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Assessment of the Heineke–Mikulicz anoplasty for skin level postoperative anal strictures and congenital anal stenosis☆,☆☆

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ABSTRACT

Introduction: Acquired skin-level strictures following posterior sagittal anorectoplasty (PSARP) and some rare cases of congenital anal stenosis can be managed using a Heineke–Mikulicz like anoplasty (HMA). We hypothesized that this procedure was an effective, safe, and durable outpatient procedure in select patients.
Methods: We retrospectively reviewed all patients who underwent HMA for skin level strictures following PSARP or for certain congenital anal stenoses from 2014 to 2017.
Results: Twenty-eight patients (19 males, 9 females) with a mean age of 5.8 years (range 0.5–24.4) underwent HMA. Twenty-six had a prior PSARP, of which 18 were redo, and 8 were primary procedures. Two patients had congenital skin level anal stenosis. The mean follow up was 1.0 years (range 0.4–2.9). The average preprocedure anal size was Hegar 8, which after HMA increased 8 Hegar sizes to 16 (95% CI 7–9, $p < 0.001$). There were no operative complications. One patient restenosed and required a secondary procedure.
Conclusion: HMA is a safe procedure for skin-level anal strictures following PSARP (primary and redo) and can also be used in some rare cases of congenital anal stenosis. Long-term follow up to determine the restricture rate is ongoing. A plan to do an HMA if a stricture develops may offer an alternative to routine anal dilations, particularly after a redo PSARP in an older child.
Type of study: Case series.
Level of evidence: Level IV.

Anal stricture is a well-described complication following posterior sagittal anorectoplasty (PSARP) and results from inadequate blood supply or tension at the anorectal anastomosis or dissection within the incorrect tissue plane leading to damage of the intramural blood supply [1]. It is the most common indication for reoperation after PSARP [1]. While the precise incidence is not known, it has been reported in up to 20% of patients [2]. Postoperative dilations have been routinely performed by most surgeons to avoid stricture at the site of the anoplasty [3], but this practice has recently been questioned [4,5]. Dilations, when implemented, can be impossible, particularly in older children, or patients can be noncompliant, leading to the need for further surgical intervention. It is important to note that postoperative anal strictures can take two forms in these patients; the first is a stricture within the anal canal located at a depth greater than 2–3 mm from the anocutaneous junction and a width of 3–4 mm. This situation requires a redo PSARP with remobilization of the distal rectum to reach healthy lumen [1]. The second, which is the subject of this review, is a thin, skin-level stricture at the circumferential anastomosis of mucosa to skin which we believe is amenable to a superficial procedure.

The Heineke–Mikulicz stricturoplasty was first described in 1886 as a technique to enlarge the strictured pylorus in patients with peptic ulcer disease. Since that time it has become a popular technique in the management of short segment small bowel strictures in patients with Crohn's disease [6], and has also been applied to a wide variety of stricturing conditions, including urethral stricture in urologic surgery [7], common bile duct strictures [8], and ostial stenosis in cardiac surgery [9]. We have described previously the Heineke–Mikulicz technique in the management of patient with skin-level anal strictures after PSARP [10], and felt its long-term durability needed to be documented.

We aimed to review all patients at our institution for whom we performed a Heineke–Mikulicz like anoplasty (HMA) done for either skin-level stricture after PSARP or in certain rare cases of congenital anal

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☆☆ How this paper will improve care: The Heineke–Mikulicz anoplasty is a safe and effective outpatient procedure for the treatment of skin-level strictures after PSARP and select cases of congenital anal stenosis and may provide an alternative treatment to routine dilations after primary PSARP.

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stenosis. We wanted to evaluate the safety and functional outcomes following this procedure. Our hypothesis was that this procedure was an effective, safe, and durable outpatient procedure in select patients.

1. Methods

We retrospectively reviewed all patients who underwent HMA at our Center for skin level strictures following PSARP or for congenital anal stenosis from 2014 to 2017. It should be noted that it is our current practice to perform routine dilations after our own primary PSARP but not after redo PSARP given that this population of patients is generally older than patients at the time of primary PSARP, and they often have a history of previous traumatic dilation. For patients in whom dilations are performed, we begin daily dilations at 2 weeks postoperatively, and gradually increase the size of the dilator until the desired size is reached. At that point, the colostomy can be closed. Dilations continue until there is no longer resistance with insertion of the final dilator. We then wean dilations over the subsequent 3–4 months [11,12].

Demographic information including age, sex, underlying diagnosis, date of previous PSARP, perioperative complications, recurrent stenosis, and need for revision was recorded. Parents (or the patient if they were older than 18) were then contacted by telephone and asked a series of questions related to their experience with the procedure. Descriptive statistics, including frequencies and percentages for categorical variables, were performed using Microsoft Excel 2010 (14.0.7015.1000), Microsoft Corporation. This study was approved by the Institutional Review Board at Nationwide Children's Hospital.

1.1. Perioperative care

Preoperatively, patients receive a single dose of broad spectrum antibiotics prior to the start of the procedure. No preoperative bowel preparation was used, but all patients had a routine bowel management program, and impaction was ruled out prior by abdominal x-ray. After the HMA, a dilation protocol is not utilized and routine wound care is practiced. Parents are advised to avoid placing anything per rectum and to avoid straddling for 1 week. From a bowel management standpoint, the family is instructed to resume their prior regimen (after 1 week if enema are involved), with any later adjustments made as a result of their clinical response. No routine check of anal size was performed, but follow up to ensure good colonic emptying was performed at 2 to 3 months.

1.2. Surgical technique

Prior to the start of the procedure, the size of the anus was measured with a Hegar dilator. With the patient in supine position and legs in stirrups, four traction sutures were placed at the 2, 4, 8, and 10 o'clock positions. Electrocautery was then used to radially incise the strictured anocutaneous junction at the 12, 3, 6, and 9 o'clock positions to a depth of 2 to 3 mm until the scar released. The incision was then closed concentric to the anal opening with absorbable sutures and repeated at each incision. After completion of the procedure, the size of the anal opening was measured with a Hegar dilator. This technique is illustrated in Fig. 1.

2. Results

28 patients (19 males, 9 females) underwent HMA over the study period. The mean age at the time of HMA was 5.8 years (range 0.5–24.4). Patients underwent HMA at a mean of 1.9 (range 0.2–13.1) years following PSARP. 25/28 (89%) of patients were symptomatic from their stricture, with all 25 patients presenting with constipation and 9/28 (32%) having one or more episodes of fecal impaction. The remaining 3 patients were diagnosed with a stricture on routine examination after PSARP. 26 had undergone a prior PSARP, 25 for an anorectal malformation and 1 for a perineal sepsis-induced anal stricture secondary to acute lymphoblastic leukemia. Patients presented after a redo PSARP in 18 cases and after primary PSARP in the remaining 8 cases. Congenital skin level anal stenosis was the indication for HMA in 2 patients. The mean follow up was 1.0 years (range 0.4–2.9). Table 1 summarizes the cohort.

Over the study period, we performed a total of 136 primary PSARPs. Of the 8 patients presenting after a primary procedure, 3 were patients who had their PSARP at our institution. Thus, the rate of skin-level stricture after primary PSARP at our institution was 2.2%. All three patients underwent routine postoperative dilations starting at two weeks [11,12].

The average preprocedure anal size, Hegar 8 (range 6–12), increased 8 Hegar sizes after HMA (95% CI 7–9, $p < 0.001$) to Hegar 16 at the completion of the procedure. There were no intraoperative complications, episodes of perineal infection, or dehiscences. 23 patients (79%) were discharged home the same day, 5/29 (17%) had the HMA as part of another procedure requiring admission, and 1 patient (3%) with a history of respiratory disease was admitted for overnight observation. At a mean follow-up of 1.0 years (range 0.4–2.9), 27/28 patients (97%) show no clinical evidence of stricture. 1 patient (3%) with a prior redo PSARP required a secondary HMA (Table 2). 9 patients (45%) returned to normal activities in <1 day, 6 (30%) returned to normal activities in 1–3 days, and the remaining 5 patients (25%) resumed full activities in 4–7 days. The surgical outcomes are summarized in Table 2.

At a mean follow-up of 14 months (range 6–42 months post-HMA), 11/20 (55%) parents described their child's severity of constipation or frequency of impaction as "significantly improved," 5/20 (25%) reported their symptoms had "slightly improved," and 4/20 (20%) described "no change" after the procedure. No parents reported any worsening of their child's stooling symptoms or new episodes of incontinence after HMA. Additionally, of children who were previously managed with laxatives, 2/8 (25%) were able to decrease their laxative dose and another 2/8 (25%) were able to discontinue laxatives altogether. 11/12 (91%) parents whose children were managed with enemas indicated that their flushes were more effective after HMA, while 2/12 (19%) were able to transition onto a laxative-based regimen. Only 3/20 (15%) parents stated that they would be willing or able to perform dilations again in the future if needed, with 15/17 citing trauma as the reason they would not perform dilations (Table 3).

3. Discussion

The optimal surgical management of a postoperative stricture after PSARP is not well defined, although a number of techniques exist [10,13–15]. We previously reported on the technique of a Heineke–Mikulicz-like anoplasty for the treatment of skin-level strictures [10]. Since the effectiveness and durability of this procedure remain unknown we chose to evaluate the technique further, focusing on its safety and longer term functional outcomes. Our results show that this procedure can be safely performed in the outpatient setting and demonstrates promising and durable functional outcomes.

Of the 23 children who underwent isolated HMA, only 1 child with a chronic history of respiratory disease required overnight respiratory management and was discharged the following day. The remaining patients who were admitted required inpatient care for reasons related to their combined procedure with this approach and resumption of diet right away. There were no intraoperative complications, episodes of wound dehiscence, wound infections, or perineal sepsis.

From a technical standpoint, an anoplasty performed for skin-level stricture after PSARP should 1) return the anal opening to a size normal for the child's age creating a supple skin to mucosa connection that grows with the child, 2) be performed with minimal dissection of the anorectum, and 3) minimize the risk of future restricturing. All children in our study experienced an immediate increase in the size of their anal opening, from a mean Hegar size of 8 to Hegar size 16, the benefit of

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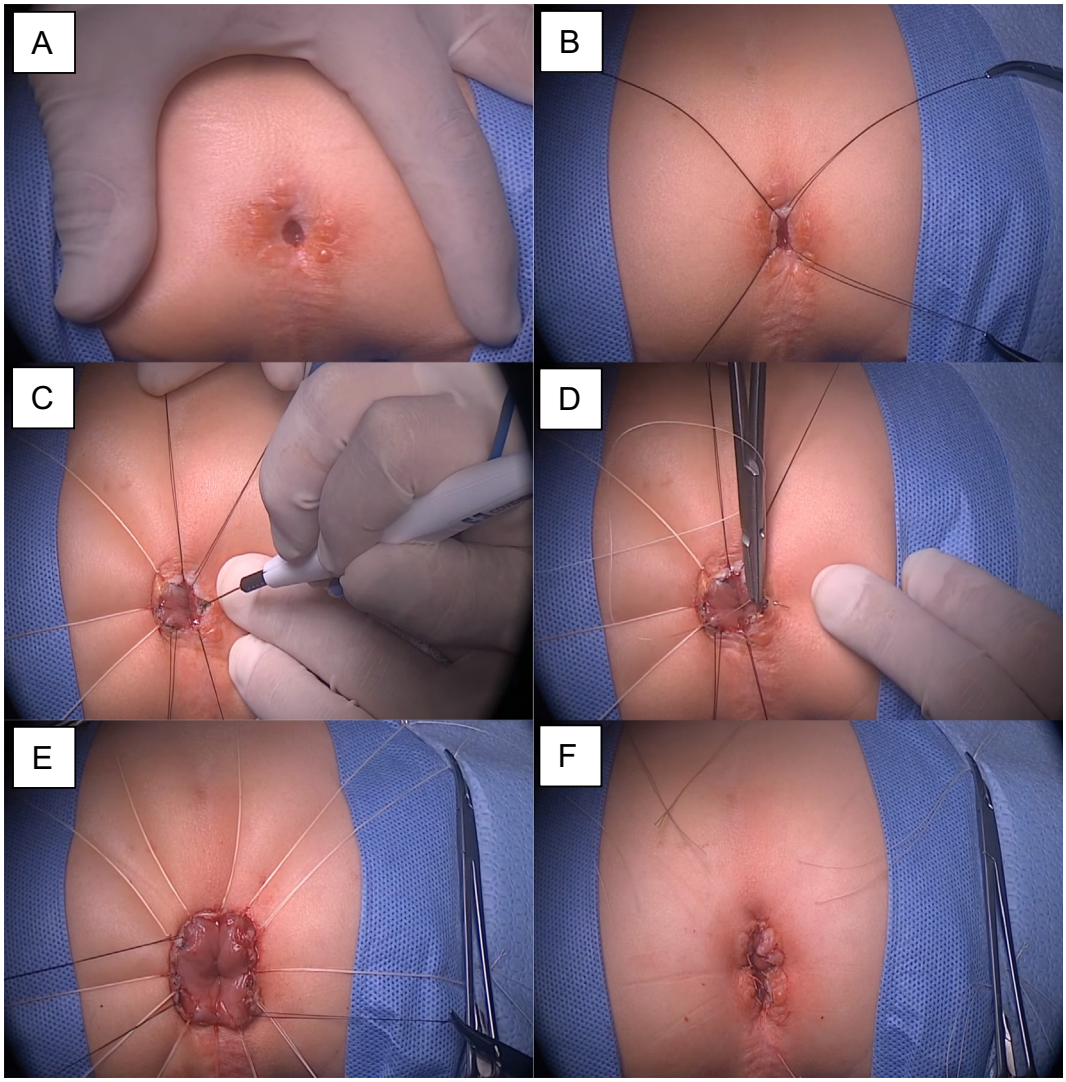

Fig. 1. Illustration of the Heineke–Mikulicz technique for skin-level stenosis after PSARP. A) Examination of the anus illustrates a skin-level stricture. B) Traction sutures are placed at the 2, 4, 8, and 10 o'clock positions. C) The skin is radially incised until the stricture releases, applying opposing tension on the adjacent traction sutures. D) The rectal mucosa is then sutured to the skin longitudinally. E) Appearance of the anoplasty after incising all four quadrants. F) Appearance of the anoplasty at the end of the procedure.

Table 1
Patient characteristics of patients undergoing Heineke–Mikulicz anoplasty from 2014 to 2017 (N = 28).

		n (%) or range
Age, years		6.1 (0.5–24.4)
Follow up, years		1.0 (0.4–2.9)
		0.8
Duration PSARP to HMA, years		(8 weeks–13.1 years)
Gender	Male	19 (68)
	Female	9 (32)
Diagnosis	Anorectal malformation	25 (89)
	Congenital anal stenosis	2 (7)
	Acute lymphoblastic leukemia	1 (4)
	Primary PSARP	8 (29)
Preceding surgery	Redo PSARP	18 (64)
	None	2 (7)
Bowel regimen preceding HMA (N = 20)	Laxatives	8 (40)
	Enemas	12 (60)
	Constipation	25 (89)
Symptoms preceding HMA	Impaction	9 (32)
	None	3 (11)

which was sustained in 97% of patients at a mean follow up of 12 months.

The median time to stricture after PSARP in our patient population was 9 months, thus our follow up is likely adequate to capture the majority of cases of restricturing. And while the majority of post-PSARP strictures occurred within a short duration (40% and 64% within 6 and 12 months, respectively), one patient was found to be strictured as

Table 2
Anatomic and surgical outcomes following Heineke–Mikulicz anoplasty from 2014 to 2017.

		n (%) or range
Anal size (Hegar)	Preoperative	8 (6–12)
	Postoperative	16 (14–18)
Intraoperative complications		0 (0)
Postoperative wound infections		0 (0)
Postoperative wound dehiscence		0 (0)
Combined procedure		5 (18)
Same day discharge		23 (82)
Recurrent stricture		1 (4)
Redo HMA		1 (4)
Return to normal activities	<1 day	9 (45)
	2–3 days	6 (30)
	4–7 days	5 (25)

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Table 3
Functional outcomes following Heineke–Mikulicz anoplasty from 2014 to 2017.

		n (%)
New symptoms	Obstruction/constipation	1 (4)
	Incontinence	0
Severity of constipation and/or frequency of impaction	Significantly improved	11 (55)
	Slightly improved	5 (25)
	No change	4 (20)
	Slightly worsened	0 (0)
	Significantly worsened	0 (0)
Laxative (n = 8)	Decrease laxative dose	2 (25%)
	Able to discontinue laxatives	2 (25%)
Enema (n = 12)	Flushes easier/more effective	11 (91)
	Able to transition to laxatives	2 (17)
"Would you repeat dilations?"	Yes	3 (15)
	No	17 (85)

late as 13 years after the preceding procedure. The diagnosis of anal stricture should be considered in any patient presenting with constipation or impaction and a history of PSARP, so the anus must routinely be examined. However, it is critical to differentiate between skin-level strictures and deeper strictures as the management differs substantially, the latter requiring a formal reoperation [1].

A thorough physical examination of the anus is usually sufficient to confirm the diagnosis. We recommend both a digital exam (as a stricture can be more easily palpated) and a check with Hegars for the size of the anal opening. In patients who will not tolerate examination in the outpatient setting, an examination under anesthesia (EUA) may be needed. A dilation may be performed for superficial strictures at the time of EUA, although the resulting trauma to the tissue may induce further fibrosis and stricturing [16]. In this setting, HMA may be useful to avoid this vicious cycle.

Similar to the subset of postoperative PSARP strictures amenable to HMA, some rare cases of congenital anal stenosis patients were amenable to repair with this same procedure. Both patients with congenital anal stenosis in our experience had stenoses involving only the skin-level confirmed on EUA that persisted despite dilations. For cases of congenital anal stenosis deeper than the skin level, we leave the distal anal canal untouched and mobilize only the posterior rectum. We then anastomose the posterior rectum to the skin [17]. The benefit of HMA in these children is that it avoids any mobilization of the posterior rectum as well as a diverting colostomy that is needed to repair a more typical case of congenital anal stenosis [17,18].

One child in our series experienced restricturing after HMA and required a second anoplasty. This child had multiple anorectal procedures related to his ARM prior to adoption, after which he underwent a third PSARP at our institution followed by a prolapse repair 6 months before developing a stricture, which required HMA. He underwent a second HMA 6 months after the first. We suspect that his restricturing was related to poor vascular supply of his distal pull-through owing to the multiple prior procedures performed. It is interesting to note that the other 17 children who presented with strictures after redo PSARP did not experience this complication. It is therefore important to continue to study this population of patients in order to identify patterns that might predict restricturing and develop approaches that may avoid this complication.

To better understand patient reported experience and outcome measures related to the procedure, we contacted parents at a minimum of 6 months postoperatively and asked a series of follow-up questions. When asked to rate the impact of the HMA on their child's severity of constipation or frequency of impaction on a 5 point Likert scale from "significantly improved" to "significantly worsened," the vast majority of parents indicated that their child's symptoms had improved, and no parents believed that the symptoms worsened after the procedure. It is important to note that stooling is a complex interaction in these patients and the ability to achieve continence depends largely on the type of original malformation, the degree of sacral development, and any spinal anomalies [19]. Thus, while an adequately sized anus is necessary to achieve social continence, the degree to which an HMA will relieve stooling difficulties is based entirely on the degree to which the stenosis accounts for the patient's symptoms. Of note, no parents complained of new-onset incontinence or the development of new stooling problems postoperatively. Interestingly, nearly all of the parents whose child was on enemas preoperatively noted that their child's flush flowed more easily after HMA, and 2 patients were able to transition off enemas entirely and onto laxatives. For patients on laxatives, one half of patients were able to decrease the laxative dose or discontinue laxatives altogether.

The practice of routine dilations has recently been called into question [4,5], citing the risk of anastomotic disruption along with the pain and psychological trauma to both the patient and the caregiver associated with this practice. In a study of patients with ARM, those who underwent dilations in childhood experienced dissociative symptoms in adolescence and adulthood that correlated with the duration of the practice [20]. Admittedly, as routine dilations remain the standard of practice in most institutions at present, the stricture rate after PSARP without dilation and therefore the influence of dilation on the rate of stricture formation are not known. This question is currently under investigation by the Pediatric Colorectal and Pelvic Learning Consortium (PCPLC) [21] in the form of a randomized controlled trial of dilation vs. nondilation after primary PSARP.

The vast majority of parents in our cohort indicated that they would choose to forego dilations, citing the severe trauma associated with the practice along with the relative ease of the HMA. It should be noted however that this cohort represents a specific subset of patients in whom dilations were not successful and thus were likely more traumatic than a typical case. We believe the HMA could provide a useful "back-up" for those patients who do not receive dilations and later form strictures, and may even be preferable in some cases to routine dilations, particularly in older patients.

Little is known overall about the optimal management of anal strictures in children, and much of the available data are derived from studies performed in adults with anal strictures, generally related to hemorrhoidectomy or fistulectomy. Given the geometric properties of the anus, we perform this procedure in 4 quadrants, which allow for a maximal concentric stricture release with minimal dissection. Alternative techniques, such as the Y-V anoplasty [14] or diamond flap anoplasty [13] have been described. The HMA described here does not require any mobilization of the rectum which could further devascularize the tissue and leave it prone to further stricture. The procedure is performed with minimal mobilization of tissue, and the superficial electrocautery (usually 3–5 mm) that is required to release the stricture lessens the chance of damage to the sphincters. We therefore believe that the HMA offers technical benefits over other described techniques. It is also quick, can be performed in the outpatient setting, and has a high success rate as demonstrated in this report.

The major limitation of our study relates to its retrospective nature and the relatively small number of patients that are investigated. Although stricture is a relatively common complication after PSARP, only a subset of strictures (the very thin ones) is amenable to HMA. We also do not know the denominator, or how many patients after PSARP would do well with no stricture either with or without a dilation protocol. It is therefore difficult to study this technique prospectively, particularly against other techniques, which limits our ability to claim superiority of the HMA. Additionally, 8/28 (29%) patients were unable

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to be reached for additional information regarding their experience with the procedure, thus possibly introducing a degree of selection bias to our results. Despite these limitations, we are encouraged by the success of this procedure and recommend it to any surgeon for the management of skin-level strictures.

4. Conclusion

The Heineke–Mikulicz anoplasty is a simple procedure that can be performed in the outpatient setting to treat skin-level anal strictures after primary and redo PSARP and in select cases of congenital anal stenosis. Our results suggest that this procedure has good durability with a low recurrence rate.

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