Journal of Pediatric Surgery xxx (2017) xxx–xxx

Contents lists available at ScienceDirect

Journal of Pediatric Surgery

journal homepage: www.elsevier.com/locate/jpedsurg

Efficacy of an anal fistula plug for fistulas-in-ano in children☆,☆☆,★,★★

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ARTICLE INFO

Article history:
Received 8 August 2016
Received in revised form 25 January 2017
Accepted 28 January 2017
Available online xxxx

Key words:
Perianal abscess
Cryptoglandular fistulas
Children

ABSTRACT

Background: In children, perianal abscesses have a good prognosis and often heal with age. However, some perianal abscesses are refractory to treatment and remain as fistulas-in-ano. Treatment with a Surgisis Anal Fistula Plug® has been reported as a new method of treatment for fistulas. In adults, the plug has been reported to cause little pain and have a high cure rate, but there have been no reported cases of its use in children.
Purpose: This study was designed to analyze the efficacy of the plug for closure of refractory fistulas in children.
Methods: Since the plug has not been approved as a medical device in Japan, application for its use was submitted to the ethics committee of our university, and approval was granted, marking the first use of the plug in Japan. We classified refractory fistulas as those treated for 6 months or longer and remaining unhealed, even after 1 year of age, despite continued conservative treatment. The plug was used in 11 refractory fistulas in 8 children.
Results: Eight of 11 fistulas (72.7%) were successfully treated. Three fistulas recurred, and fistulectomies were performed. No sequelae were observed after AFP treatment.
Conclusion: The plug was effective even for closure of refractory fistulas without sequelae in children.
Levels of evidence: Treatment Study, Level IV

A perianal abscess (PA) is a condition that develops overwhelmingly in male infants younger than one year [1–3]. Most PAs heal with incision and drainage or medication. Some PAs form fistulas-in-ano (FIAs). The incidence of PAs evolving toward FIAs has been previously reported to range from 20% to 77% [2–8]. However, once an FIA is formed, spontaneous resolution is expected in a high percentage of infants. Therefore, several reports have advocated that early fistulotomy or fistulectomy should be avoided in infants [3–5,9]. We classified refractory FIAs as those treated for 6 months or longer and remaining unhealed, even after 1 year of age, despite continued conservative treatment. For refractory FIAs, surgical intervention with fistulotomy and/or fistulectomy was performed. In cases of multiple fistulas or a fistula with a long tract, surgery is even more invasive and has a risk of a damage to the sphincter muscle [10]. Another treatment, fibrin glue injection, has been reported. Studies of fibrin glue showed wide variability in healing rates [11–13]. Use of the Surgisis Anal Fistula Plug® (AFP; Cook Surgical Inc., Bloomington, IN) has been reported a number of times in cases involving adults [14–23]. The AFP is a golf tee-shaped porcine collagen matrix. Once the AFP is inserted in the fistula tract and secured, it is then naturally absorbed by the body. This absorption process leads to closure of the anal fistula. Use of the AFP involves reduction in operating time, no risk of damage to the sphincter muscle, and a high cure rate. However, there have been no reports of its use in children. In this study, AFPs were used to treat refractory FIAs in children.

1. Patients and methods

1.1. Patients

Between December 2006 and December 2015, 135 children (133 males and 2 females) with PAs and FIAs were treated at our hospital. None of the children had anorectal or inflammatory disease. Initially, all children were treated by conservative treatment with medication and local treatment. Children with PAs or FIAs had received conservative treatment with Juzentaihoto, a traditional herbal medicine that acts on the intestinal immune system. Antibiotics and stool softeners were not used. Local treatment (incision or aspiration for abscess) was required for children who had an enlarged PA and pain. During conservative treatment, some formed fistulas. FIA was defined by the opening hole persisting for more than three weeks [2,3]. Refractory FIAs were defined as those treated for more than 6 months and remaining unhealed, even after one year of age, despite continued conservative treatment. Ultimately, 8 patients with refractory FIAs were chosen to undergo treatment with an AFP.

The AFP has yet to be approved as a medical device in Japan. Its use in this study was approved by the Ethics Committee of Tokyo Women's Medical University. The AFP was explained to patients' families, and informed consent was obtained prior to its use.

☆ Financial disclosures, if any: Nothing
☆☆ The source(s) of support in the form of grants, equipment drugs, or all of these: Nothing
★ State if meeting presentation-name of meeting, date and year: None
★★ Correspondence author performed all operation of the anal fistula plug insertion for children and each coauthor supported those.

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http://dx.doi.org/10.1016/j.jpedsurg.2017.01.062
0022-3468/

2. Methods

All of the patients were treated under general anesthesia in the supine jackknife position. No seton was inserted prior to the operation. The fistula tract was washed with hydrogen peroxide, and a nylon string was then passed through the fistula tract. Using the nylon string as the guideline, an AFP was inserted along the full length of the fistula tract via the external opening of the tract. The AFP was sutured close to the internal opening with 5–0 absorbable suture. The internal opening was closed by this suture. The excess portion of the AFP was removed via the external opening of the tract. Since the AFP is golf tee shaped, the insertion methods were the same as for adult patients, and no plug modification was required for use in children. No antibiotics and no stool softeners were administered before or after the operation. Oral intake began 2 h after surgery, and all patients were discharged the following day. After the AFP treatment, FIAs were observed at 1 week, and 1 and 6 months later.

3. Results

A total of 135 patients with PAs and FIAs were treated at this hospital. The male: female ratio was 133:2. Eighty of 135 patients (59.3%) had the first clinical symptoms younger than 5 months, and most patients (122 of 135: 90.4%) had them less than 11 months (Fig. 1). The overall formation rate of FIAs was 24 of 80 (30.0%) younger than 5 months at the first clinical symptom; 16 of 42 (38.1%) in 6 to 11 months old; 2 of 9 (22.2%) in one-year-olds; and 2 of 4 (50.0%) older than 2 years old. Refractory FIAs occurred in 8 of 135 (5.9%) children. The incidence of refractory FIAs had a tendency to increase with the age of the first clinical symptom: younger than 5 months, 3 of 80 (3.8%); 5–11 months, 4 of 42 (9.5%); and 1 year, 1 of 9 (11.1%). However, there was no significant correlation between the age of first clinical symptoms and the development of a refractory fistula. In children more than 2 years old, no patients extended to a refractory FIA and required an AFP. In our hospital, only two female patients had FIAs. Both patients had FIAs that extended to the vagina. One was a 1-year-old girl who had a recurring FIA since 1 month of age. In the other female patient, the FIA was cured spontaneously during conservative treatment. AFPs were inserted in 11 refractory anal fistulas in 8 children (Table 1). Only one child had postoperative pain and was given an analgesic, but the other 7 children did not have pain, and no analgesic was given. Regarding complications, early dislodgement of the AFP was noted in 1 child, but infection of the insertion site was not noted in any of the other children. Six months after surgery, 8 of 11 anal fistulas (72.7%) had healed. All 3 recurrences occurred within 6 months after AFP treatment. One FIA in a child had early dislodgement of the AFP, which recurred at 3 months postoperatively. For 3 recurrent fistulas, fistulectomy was performed. Subsequent recurrence was not noted 1–9 years after the last surgical treatment.

4. Discussion

In children, PAs and FIAs occur overwhelmingly in boys [1,2]. Conservative treatment involves administration of medication, such as antibiotics or stool softeners. Local treatment by needle aspiration or incision was performed. In infants, several papers have reported that the majority of PAs and FIAs resolved with conservative or local treatment [3–5,9]. The overall incidence of PAs extending toward FIAs reported in the literature ranges from 20% to 77% [2–8]. In the present series, the overall incidence of FIA development was 32.6% (44 of 135). This percentage is not different from other published data. Only a few cases with FIAs recurred and were not cured spontaneously. In the present series, only 8 of 135 PAs/FIAs (5.9%) developed refractory FIAs and required surgical treatment. This percentage is smaller than the overall operation rate of previous reports (6.2–84.6%) [2,3,5,6,8,9]. For refractory FIAs, surgical management, such as fibrin glue injection or surgical removal, is needed. Fibrin glue injection does not cause pain, but its healing rate ranges from 14% to 76% [11–13]. The recurrence rate of fistulectomy in infants was reported to range from 0% to 25% in some series [1,2,4–10,24–26]. In cases of a fistula with a long tract from the anal edge or multiple fistulas, fistulectomy is invasive.

Numerous overseas reports have described the use of AFPs in adults. Its healing rate ranges from 20% to 86% [14–23]. There have been no reports of AFP use in children, either in Japan or overseas. AFP treatment has the advantage of its surgical technique compared with fistulectomy and/or fistulotomy. AFP insertion requires a short operation time and has no risk of damaging the sphincter muscle. This advantage is mostly apparent in patients with a long and/or multiple FIAs. In the present series, the recurrence rate with AFP treatment was 27.3% (3 of 11 refractory FIAs). This incidence is not much higher than with fistulectomy. All insertion procedures were finished within 15 min in the present study.

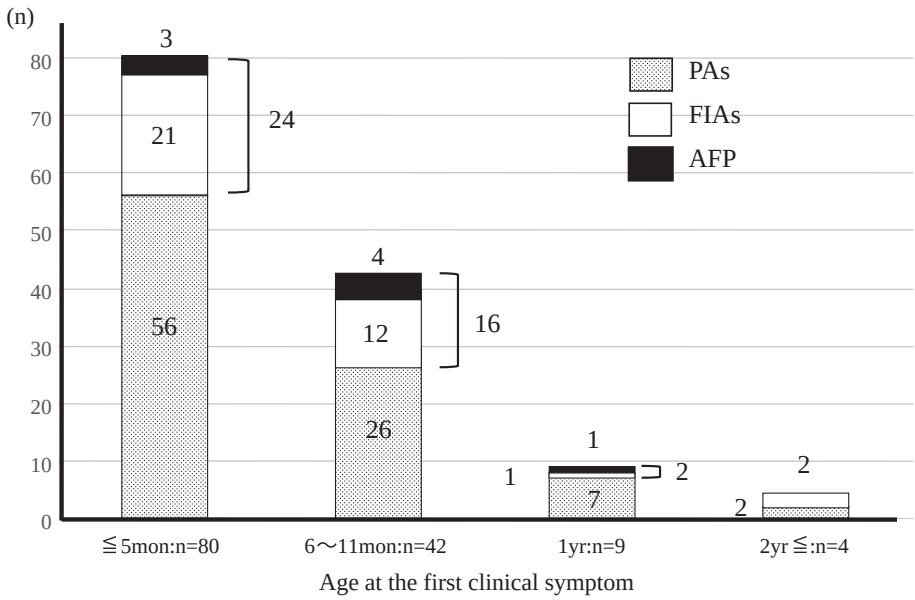

Fig. 1. Outcomes of PAs and FIAs treated in our hospital. Abbreviations: PA, perianal abscess; FIA, fistula-in-ano; conservative, conservative treatment; AFP, anal fistula plug; mon, month; yr., year.

Please cite this article as: Kouchi K, et al, Efficacy of an anal fistula plug for fistulas-in-ano in children, J Pediatr Surg (2017), http://dx.doi.org/10.1016/j.jpedsurg.2017.01.062

Table 1
Clinical features of children and outcomes.

Patient	Sex	First clinical symptom	Age at time of admission	Fistula	Pre-treatment	AFP Insertion	Sequela	Healing
1	M	6 mon	6 mon	1	Medication; 9 mon	1 yr 5 mon	Non	1/1
2	M	1 mon	6 yr	1	Medication; 5 yr	6 yr	Non	1/1
3	M	8 mon	8 mon	2	Medication; 10 mon	1 yr 6 mon	Non	2/2
4	M	4 mon	3 yr	2	Medication; 3 yr	3 yr	ED in 1	1/2
5	M	10 mon	5 yr	2	Medication; 5 yr	5 yr	Non	2/2
6	F	1 mon	1 yr	1	Medication; 1 yr	1 yr 9 mon	Non	1/1
7	M	1 yr	6 yr	1	Non	6 yr	Non	0/1
8	M	6 mon	8 yr	1	Non	8 yr	Non	0/1

Abbreviations: AFP, anal fistula plug; M, male; F, female; yr., year; mon, month; ED, early dislodgement.

Complications of AFP have been reported to include dislodgement of the AFP and infection at the insertion site. Early dislodgement is when the AFP dislodges 1–2 days after insertion and occurs in about 3–7% of adult cases [15,18]. This is primarily attributed to the fact that the AFP was inadequately secured at the primary opening of the tract and the mismatching of the diameters of the plugs and fistulas. In cases with wide-diameter fistulas not suitable for an AFP, insertion of a seton was recommended for 6–8 weeks before AFP insertion [27]. In children, no FIAs showed enlargement of their diameters, and there were no cases of mismatching of the diameters between a plug and an FIA. Therefore, plug modification was not required for AFP insertion. In the present study, the AFP dislodged in 1 child, and recurrence of the anal fistula was noted. Infection develops in the form of redness and swelling at the insertion sites of AFPs and is noted in 0–3% of cases [12,15,21]. In our hospital, infection caused by AFP insertion was not noted in any of the patients. Postoperative pain was minimal, and an analgesic was used in only one patient. All of the patients were discharged on the day after surgery.

Using an AFP to treat refractory anal fistulas in children involves a simple surgical procedure, short operation time, and little pain, and it has a high cure rate. We propose that AFP treatment as the first-choice treatment in children with refractory anal fistulas.

References

[1] Longo WE, Touloukian RJ, Seashore JN. Fistula in ano in infants and children. Pediatrics 1991;87:737–9.
[2] Serour F, Somekh E, Gorensterin A. Perianal abscess and fistula-in-ano in infants: a different entity? Dis Colon Rectum 2005;48:359–64.
[3] Watanabe Y, Todani T, Yamamoto S. Conservative management of fistula in ano in infants. J Pediatr Int 1998;13:274–6.
[4] Poenaru D, Yazbeck S. Anal fistula in infants: etiology, features, management. J Pediatr Surg 1993;28:1194–5.
[5] Afsalar CE, Karaman A, Tanir G, et al. Perianal abscess and fistula-in-ano in children: clinical characteristic, management and outcome. Pediatr Surg Int 2011;27:1063–8.
[6] Al-Salem AH, Laing W, Talwalker V. Fistula-in-ano in infancy and childhood. J Pediatr Surg 1994;29:436–8.
[7] Festen C, van Harten H. Perianal abscess and fistula-in-ano in infants. J Pediatr Surg 1998;33:711–3.
[8] Ezer SS, Oğuzkurt P, Ince E, et al. Perianal abscess and fistula-in-ano in children: aetiology, management and outcome. J Paediatr Child Health 2010;46:92–5.
[9] Chang HK, Ryu JG, Oh JT. Clinical characteristics and treatment of perianal abscess of fistula-in-ano in infants. J Pediatr Surg 2010;45:1832–6.
[10] Schouten WR, van Vroonhoven TJMV. Treatment of anorectal abscess with or without primary fistulectomy. Dis Colon Rectum 1991;34:60–3.
[11] Aitola P, Hiltunen KM, Matikainen M. Fibrin glue in perianal fistulas-a pilot study. Ann Chir Gynaecol 1999;88:136–8.
[12] Johnson EK, Gaw JU, Armstrong DN. Efficacy of anal fistula plug vs. fibrin glue in closure of anorectal fistulas. Dis Colon Rectum 2006;49:371–6.
[13] Haim N, Neufeld D, Ziv Y, et al. Long-term results of fibrin glue treatment for cryptogenic perianal fistulas: a multicenter study. Dis Colon Rectum 2011;541279-83.
[14] Champagne BJ, O'Connor LM, Ferguson M, et al. Efficacy of anal fistula plug in closure of cryptoglandular fistulas: long-term follow-up. Dis Colon Rectum 2006;49:1817–21.
[15] van Koperen PJ, D'Hoore A, Wolthuis AM, et al. Anal fistula plug for closure of difficult anorectal fistula: a prospective study. Dis Colon Rectum 2007;50:2168–72.
[16] Christoforidis D, Etzioni DA, Goldberg SM, et al. Treatment of complex anal fistulas with the collagen fistula plug. Dis Colon Rectum 2008;51:1482–7.
[17] Christoforidis D, Pieh MC, Madoff RD, et al. Treatment of transsphincteric anal fistulas by endorectal advancement flap or collagen fistula plug: a comparative study. Dis Colon Rectum 2009;52:18–22.
[18] Schwandner T, Roblick MH, Kierer W, et al. Surgical treatment of complex anal fistulas with the anal fistula plug: a prospective, multicenter study. Dis Colon Rectum 2009;52:1578–83.
[19] Ellis CN, Rostas JW, Greiner GF, et al. Long-term outcomes with the use of bioprosthetic plugs for the management of complex anal fistulas. Dis Colon Rectum 2010;53:798–802.
[20] Neal EC. Outcomes with the use of bioprosthetic grafts to reinforce the ligation of the intersphincteric fistula tract (BioLIFT procedure) for the management of complex anal fistulas. Dis Colon Rectum 2010;53:1361–4.
[21] van Koperen PJ, Bemelman WA, Gerhards MF, et al. The anal fistula plug treatment compared with the mucosal advancement flap for cryptoglandular high transsphincteric perianal fistula: a double-blinded multicenter randomized trial. Dis Colon Rectum 2011;54:387–93.
[22] Han JG, Wang ZJ, Zhao BC, et al. Long-term outcomes of human acellular dermal matrix plug in closure of complex anal fistulas with a single tract. Dis Colon Rectum 2011;54:1412–8.
[23] de la Portilla F, Rada R, Jimenez-Rodriguez R, et al. Evaluation of a new synthetic plug in the treatment of anal fistulas: results of a pilot study. Dis Colon Rectum 2011;54:1419–22.
[24] Fitzgerald RJ, Harding B, Ryan W. Fistula-in-ano in childhood: a congenital etiology. J Pediatr Surg 1985;20:80–1.
[25] Shafer AD, McGlone TP, Flanagan RA. Abnormal crypts of Morgagni: the cause of perianal abscess and fistula-in-ano. J Pediatr Surg 1987;22:203–4.
[26] Grant CS, Al-Salem AH, Anim JT, et al. Childhood fistula-in-ano; a clinic-pathological study. Pediatr Surg Int 1991;6:207–9.
[27] O'Connor L, Champagne BJ, Ferguson MA, et al. Efficacy of anal fistula plug in closure of Crohn's anorectal fistulas. Dis Colon Rectum 2006;49:1569–73.

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