	+	-	-	-	-	-	-	-
Total n (%)	16 (13.1)	1 (0.8)	1 (0.8)	8 (6.5)	3 (2.4)	1 (0.8)	5 (4.0)	2 (1.6)

anesthesia had no effect on complication rates. However, another report concluded that patients undergoing CB were more likely to experience complications than those who received a dorsal penile block.[11] Meanwhile, Braga et al.[12] concluded that the severity of hypospadias, and the type of regional anesthesia, was the only risk factor significantly associated with postoperative complications. All patients underwent caudal-type regional anesthesia, which prevented us from evaluating its effects in these children. Hence, other causes of UCF are discussed in our study.

Although the exact causes of fistula formation are unknown, the type of hypospadias and the surgical technique, penile size, age of the patient, experience of the surgeon, presence of postoperative edema, penile engorgement, local infection, and local ischemia have been associated with its development.[11] Kundra et al.[5] reported that the presence of tissue edema was associated with postoperative UCF. They proposed a causal relationship between CB and UCF formation. In addition, they concluded that CB resulted in penile engorgement, up to a 27% increase from the baseline, and this increased penile edema caused by delayed tissue healing, which in turn led to UCF. The incidence of urethrocutaneous fistula after hypospadias repair in their study was 19.2% and all patients had received caudal block. However, the rate of penile engorgement in our study was 2.4% and all patients had received CB, in contrast to the higher rates of fistula formation in the CB group in the study by Kundra et al.[5] The results of our study showed a lower rate compared to the results of Kundra et al.[5] In addition, since the rate of penile engorgement was 2.4%, it was thought that penile fistula was associated with surgical technique, rather than CB.

Some authors have suggested that the duration of surgery and surgeon's experience are associated with a higher incidence of UCF.[2,9] It was also reported that similar fistulae rates were observed for different repair methods.[12] In this study, neither the duration of surgery and tourniquet application nor hospitalization were associated with the development of UCF. Hypospadias is often classified as posterior, penile, or anterior, depending on the preoperative position of the meatus.[13] Similar to our study, the mid-penile type was one of the most common types in the study by Braga et al.[12] The type of hypospadias, hence

the place of the meatus, is a critical factor for the development of postoperative fistula formation. Proximal types require longer operative time and have a higher risk of subsequent tissue edema and fistulae formation. In contrast to our study, Zaidi et al.[9] reported that wound infection was associated with fistula formation and that meatal location and the use of subcutaneous epinephrine were associated with higher rates of UCF.

One of the main rules of successful hypospadias repair is to select the most appropriate technique for a given patient to achieve the best result, both functionally and cosmetically. Meatal advancement and glanuloplasty (MAGPI), which was one of the techniques used in our study, is commonly used in hypospadias surgery despite some disadvantages such as postoperative meatal regression and meatal stenosis. However, tubularized incised plate urethroplasty (Snodgrass) is less influenced by such issues. Fistulas are the most common problem in most series and can be prevented via the interposition of a barrier layer between the neourethra and the overlying glans and shaft skin closures. Duckett and Snyder reported problems in 1.2% of 1000 boys 2 months after MAGPI.[14] In our study, the type of surgery was Snodgrass in 90 (73.8%) children and MAGPI in 32 (26.2%) children, which were performed by an expert pediatric surgeon, specialized in pediatric urology. The rate of fistula was 15/90 (16.6%) in Snodgrass and 1/32 (3.1%) in MAGPI procedures. Alsharbaini et al.[15] reviewed the results of 320 cases treated by Snodgrass urethroplasty and found the UCF rate to be 2.5%. We found a higher rate (16.6%), which may be due to the lower number of cases. Stenting material may have an impact on the UCF rates. Saraç et al.[16] compared the outcomes of hypospadias repair with respect to the stenting tube/catheter types. They used the Snodgrass method in all patients and found fistula development in 31.3% of the patients who underwent stent placement using a feeding tube, and 4.39% of patients who underwent stent placement stenting using a Foley catheter. However, we used the same stenting material for all patients.

Guidelines suggest that the optimal age for hypospadias repair is between 6 and 18 months; however, the mean age of our study population was significantly higher.[6] In general, penile size is not a limiting factor for the optimal timing of hypospadias repair, since penile growth is moderate in the first few years of life. Therefore, there is no particular advantage in delaying surgery. Additionally, surgery and hospitalization are less desirable after 18 months of age, when genital awareness begins[17] Complications can occur at any age. The increased complication rate in this study cannot be attributed to the higher mean age of the study population but may instead reflect the limited interest of the families, who are primarily responsible for the care of the children.

The results of our study must be interpreted in the context of its limitations. First, due to the retrospective nature of this study, data could not be obtained for some other potential confounding factors, such as underlying disorders and anatomical properties of the penis (ventral curvature degree, glans width, etc.).[1] The degree of chordee and the length of the urethral defect or fistula formation were not evaluated in the manuscript. Additionally, our study was a single-center study reflecting the experience of a single surgeon, which limits the generalizability of the results. More extensive prospective trials are needed to identify the risk factors for UCF in hypospadias surgery.

CONCLUSION

In conclusion, hypospadias repair is a challenging procedure with potential complications. UCF remains a significant problem in the postoperative period. We found no association between the incidence of postoperative UCF and patient variables. All patients underwent caudal block and despite previous literature, we experienced lower rates of penile engorgement and postoperative UCF. These results showed that there was no cause-and-effect relationship between the caudal block and UCF. We think that the development of a urethrocutaneous fistula is mostly related to surgical causes and that well-designed, prospective, and controlled studies are required to elucidate this issue.

Ethical approval

This study has been approved by the Aydın Adnan Menderes University Non-invasive Clinical Research Ethics Committee (approval date 12/09/2018, number 2018/1456). Written informed consent was obtained from the participants.

Author contribution

Surgical and Medical Practices: DK, AOE, PDE, BM, SKÖ, MY, FG; Concept: DK, AOE, PDE, BM, SKÖ, MY, FG; Design: DK, MY, FG; Data Collection or Processing: DK, AOE, PDE, BM, SKÖ, MY, FG; Analysis or Interpretation: DK, SKÖ, İKÖ, FG; Literature Search: DK, PDE, BM, SKÖ, FG; Writing: DK, FG. All authors reviewed the results and approved the final version of the article.

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Conflict of interest

The authors declare that there is no conflict of interest.

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