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ORIGINAL ARTICLE

Risk factors for the recurrence of perineal canal

Keisuke Kajihara[1] · Hiroaki Fukuzawa[1] · Koji Fukumoto[2] · Naoto Urushihara[2] · Yoshitomo Samejima[1] · Kotaro Uemura[1] · Kozo Nomura[1] · Insu Kawahara[1] · Kaori Isono[1] · Keiiti Morita[1] · Makoto Nakao[1] · Akiko Yokoi[1] · Kosaku Maeda[1]

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Abstract

Purpose The aim of this study was to investigate risk factors for recurrence in the perineal canal (PC).

Methods Patients with PC who underwent operations were enrolled in this study and were divided into recurrence and non-recurrence groups. Preoperative infection, the age at the operation, the presence of colostomy and the treatment procedure for fistula were retrospectively investigated. Regarding the treatment procedure for fistula, either closure of the rectal wall with stitches or ligation of fistula in the rectum was performed. These factors were compared between the two groups.

Results Six of 17 patients with PC who underwent surgical treatment had recurrence. There were no significant differences in the incidence of preoperative infection, age at operation or presence of colostomy ($p = 0.60$, 0.38, 1.00, respectively). In the recurrence group, all patients were treated by closure of the rectal wall. In the non-recurrence group, five were treated by the closure of the rectal wall with stitches and six by ligation of the fistula. There was a significant association between recurrence and the treatment procedure for fistula ($p = 0.04$).

Conclusion Closure of the rectal wall with stitches is a risk factor for the recurrence of PC.

Keywords Perineal canal · Vestibulo-anal pull-through · Ligation of fistula · Risk factor for recurrence

Introduction

Perineal canal is a relatively rare entity in congenital anorectal malformations, so-called recto-vestibular fistula with a normal anus, H-type anorectal malformation or double termination of the alimentary tract in females [1]. This malformation can only be treated by surgery. Various surgical procedures have been reported; however, the high frequency of recurrence has been a grave problem. To date, no studies have described the factors associated with the recurrence of fistula.

In the present study, we investigated the risk factors for recurrence in the surgical management of perineal canal.

Materials and methods

We retrospectively reviewed the medical records of patients with perineal canal who underwent surgical management at Kobe Children's Hospital and Shizuoka Children's Hospital from January 2004 to June 2018. Patients who had complicated fistulas (e.g., multiple fistulas or an incomplete fistula) were excluded from the study. The patients were divided into two groups: the recurrence group and the non-recurrence group. In each group, the following factors were ascertained: preoperative infection of the vulva or periphery of the vulva, age in months at the operation, presence or absence of colostomy at the radical operation and the procedure used to treat the fistula on the rectal side.

The distribution of variables was compared using Fisher's exact test and continuous variables were compared using an unpaired *t* test. *p* values or < 0.05 were considered to indicate statistical significance.

This study was approved by each local institutional ethics committee (no. R30-24, 2019087) and complied with the Helsinki Declaration of 1964 (revised in 2013).

✉ Keisuke Kajihara
ballack13_kay@yahoo.co.jp

[1] Department of Pediatric Surgery, Kobe Children's Hospital, 1-6-7 Minatojimaminamimachi, Chuo-ku, Kobe 650-0047, Japan

[2] Department of Pediatric Surgery, Shizuoka Children's Hospital, Shizuoka, Japan

Operative technique

In our study, regarding the procedure used to treat the fistula on the rectal side, two techniques were performed. In one procedure, we resected the fistula from the rectal wall and closed the anterior rectal wall with stitches. In the other procedure, we threaded a thin catheter through the fistulous tract (Figs. 1 and 3a), peeled off the fistulous tract from the vestibular side until the anterior rectal wall (Fig. 2), pulled the fistulous tract inside-out into the rectum (Fig. 3b, d) and only ligated the fistula in the rectum, without the resection of the fistula (Fig. 3c, e).

Results

A total of 19 patients with perineal canal were treated at Kobe Children's Hospital and Shizuoka Children's Hospital from January 2004 to June 2018. Two of these patients whose medical records indicated that they had complicated fistulas (multiple fistulas and an incomplete fistula) were excluded from the study. Thus, 17 patients were enrolled in this study (Table 1). Six patients had recurrent fistula; the other 11 patients had no recurrence. As the primary symptom feces passing through the vestibular fistula was seen in 15 cases, and infection of the vulva or periphery of the vulva was seen in six (vulva, $n=4$; perineum, $n=1$; right femur, $n=1$). Three patients each in the recurrence and non-recurrence groups had preoperative infection of the vulva or periphery of the vulva. There was no significant difference in the rate of preoperative infection ($p=0.60$) (Table 2). The age at the operation ranged from 2 to 10 months. The mean age at the operation in the recurrence and non-recurrence groups was 5.7 months and 6 months, respectively, and did not differ to a statistically significant extent ($p=0.38$). In the non-recurrence group, one patient underwent colostomy before the perineal canal operation.

Regarding the procedure used to treat the fistula on the rectal side, all six patients in the recurrence group were treated by closure of the anterior rectal wall with stitches. On the other hand, in the non-recurrence group, five patients were treated by closure of the anterior rectal wall with stitches and six patients were treated by vestibular-rectal pull-through inside-out and ligation of the fistula in the rectum. A univariate analysis demonstrated a statistically significant association between the recurrence of perineal canal and the procedure used to treat the fistula on the rectal side. The rate of recurrence after ligation (only) of the fistula in the rectum was significantly lower than that after closure of the rectal wall with stitches ($p=0.04$).

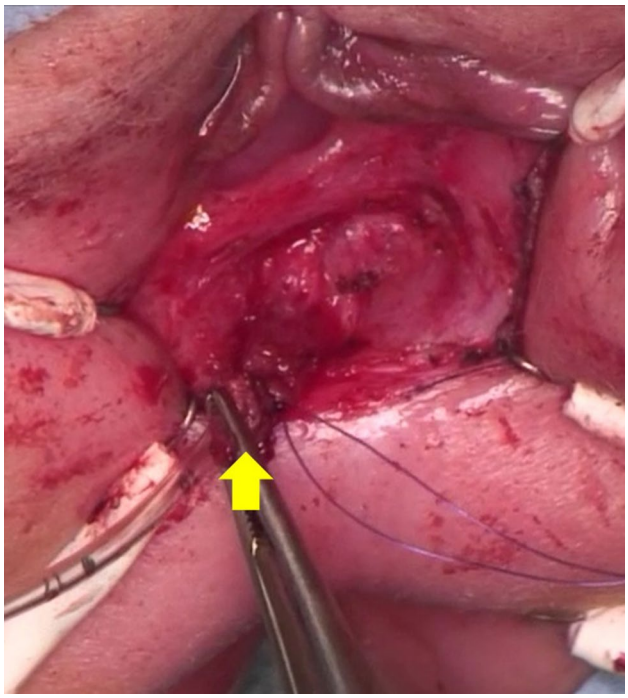

Fig. 2 The fistulous tract (arrow) was peeled off until the anterior rectal wall

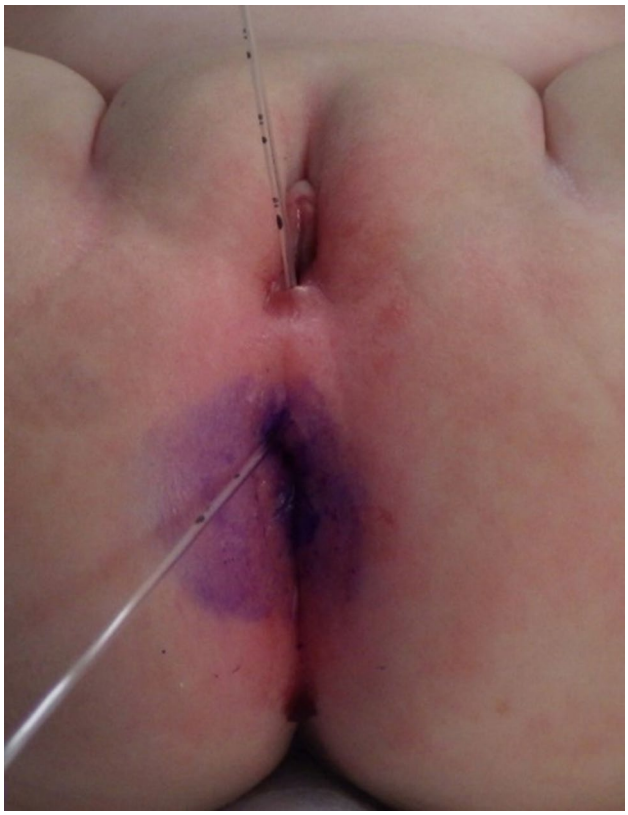

Fig. 1 A 4 Fr. catheter was threaded through the vesutibulo-anal fistula

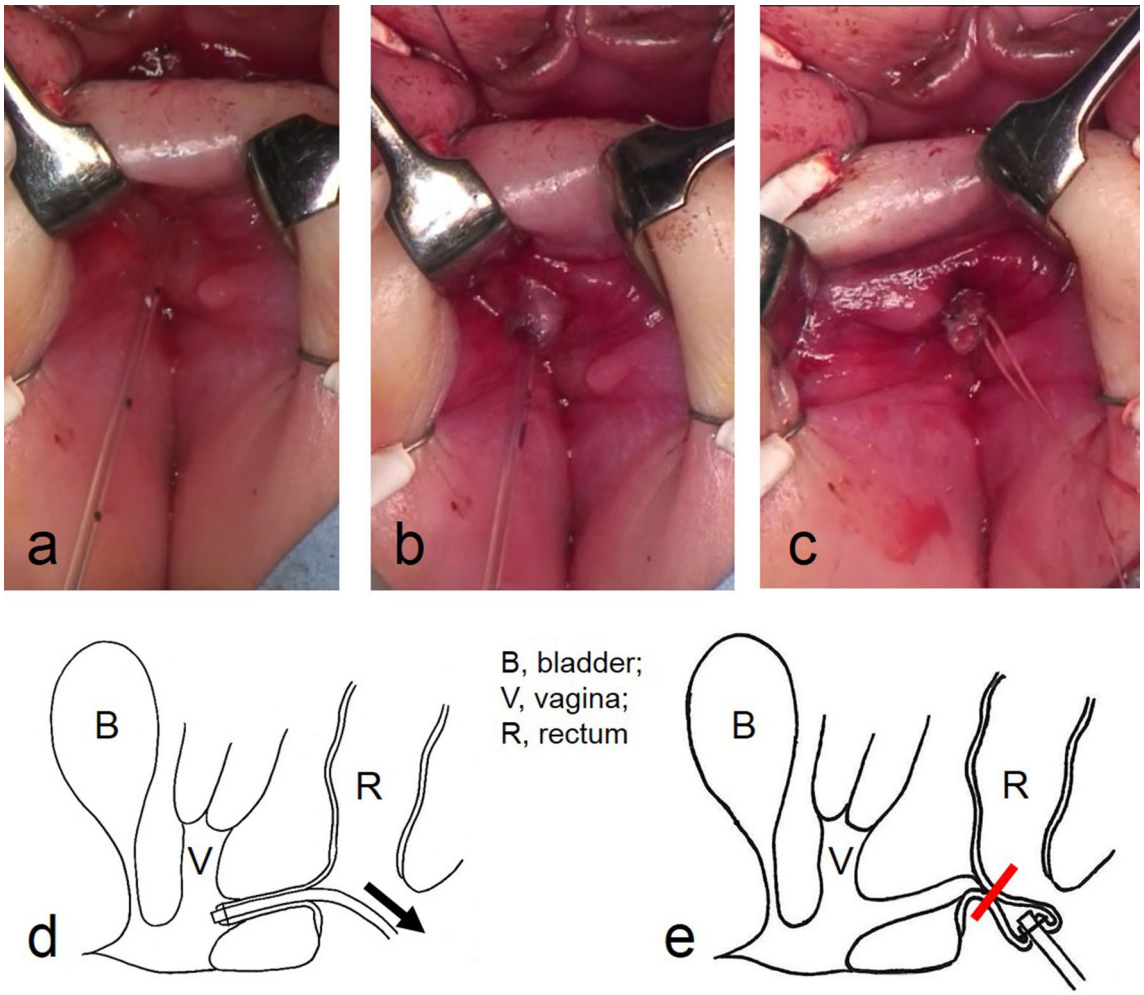

Fig. 3 **a** Fistula in a rectum with penetration of a catheter. **b** The fistulous tract was pulled inside-out into the rectum. **c** The fistula was only ligated in the rectum, without resection. **d**, **e** Schematic illustrations of the technique. The fistulous tract was pulled inside-out into the rectum by pulling the catheter (arrow). The fistula was ligated in the rectum on the rectal side (red line)

Discussion

Perineal canal is a rare congenital anorectal malformation. This entity is seen more frequently in Asia than it is in Europe and United States [1, 2]. Various surgical managements have been reported in the literature, including vestibulo-anal pull-through (VAP), recto-vestibular pull-through, pull-through of the anterior wall of the rectum, the perineal transverse procedure, posterior sagittal anorectoplasty, anterior sagittal anorectoplasty, and others [1–10]. In Japan, the major procedures seem to be VAP and the perineal transverse procedure. Some authors have reported no recurrence [2, 3]; however, in most reports, recurrence has been a major problem. Sharma et al. reported that the recurrence rate ranged from 5 to 60% in a systematic review [1]. Our study is the first report to investigate risk factors for recurrence in patients with perineal canal.

Tsugawa et al. [4] hypothesized that recurrence was due to local ischemia in the rectal tissue or failure to separate the divided ends of the fistula and that recurrence might originate from the rectum due to anal pressure or local infection. They performed a perineal transverse procedure, which included the closure of the healthy rectal wall with stitches and the attachment of the external sphincter muscle to the anterior rectal wall above the divided fistula [4]. For the purpose of leaving no sutures inside the anal canal, Tsuchida et al. reported pull-through of the anterior wall of the rectum [5]. Some authors reported similar techniques [3, 10]. All techniques that have been described in previous reports have included steps to close the anterior rectal wall with stitches. We presumed that failure of the sutures on the rectal wall was the main cause of recurrence. Thus, to minimize injury of the rectal wall, we tried to pull the fistulous tract inside-out into the rectum and to only ligate the fistula in the rectum without resection of the fistula. Actually, no recurrence was

Table 1 The clinical features of patients

Case	Age at primary symptom (months)	Primary symptom	Colostomy	Age at operation (months)	Procedure for treating a fistula on the rectal side	Recurrence	Treatment of recurrence
1	1	Swelling of vula, feces passing through the vestibular fistula	–	2	Closure of rectal wall with stitches	+	Lay open
2	2	Swelling and redness of vula, feces passing through the vestibular fistula	–	3	Closure of rectal wall with stitches	+	Lay open
3	2	Feces passing through the vestibular fistula	–	9	Closure of rectal wall with stitches	+	Core out of fistula
4	1	Fever, swelling and redness of vulva, feces passing through the vestibular fistula	–	4	Closure of rectal wall with stitches	+	Pull-through of anterior rectal wall
5	1	Feces passing through the vestibular fistula	–	6	Closure of rectal wall with stitches	+	Lay open
6	2	Feces passing through the vestibular fistula	–	10	Closure of rectal wall with stitches	+	Perineal transverse procedure
7	1	Feces passing through the vestibular fistula	–	8	Closure of rectal wall with stitches	–	
8	1	Cellulitis of right femur	–	4	Closure of rectal wall with stitches	–	
9	5	Swelling and redness of vulva, feces passing through the vestibular fistula	–	6	Closure of rectal wall with stitches	–	
10	1	Feces passing through the vestibular fistula	–	3	Closure of rectal wall with stitches	–	
11	1	Swelling and redness of vulva, feces passing through the vestibular fistula	–	5	Closure of rectal wall with stitches	–	
12	2	Feces passing through the vestibular fistula	–	5	Ligation of fistula	–	
13	2	Feces passing through the vestibular fistula	–	5	Ligation of fistula	–	
14	0	Feces passing through the vestibular fistula	–	5	Ligation of fistula	–	
15	1	Feces passing through the vestibular fistula	–	9	Ligation of fistula	–	
16	6	Feces passing through the vestibular fistula	–	8	Ligation of fistula	–	
17	2	Feces passing through the vestibular fistula	–	8	Ligation of fistula	–	

observed among patients who were treated with this technique, and the rate of recurrence after ligation (only) of the fistula in the rectum was significantly lower than that after closure of the rectal wall with stitches. These findings suggested that vestibulo-anal pull-through inside-out and ligation (only) of the fistula in the rectum were sufficient for the effective treatment of perineal canal.

In our study, all patients with preoperative infection of the vulva or periphery of the vulva were treated medically, and the operation was performed after the infection was controlled. Generally, local infection at the time of an operation is a risk factor for wound dehiscence. This study demonstrated no significant association between the recurrence of perineal canal and preoperative infection. However, it seems to be better to perform operation of perineal canal after the control of infection.

Most cases reported in the literature were treated without colostomy. Sharma et al. asserted that colostomy did not

Table 2 A comparison between the recurrence and non-recurrence groups (a univariate analysis)

	Recurrence		*p* value
	+	−	
	n = 6	*n* = 11	
Preoperative infection			
+	3	3	0.60
−	3	8	
Colostomy			
+	0	1	1.00
−	6	10	
Age at operation (months)	5.7	6	0.38
Procedure for treating a fistula on the rectal side			
Ligation of fistula	0	6	0.04
Closure of the rectal wall with stitches	6	5	

reduce the rate of recurrence [1]. Similarly, in our study, there was no significant association between recurrence and the presence of colostomy.

The relationship between the age at the operation and the outcome of treatment in patients with perineal canal has not been discussed in previous reports. Our study demonstrated no significant association between recurrence of perineal canal and the age of the operation. As Tsugawa et al. hypothesized [4], high anal pressure can be a risk factor for recurrence from the rectum. Because food consumed during weaning will make the stool harder, we believe that the treatment of perineal canal should be performed before a start of weaning.

Limitations

The present study was associated with some limitations. The number of patients with perineal canal was relatively small. As a consequence, a multivariate analysis could not be performed. Thus, we only performed a univariate analysis. As a result, any confounding factor could have influenced our results. All of our cases were female infants with a single fistula. Banu et al. and Rintala et al. reported on patients with multiple fistulas [6, 7]. The procedure for treating a fistula on the rectal side, vestibule-anal pull-through inside-out and ligation of fistula in the rectum, is difficult to perform for patients with multiple fistulas.

Conclusion

In patients with perineal canal, closure of the rectal wall with stitches was found to be a risk factor for recurrence.

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Compliance with ethical standards

Conflict of interest The authors declare no conflicts of interest in association with the present study.

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