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ORIGINAL ARTICLE

Bowel vaginoplasty in children and young women: an institutional experience with 55 patients

Sundeep Kisku[1] · Lilly Varghese[2] · Aruna Kekre[2] · Sudipta Sen[1] · Sampath Karl[1] · John Mathai[1] · Reju Joseph Thomas[1] · Ravi Kishore[1]

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Abstract
Introduction and hypothesis Absence of a vagina owing to congenital Mullerian defects or other acquired causes requires reconstruction of the female genital passage. We present our experience using various bowel segments.
Methods Bowel vaginoplasty was performed in 55 patients from January 2004 through May 2014 for cervicovaginal atresia (20), Mayer–Rokitansky–Küster–Hauser (MRKH) syndrome (20), distal vaginal atresia (8), cloaca (2), cervical atresia (1), complex urogenital sinus (1), transverse vaginal septum (1), rhabdomyosarcoma of the vagina (1), and traumatic stricture of the vagina (1). The bowel segments used were sigmoid (50), ileum (2), anorectovestibular fistula (2), and loop rectovaginoplasty (1).
Results Thirty-nine patients who had the proximal vagina or uterus anastomosed to the bowel segment reported regular menstrual flows. Three patients are sexually active with satisfactory coital function. None of our patients developed pyometra. Five patients had neovaginal mucosal prolapse. Two patients had severe stenosis requiring excision of the neovagina. Seven patients had mild stenosis requiring dilatations in 6 patients and V-Y meatoplasty for 1 patient. One patient had a descending colon anastomotic leak requiring a diversion ileostomy.
Conclusions Genital reconstruction with bowel vaginoplasty is a highly skilled operation that provides a durable and lubricated replacement of the vagina with good outcomes. Uterocoloneovaginoplasty is a safe procedure preserving the menstrual flow in patients with a functional uterine fundus.

Keywords MRKH · Rhabdomyosarcoma of the vagina · Colovaginoplasty · Vaginal reconstruction

✉ Sundeep Kisku
skisku@yahoo.com

[1] Department of Paediatric Surgery, Christian Medical College, Vellore, India 632004

[2] Department of Obstetrics and Gynaecology, Christian Medical College, Vellore, India 632004

Introduction

The absence of a vagina may be due to congenital (Mullerian defects) or acquired causes [1, 2]. Congenital Mullerian defects include Mayer–Rokitansky–Küster–Hauser syndrome (MRKH), cervicovaginal atresia and isolated total or segmental vaginal atresia. The incidence of MRKH is 1 in 4,500, while it remains unknown in cervicovaginal atresia and vaginal atresias [3, 4]. The acquired causes are rare and include trauma and malignancy [1]. Vaginal reconstruction can be achieved by non-surgical and surgical methods. The non-surgical methods essentially create a deeper vaginal dimple by graded vaginal dilators (Frank's technique and Ingram's technique) [3]. Surgical methods of vaginoplasty include the vulvar flap (Williams), groin flaps, Vecchietti procedure (vaginal depth increased by applying pressure on the vaginal vault), Davydov's procedure (peritoneal pull through), intestinal vaginoplasty and tissue-engineered biomaterial graft vaginoplasty [5]. Bowel vaginoplasty provides an epithelially lined, lubricated passage as a conduit for menses and coitus. We present our experience with bowel vaginoplasty for various vaginal anomalies and defects.

Material and methods

Fifty-five patients required vaginal reconstruction in our tertiary care hospital over a period from January 2004 through April 2014. Ethical clearance was obtained from the institutional review board (IRB). The clinical records of these patients were reviewed for the nature of the anatomical defect, type of surgery performed and functional outcome.

The patients presented at the age of 1 month to 34 years (median 16 years). Six (11 %) patients were lost to follow-up. Forty-seven patients were followed up for a median of 32 months (1 month to 120 months).

Thirty-four patients (62 %) underwent single or multiple preoperative procedures. These included vaginoplasty (n=29), examination under anaesthesia (n=8), laparoscopy (n=8), anoplasty (n=3), sigmoid diversion colostomy (n=3), vesicostomy (n=1), salpingectomy (n=1) and urethral reconstruction with a Yang–Monti ileal segment (n=1). Two patients (1 and 3 months of age) presented with rectovestibular fistula for correction. In both these patients, the diagnosis of vaginal agenesis was made at surgery.

Bowel vaginoplasty was performed for various disorders as listed in Table 1. The bowel segments used were sigmoid (n=50), ileum (n=2), anorectovestibular fistula (n=2) and loop rectovaginoplasty (n=1). Sigmoid colon was the segment of choice in most patients as the ileum was reportedly associated with a greater risk of post-operative stenosis [2] The ileum and the other bowel segments were used for specific indications, as described later in the study.

All adolescent and adult patients with uterine agenesis underwent sigmoid neovaginoplasty. Patients with distal vaginal atresia (n=8) underwent sigmoid replacement of the vagina, bridging the proximal cuff of the vagina to the vestibule. Nine patients with Mayer–Rokitansky–Küster–Hauser (MRKH) syndrome had some functional endometrium in the hypoplastic uterus (n=4) or hemiuterii (n=5). These 9 patients and the other patients, who had a functional endometrium with an obstructed cervical outflow channel (cervicovaginal atresia, n=20, cervical atresia, n=1), had the sigmoid colon anastomosed to the uterus or hemiuterii above and the vestibule below (Fig. 1). This technique was described earlier by authors from our institution, Kanniayan and Sen, in 2009 [4]. The reconstruction was oriented antiperistaltic (n=34) or isoperistaltic (n=16) based on the surgeon's preference and the vascular anatomy. Endometriosis was noted in 14 patients of the 20 patients with cervicovaginal atresia (70 %). These endometrial lesions were ablated during surgery.

Ileal segments were used to reconstruct the vagina in 2 patients. One of these patients had a cloacal anomaly, where the sigmoid colon was used for the anal pull-through, and the other patient had rhabdomyosarcoma of the vagina.

The 2 patients, aged 1 and 3 months respectively, as previously mentioned, presented for surgical correction of an anorectal malformation. They were diagnosed intraoperatively to have rectovestibular fistula with vaginal atresia. Here, the anorectovestibular fistula (ARVF) was left in situ as the neovagina and the rectosigmoid were pulled down for a neoanus (Fig. 2). One patient with a high cloacal anomaly had loop colon rectovaginoplasty (Fig. 3) performed as described by Harrison et al. [6]. One patient presented with vaginal stricture that manifested 4 years after urethral reconstruction with a Yang–Monti ileal segment for traumatic urethral injury. She underwent sigmoid colon replacement of the strictured segment. One patient with transverse vaginal septum underwent several failed attempts at resection of the vaginal septum and vaginoplasty. She underwent perineal

Table 1 Distribution of patients who underwent bowel vaginoplasty.(N=55)

Anomaly	Number of patients (%)	Pre-pubertal		Post-pubertal		Lost to follow-up	Sexually active
		Median age (months/years)	Follow-up (months)	Median age (years)	Follow-up (months)		
Cervicovaginal atresia	20 (36)			15 (11–34)	56 (1–120)	4	1
Mayer–Rokitansky–Küster–Hauser syndrome	20 (36)	n=2 2 months (1–3)	40 (40, 41)	n=18 18 (11–32)	17 (1–108)	1	2
Distal vaginal atresia	8 (14)			14.5 (11–20)	38 (4–90)	1	
Cloaca	2 (4)			13.5 (11, 16)	34 (31, 36)		
Cervical atresia	1 (2)			15	34		
Complex urogenital sinus	1 (2)			16	57		
Transverse vaginal septum	1 (2)			15	2		
Rhabdomyosarcoma of the vagina	1 (2)	3	65				
Post-traumatic vaginal stricture	1 (2)			11	2		
Total	55	2 months (1 month to 3 years), n=3	41 (40–65)	15 (11–34) n=52	31 (1–120)	6	3

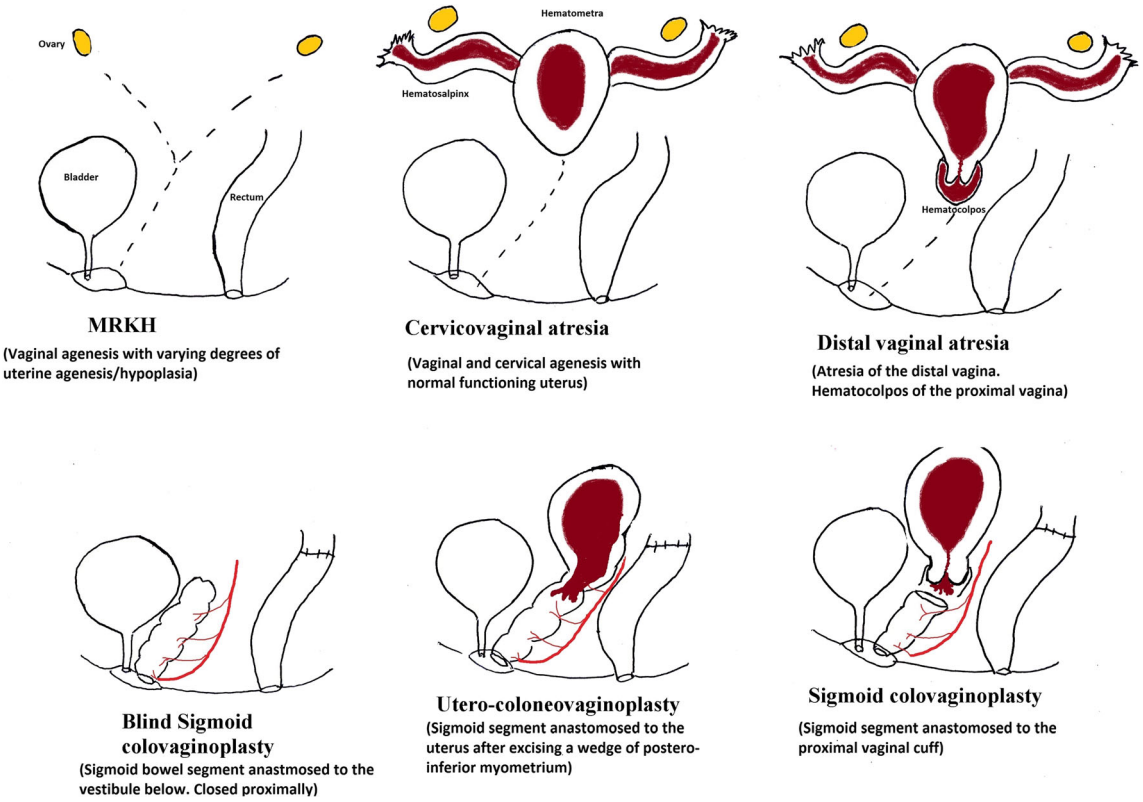

Fig. 1 Colonic conduit for the various disorders

exploration and laparotomy with excision of the septum and the fibrous scar along the posterior wall of the vagina. A sigmoid bowel patch was placed onto the posterior wall of the vagina (Fig. 4).

All patients were reviewed in the outpatient clinic 2 weeks after the vaginal reconstruction. A genital examination was performed and the health and patency of the neovagina were confirmed. Patients were instructed to maintain good local hygiene by gentle washes. Patients with extensive endometriosis were placed on hormonal therapy. Sexual intercourse was allowed 3 months after surgery.

Results

Thirty-nine of the forty-two patients who had the proximal vagina or uterus/uterii anastomosed to the bowel segment reported regular cyclical menstrual flows. Three patients are sexually active and report satisfactory coital function with no dyspareunia or post-coital bleeding. One of them became pregnant and aborted at 2.5 months' gestation. None of our patients developed pyometra or sepsis. One patient requires weekly vaginal douches to avoid a bad odour.

Five patients had neovaginal mucosal prolapse requiring mucosal trimming in 3 patients and expectant management in 2 patients. Two patients had severe stenosis requiring excision of the neovagina. Seven others had mild stenosis requiring dilatations in 6 patients and V-Y meatoplasty for introital stenosis in 1 patient. A total of 12 patients are on daily precautionary self-calibration.

One patient had a descending colon anastomotic leak on the fifth postoperative day requiring a diversion ileostomy. Two patients developed wound infection requiring delayed wound suturing. Two patients (aged 11 and 14 years) had features of neurogenic bladder in renal failure. Both had painful cyclical abdominal pain due to haematometra. They were treated for the renal failure and underwent vaginoplasty to relieve them of the obstructed menses. They were counselled about the need for renal transplantation in the future. One was followed for 9 years and died while waiting for a transplant. The other has completed 10 years of follow-up and is currently being prepared for transplantation. Another patient with cloaca also died after discontinuing psychiatric counselling and therapy for severe depression and other psychological problems. The complications are summarised on Table 2.

Excessive mucous secretion was not a problem in our patients. None of the patients developed diversion colitis, ulcerative colitis or malignancy of the neovagina. All patients, except the children who had the ARVF used for neovaginal reconstruction as mentioned above, are continent for both urine and stool.

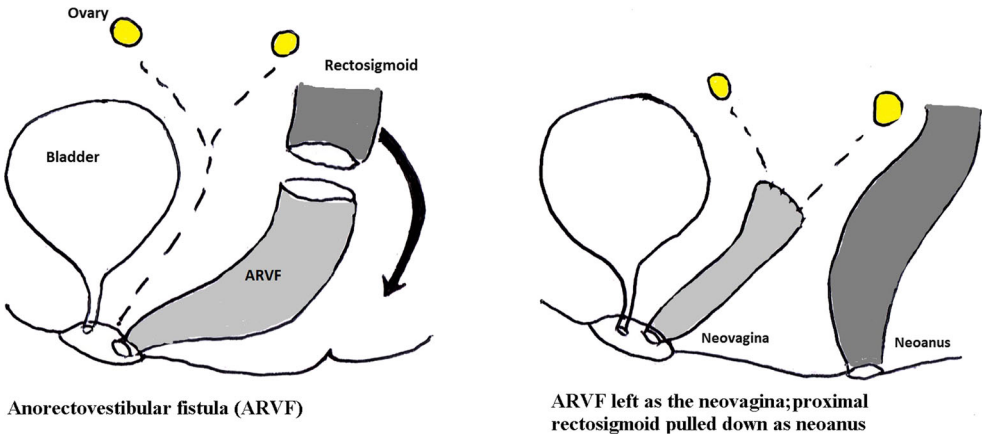

Fig. 2 Using the anorectovestibular fistula (ARVF) as the neovagina

Discussion

Vaginal reconstruction is challenging and aims to provide an anatomical and functional passage for the passage of menses, coitus and fertility. Successful treatment outcomes have improved various psychological aspects of the patient [7].

Frank's technique of creating a neovagina by dilatations has been suggested as a first-line treatment for patients with vaginal atresia as it is non-invasive, has no complications and has good functional outcomes [8]. However, the vagina is not lubricated and requires a great deal of motivation to persist in dilatation. Refusal and non-compliance regarding such a method of vaginal creation is not uncommon [9]. Patients following the popular McIndoe procedure require prolonged dilatation and night-time stenting. There have been reports of stenosis, graft shortening, dyspareunia, vaginal and rectal prolapse, and rectovaginal and rectovesical fistulae [10]. The Vecchietti procedure is suitable for those patients who have had no previous vaginal surgery. Bladder injury, perforation of the olive from the upper pole of the vagina and vaginal prolapse are some complications of the Vecchietti procedure [11]. Davydov's procedure is suitable for patients with a non-elastic vaginal dimple or previous vaginal surgery and may be unsuitable for those with previous abdominal surgery and adhesions [5, 12]. Dyspareunia and lubrication are also of some concern [5]. Experience with flaps is limited to a few cases. Flap necrosis and disfigurement of the groin have been reported [13].

Baldwin was the first to describe bowel replacement of the vagina in 1904. He finally performed a bowel vaginoplasty

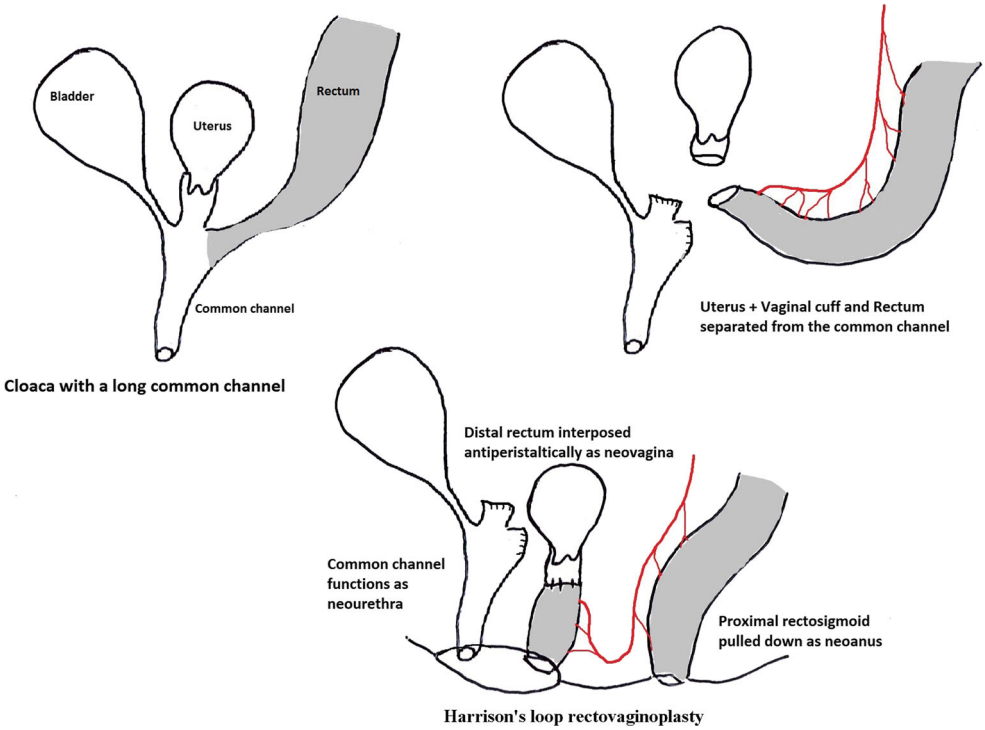

Fig. 3 Constructing Harrison's loop rectovaginoplasty

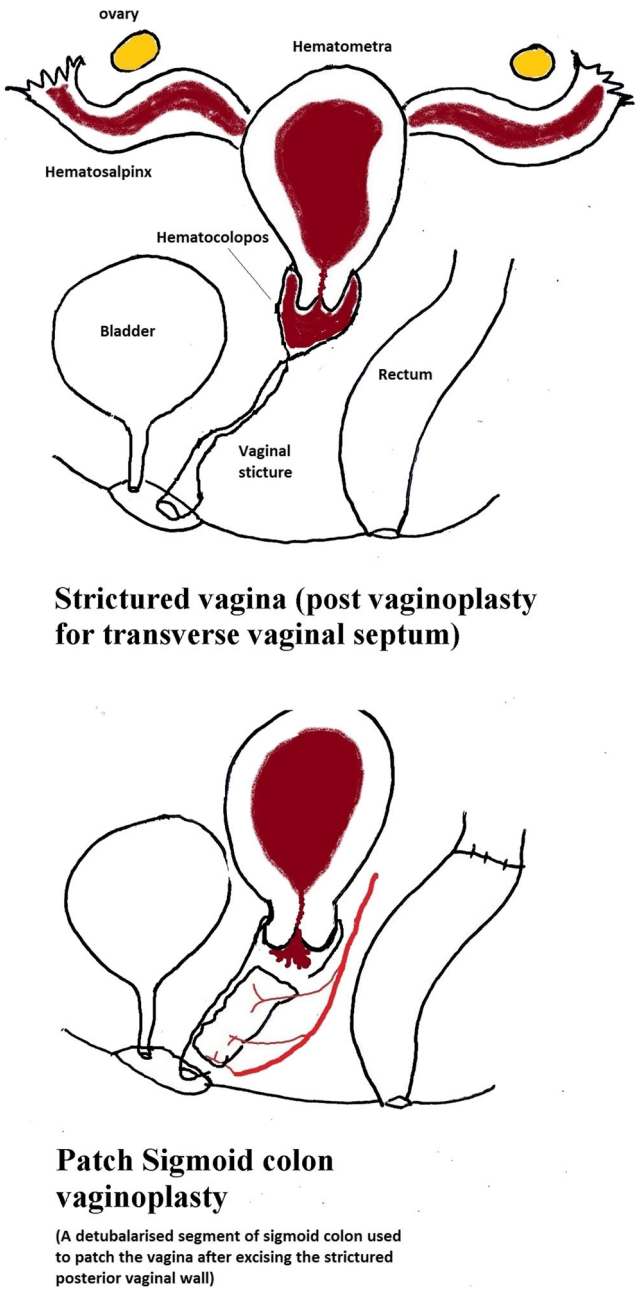

Fig. 4 Patch colovaginoplasty

using an ileal segment in 1907. Wallace performed the first sigmoid colon vaginoplasty in 1911. Subsequently, these procedures were abandoned because of a high mortality rate in the pre-antibiotic era [2]. Turner-Warwick and Kirby in 1990 reported their experience with the use of colocaecum for vaginal reconstruction [1]. Ikuma et al. reported the first laparoscopic sigmoid replacement of the vagina in 1997 [14].

Bowel vaginoplasty is a durable, vascularised, epithelially lined, self-lubricating channel that grows with the patient. Generally, there is no need for long-term stenting. Bowel replacement, however, is a complex surgery with several potential short- and long-term complications. These include excessive mucosal discharge, bowel obstruction, anastomotic leaks, bladder/ureteral injury, stenosis, prolapse, polyp formation, diversion colitis, ulcerative colitis and rarely malignancy [1, 15]. Despite these complications, several authors have reported successful bowel vaginoplasties [1, 2, 4, 15, 16]. Clearly, the multiplicity of procedures for this complex anomaly indicates that there is no perfect solution. Further, there is no consensus currently on the ideal procedure.

The management of cervicovaginal atresia and cervical atresia is still evolving. There has been a paradigm shift from the historical recommendation of hysterectomy to recent research into cervicovaginal reconstruction. Genital reconstruction in this anomaly has been associated with very low fertility, significant morbidity and mortality in the past [17]. With the advances in antimicrobials and surgical techniques the morbidity in recent times has been lower [18]. This is attributed in part to the loss of the cervix and endo-cervical mucus, and also the associated genital anomalies, previous surgical procedures and endometriosis [17]. Our patients are from the middle and lower socioeconomic status. Many patients cannot afford more than a single definitive procedure. Further, ours is a conservative society where the loss of the vagina and fertility makes it very unlikely for the patient to get married [19]. The utero-coloneovaginoplasty, described earlier by Kanniayan and Sen, is tailored to meet this need—ensuring a safe, wide, epithelially lined, lubricated conduit for menses and coitus, at the cost of infertility [4]. The wide anastomosis provides free drainage of the menstrual blood, thus preventing haematometra and pyometra [4, 19]. We have performed utero-coloneovaginoplasty on 30 patients (cervicovaginal atresia, n=20, MRKH, n=9, cervical atresia, n=1).

Cervical atresia and MRKH are two different entities. Patients with MRKH have vaginal atresia with varying degrees of uterine hypoplasia, while patients with cervicovaginal atresia have vaginal and cervical atresia with a normally functioning uterine fundus [17]. About 10 % of patients with MRKH have functioning endometrium [19]. The patient in our study with cervical atresia had undergone several failed previous attempts at canalisation. Intraoperatively, the stenosed upper vagina and the hypoplastic cervix were excised and a segment of sigmoid colon was used to bridge the gap between the uterus and the lower vagina. All patients with cervicovaginal atresia successfully underwent the utero-coloneovaginoplasty and none of the patients developed pyometra or sepsis.

We concur with several authors that in patients with MRKH, Frank's technique of using vaginal dilators to create a neovagina should be the first line of management [5, 8]. Of the 20 patients with MRKH, those patients (n=9) with functional endometrium underwent utero-coloneovaginoplasty, thus allowing them to have regular menstrual cycles. Four patients had previous failed attempts at vaginoplasty and hence a blind ending sigmoid vaginoplasty was performed. Five other patients underwent primary sigmoid vaginoplasty

Table 2 Complications following bowel vaginoplasty (N=55)

Anomaly	Number of patients	Mucosal prolapse	Stenosis	Neovaginal excision	Anastomotic leak+ diverting ileostomy	Secondary suturing	Died
Cervicovaginal atresia	20	1	2			1	
Mayer–Rokitansky–Küster–Hauser) Syndrome	20	3	2	2	1	1	1
Distal vaginal atresia	8	1	2				
Cloaca	2		1				1
Cervical atresia	1						
Complex urogenital sinus	1						
Transverse vaginal septum	1						
Rhabdomyosarcoma of the vagina	1						
Post-traumatic vaginal stricture	1						
Total	55	5 (9 %)	7 (13 %)	2 (4 %)	1 (2 %)	2 (4 %)	2 (4 %)

after being counselled about the various options and non-surgical methods of vaginoplasty suggested. Following discussions within the family, the patients had opted for sigmoid vaginoplasty.

Two infants with MRKH had the ARVF left as the neovagina and the rectosigmoid pulled down as the neoanus. Intraoperatively, the uterine structures were found to be rudimentary and hence an anastomosis between the bowel and uterine structure was not performed. Both these patients had difficulty with toilet training at the age of 3 years. In addition, both patients had anal mucosal prolapse that required anal mucosal trimming. Sarin et al. described good results with ARVF as a vaginal replacement in this subset of patients [20]. Levitt et al. reported good results initially with the use of ARVF in these patients, but subsequently advised against it owing to the delay in toilet training and the need for further augmentation of the neovagina when the patient became sexually active [21]. Our experience is in agreement with that observed by Levitt et al. in that it would be better to use the ARVF as the neoanus. The neovagina may be created at the same operation, or in our opinion, at puberty, when the uterine structures can be assessed for anastomosis with the bowel segment [16].

Distal vaginal atresia is managed by vaginal pull-through if the proximal vagina can be mobilised to the perineum. Authors from our institution had previously reported 4 patients with distal vaginal atresia treated with a sigmoid bowel interposition [4]. We report a total of 8 patients (4 from the previous study) who underwent successful sigmoid colon interposition between the proximal vaginal cuff and the vestibule. In all cases, the proximal vagina and uterus were mobilised and it was determined that a vaginal pull-through was not possible because of the distance between the vaginal cuff and the vestibule.

Both patients with cloacal anomalies underwent bowel and genital reconstruction in stages. One patient required a segment of ileum to be used to anastomose the uterus to the vestibule, as the colon was used for the bowel pull-through. The second patient, was reared as a male from the birth owing to the appearance of a pseudophallus. This patient underwent a colostomy in the neonatal period for an imperforate anus and presented to us at puberty with the commencement of thelarche and painful cyclical lower abdominal pain. The patient was evaluated and diagnosed to have a cloaca, vaginal atresia, haematometra with bilateral haematosalpinx, sacral agenesis and neurogenic bladder. The patient was extensively counselled by an adolescent psychiatrist and subsequently decided to change to a female gender. She was treated with appropriate medications for the distress she now faced with a gender change. When she was psychologically fit , she underwent Harrison's loop rectovaginoplasty along with Malone's procedure and appendicular mitrofanoff, reconstructing the neovagina and neoanus from the same loop of terminal rectum [6]. She developed retraction of the neoanus and neovagina, which required regular dilatations. She reported regular menstrual cycles. She continued her psychological counselling postoperatively for 2 years and thereafter discontinued therapy. She died a year later, at her hometown, isolated, withdrawn, neglected and untreated for severe depression, psychological disturbances and progressive ill health.

One patient with complex urogenital sinus underwent excision of the left hemi-uterus and utero-coloneovaginoplasty to the right distended hemi-uterus. She developed stenosis of the vagina after 5 years of follow-up. She did not want further reconstruction and hence underwent excision of the neovagina.

One child who had neoadjuvant chemotherapy for rhabdomyosarcoma of the vagina underwent excision of the vagina and genital reconstruction with a segment of ileum. The surgeon was mindful of the fact that the child would probably require radiation therapy and develop secondary vaginal

stenosis. Hence, an ileal segment was used, reserving the sigmoid colon for use later. The child did not, however, require radiation therapy and is doing well 5 years later. Hensle et al. reported 3 patients with rhabdomyosarcoma treated successfully by bowel vaginoplasty [2].

One patient with a transverse vaginal septum who underwent several failed attempts at resection of the vaginal septum and vaginoplasty was treated by a colonic patch to the posterior wall of the vagina. She is menstruating regularly and has been followed up for 9 months at the time of writing. We believe that this is the first published report of a patch of sigmoid colon used to treat recurrent vaginal stenosis.

Sigmoid colon has been the most popular segment of bowel used for vaginoplasty. Ileum, caecum and the ascending colon have also been used. Ileum as a vaginal replacement has been associated with a higher risk of stenosis [2]. In addition, ileal segments produce copious mucous, which is not as lubricating as the colonic mucous, leading to dyspareunia. Post-coital bleeding also occurs with the ileum owing to a more fragile mucosal lining [1]. Ileum is a good alternative to sigmoid colon when the latter is unavailable. We have used it in 2 patients with good results.

Vaginal stenosis following bowel vaginoplasty has been described by several authors [1, 2]. While most patients were treated with dilatations, some required V-Y vaginoplasty for introital stenosis [1], and severe cases have required excision of the neovagina [1, 2]. Most patients have stenosis of the bowel graft at the introitus. This may be due to ischaemia, which results from constructing the anastomosis under tension. Great care must be taken to mobilise and orient the bowel segment during the procedure.

Neovaginal mucosal prolapse has been treated by excision or fulguration [1, 2]. Anchoring the proximal bowel neovagina to the sacral promontory is thought to prevent prolapse of the neovagina. Laparoscopic promontofixation for the treatment of neovaginal prolapse has been described [22].

Bowel anastomotic leak is a rare complication often requiring proximal faecal diversion [23]. Our patient had a contained faecal leak from an unrecognised "stay suture" perforation proximal to the bowel anastomosis. Following the repair of the small tear, a protective diversion ileostomy was performed as a precaution. The ileostomy was closed 6 weeks later.

The simultaneous development of ulcerative colitis in the colon and sigmoid neovagina has been reported in a 17-year-old woman [24]. Diversion colitis has been reported in children following bowel vaginoplasty [25]. Benign and malignant lesions are very rare in the neovagina [26]. None of our patients developed diversion colitis, ulcerative colitis or malignancy of the neovagina.

Patients in whom the uterus had been preserved reported successful regular menstrual cycles. The ability to menstruate, even in the absence of fertility, is known to provide positive psychological support to the patient as she confronts challenging gender role identity issues [7]. Three patients reported satisfactory peno-vaginal intercourse with no dyspareunia. One patient reported a pregnancy that resulted in an abortion. Live births following bowel vaginoplasties have been reported [1, 2]. Excessive mucous production is not a problem in our patients and none of them requires the use of sanitary pads [1, 27]. Vaginal douche was required by only 1 patient. It may be recommended for those patients with a foul-smelling discharge. A thorough evaluation of the quality of life with regard to the urinary, anorectal and sexual functions was not carried out as it was a retrospective study.

Conclusion

Genital reconstruction with bowel vaginoplasty is a highly skilled operation that provides a durable and lubricated replacement of the vagina with good outcomes. Uterocoloneovaginoplasty is a safe procedure preserving the menstrual flow in patients with a functional uterine fundus.

Conflict of interest None.

Authors' participation Sundeep Kisku: protocol development, data collection and management, data analysis, manuscript writing; Lilly Varghese: data collection and management, data analysis, manuscript editing; Aruna Kekre: data analysis, manuscript editing; Sudipta Sen: protocol development, data management and data analysis, manuscript editing; Sampath Karl: data analysis, manuscript editing; John Mathai: data analysis, manuscript editing.

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