

Management of Vaginal Hypoplasia in Disorders of Sexual Development: Surgical and Non-Surgical Options

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Key Words

Disorders of sexual development • Surgery • Vaginal hypoplasia

Abstract

Patients with disorders of sexual development (DSD) requiring vaginal reconstruction are complex and varied in their presentation. Enlargement procedures for vaginal hypoplasia include self-dilation therapy or surgical vaginoplasty. There are many vaginoplasty techniques described, and each method has different risks and benefits. Reviewing the literature on management options for vaginal hypoplasia, the results show a number of techniques available for the creation of a neovagina. Studies are difficult to compare due to their heterogeneity, and the indications for surgery are not always clear. Psychological support improves outcomes. There is a paucity of evidence to inform management regarding the optimum surgical technique to use, and long-term data on success is lacking, particularly with respect to sexual function. In conclusion, vaginal dilators remain the cornerstone of treatment of women with vaginal hypoplasia and should be used as the first-line technique. Surgical vaginoplasty has a role in complex patients with previous failed dilation and surgical intervention, particularly those cases where there is significant scarring from previous surgery. Re-

gardless of the vaginal reconstruction technique, patients should be managed in a multidisciplinary team where there is adequate emotional and psychological support available.

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Introduction

Patients with disorders of sexual development (DSD) requiring vaginal reconstruction are complex and varied in their presentation. Enlargement procedures for vaginal hypoplasia include self-dilation therapy or surgical vaginoplasty. These interventions are offered to improve psychological and sexual outcomes. The concept of surgery for DSD conditions has become increasingly controversial in the last decade. Clinicians and patients have become involved in the debate, with strong views on both sides of the fence, with minimal evidence to inform management. There is now a consensus that vaginal dilation therapy is the first-line treatment for vaginal hypoplasia [ACOG, 2002] due to the absence of surgical risk, but success depends on the motivation of the patient, and the appropriate time to start treatment must be decided on an individual basis. Concomitant psychological support is necessary and improves outcomes. Surgical vaginoplasty methods depend on the genital configuration, pre-

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vious attempts at genital surgery and the surgeon's personal expertise and preference. There are many vaginoplasty techniques described: tension via an external traction device, peritoneal grafting, amnion grafting, skin grafting, bowel grafting, and muscle and skin flaps. Each method has different risks and benefits. The surgical risks include malignancy (in graft material), contracture leading to introital stenosis or loss of vaginal length, vaginal prolapse, dry vagina, or excessive vaginal discharge. There is a lack of evidence to inform management regarding the optimum surgical technique to use, and reliable long-term data on success is not available in the literature, particularly with respect to sexual function.

Indications for Surgery

Currently, surgery is performed in 2 instances, first to correct the appearance of ambiguous genitalia at birth, and second to reconstruct a vagina where the genital appearance is unambiguously female, but little or no natural vagina is present, the later being performed in the majority of cases during adolescence or adulthood.

Surgery for Ambiguous Genitalia

The concept underlying the surgical management of DSD conditions arises from the Optimal Gender Policy proposed by John Money in the 1950s, in which it was stated that the phenotype must match the sex of rearing for optimal gender development [Money et al., 1955]. In addition, they hypothesised that children were sexually neutral until the age of 2 and therefore any corrective surgery should take place before this time. Further arguments for the surgical correction in childhood of ambiguous genitalia included the belief that the child will be psychologically damaged by the virilised external appearance and correction before the age of permanent memory was desirable [Engert, 1989; Hrabovszky and Hutson, 2002]. Many neonates with ambiguous genitalia are assigned as females. Traditional rationale for early feminising genital surgery for the more severely virilised cases included relative technical ease of surgery, negating the need to disclose the disorder with patient, and an assumed 'one stage' procedure, with the aim of aiding parental acceptance of the child's assigned gender, and improving the psychological outcomes for the child [Crouch et al., 2004; Warne et al., 2005]. The majority of infants undergoing genital surgery will require repeat genital treatment (surgery or vaginal dilatation therapy) at or after puberty, mainly for vaginal introital stenosis but also

for cosmesis [Alizai et al., 1999; Creighton et al., 2001]. All of the indications for early genital surgery are now being re-evaluated, and new recommendations suggest delaying unnecessary genital surgery till an age of informed consent and to better individualise care [Hughes et al., 2006; Lee et al., 2006].

Surgery in the Adolescent and Adult

Gynaecologists are most often involved in the care of the adolescent patient developing ambiguous genitalia at puberty, vaginal hypoplasia or in follow-up of adults who underwent feminising genital surgery as children. In many subjects born with ambiguous genitalia, there will be an associated vaginal hypoplasia or agenesis, and the gynaecologist will need to discuss the treatment options at the appropriate time. Where childhood surgery has been performed, there is a strong possibility that repeat surgery may be required for vaginal stenosis, hypoplasia or genital cosmesis. In cases of failed dilation, surgery is also indicated. Treatment is indicated to improve psychological and sexual outcomes; however, there have been no studies to provide evidence that improvement in these outcomes are achieved, particularly in the long term, and in a number of these procedures ongoing vaginal dilatation is required.

Vaginal Dilation

Non-surgical vaginal dilation for vaginal agenesis was first reported by Frank [1938], who described the use of vaginal moulds of increasing width and length to successfully create a neovagina for 5 out of 6 women. Since this time, it has been shown to be an effective technique [Ismail-Pratt et al., 2007], although dilation treatment may take several months to achieve the final result. This regime has been criticised as 'distasteful' in interview studies with women [Boyle et al., 2005], and it has been suggested that compliance and patient satisfaction is generally low [Minto et al., 2003]. Motivation for reconstructive surgery is often based on aspirations for patient normality not just in terms of sex anatomy, but also sexual activities and experiences [Minto et al., 2003; Boyle et al., 2005; Liao et al., 2006]. Some women may perceive surgery to be a quicker and less emotionally involved approach to create a neovagina. However, post-operative surgical dilation is almost always required and thus surgery is not necessarily the 'quick fix' operation that it initially appears to be. Given the stigma attached to genital anomalies and the potential drawbacks in approaches to

vaginal reconstruction, specialist emotional and psychological support must be planned alongside expert medical care. Within such a broad approach to care provision, barriers to dilation treatment and adherence are more likely to be overcome, rendering it viable as an alternative to surgery as a first-line approach.

Vaginal dilator therapy has been shown as highly successful in a recent prospective study by Ismail-Pratt et al. [2007]. A standardised approach was used where patients were shown how to apply gentle pressure to vaginal dimple using graduated dilators for 30 min daily and attend up to 5 follow-up visits at 6–8 weeks apart with a clinical nurse specialist. Psychological support was available as needed before, during or after treatment, as were opportunities for education in sexual health and wellbeing. The success of dilator therapy was as high as 86% for achieving a normal vaginal length, and 81% of patients were able to have intercourse free of pain [Ismail-Pratt et al., 2007]. Therefore, dilation delivered in a multi-disciplinary context that is sensitive to patients' perspectives is highly successful in terms of objective measurement and ability to achieve coitus without dyspareunia and therefore should be a first-line treatment for all women with a short or absent vagina, avoiding surgical risks. Failure is usually due to patient unwillingness or inability to dilate or to anatomical reasons that prevent dilation success such as genital scarring due to previous surgery.

Surgical Techniques

Surgical creation of a neovagina is required when non-operative methods, mainly dilation, have failed or are inappropriate and likely to fail due to previous scarring or an absent vaginal dimple. With the increasing recognition of vaginal dilation efficacy, women who need an operative method are often more complex. So far, among the international scientific committee, there is no common agreement on the gold standard technique for vaginoplasty. In the dedicated literature, there is lack of standardisation among centres in terms of patient selection, outcomes and long-term follow-up. There are several reasons underlying the difficulties of finding a standardised approach to this intervention. Firstly, the variability of the medical condition leading to the need of vaginal reconstruction, such as XY females with a degree of virilisation may present with narrow android shape pelvis that might make surgery more difficult because of the restricted space available [Davies et al., 2005]. Also, women who have previously undergone vaginal or pelvic surgery who

may have a scarred perineum and inelasticity of the tissue or significant pelvic adhesions make surgery more technically complicated [Davies et al., 2005]. Comparing the outcomes of studies where prior complex procedures have been performed such as the aforementioned group, against studies where women with isolated vaginal agenesis are treated surgically without prior dilation therapy gives a biased picture of the results of the techniques reported. Secondly, individual surgeon ability and experience affects the choice of the procedure. Not least, there is still no concordance in defining and recording the main outcomes and long-term results of the different vaginoplasties. All these factors play a role in the actual impossibility of providing a comprehensive review of the proposed surgical approaches. With the limitations above mentioned, we will attempt to give an updated review of the current techniques.

Intestinal Vaginoplasty

The creation of neovagina from a segment of intestine was the first surgical attempt in this field to be reported. Baldwin [1904] employed a segment of small intestine to create a neovagina. Currently, the most common operative procedure involves a segment of distal sigmoid colon that is isolated on its vascular pedicle and then transposed down to the perineum. Small bowel segments may also be used for creating a neovagina with the added benefit of less mucous production and colitis in the graft. A bowel anastomosis is then required. This procedure is most commonly described via laparotomy; however, laparoscopic bowel vaginoplasties have also been described by Liguori et al. [2005] and Imperato et al. [2007]. More recently a robotic approach to sigmoid vaginoplasty has been illustrated by Kim et al. [2008]: the authors claim that the assistance of robotics allows higher degrees of movement and accelerates the procedure allowing free-hand sutures for the bowel anastomosis.

Long-term results evaluating sexual function are quite encouraging albeit non-validated: Imperato et al. [2007] reported that among 62 women who underwent bowel vaginoplasty 51.6% declared regular sexual activity (intercourse at least once a week) and 48.4% occasional activity (sexual intercourse less than once per week). Hensle et al. [2006] similarly reported 57 patients, of whom 78% were sexually satisfied after surgery. However, 42 of 57 patients in Hensle's study (74%) and 58 of 62 (94%) of patients in Imperato's study were diagnosed with Meyer Rokitansky Küster Hauser syndrome (MRKH), and it is not stated in these papers whether these patients had any attempt with dilator therapy prior to surgery. Therefore,

these women may have been subject to unnecessary major complex reconstructive surgery when dilators may have worked equally well.

Complications of intestinal neovagina include stenosis of the bowel segment which appears more common when small bowel is employed [Hensle and Reiley, 1998], mucosal or entire neovaginal prolapse [Hensle and Reiley, 1998; Parsons et al., 2002; Rajimwale et al., 2004] excessive mucus production, and bleeding which can require removal of the bowel segment [Parsons et al., 2002; O'Connor et al., 2004; Rajimwale et al., 2004; Kapoor et al., 2006]. Multiple attempts at creation of the neovagina or concomitant use of bowel for other organs including Mitroffanoff formation can result in short bowel syndrome and its associated sequelae. Although the incidence is low, ulcerative colitis as well as adenocarcinoma in a bowel neovagina have also been reported [Syed, 2001; Rajimwale et al., 2004; Schober, 2007]. The advantages include a low incidence of dyspareunia, no need for post operative dilation, and lubrication.

Despite the reported success rates of intestinal vaginoplasty [Hensle et al., 2006; Imparato, 2007], it remains a complex surgical procedure with important morbidity and long-term effects; it is now commonplace to reserve this procedure for specially selected patients who pose technically difficult, for instance where there is no vaginal dimple or where less invasive approaches cannot be performed (for example complex cloacal anomalies) or have failed.

Surgical Traction Techniques

The Vecchietti Procedure

The Vecchietti procedure dilates the vagina by passive traction on an ovoid bead placed in contact with the vestibule and attached to the abdominal wall by traction wires. Increasing traction is applied to the threads and the neovagina is approximately stretched 1 cm per day for 7–8 days with a final vaginal length of 10–12 cm [Vecchietti, 1979]. The lining of the neovagina becomes re-epithelialised with normal vaginal mucosa. It was first described in 1965 via laparotomy and it is now mainly performed laparoscopically [Fedele et al., 1994; Brucker et al., 2008]. The major problem related to this procedure is the pain associated to the daily traction and the need to perform vaginal dilation after surgery to prevent neovaginal collapse. However, the results are certainly favourable. Fedele et al. [1994] reported that among 110 women with MRKH who underwent laparoscopic Vecchietti procedure 97% had normal anatomical findings and 97% good functional results at 12 months after surgery. In a

similar follow-up study among 101 patients, Brucker et al. [2008] reported that 99% of patients had an adequate vaginal length and width on clinical examination following surgery, with only 1 patient requiring repeat surgery. However, sexual function at 6-month follow-up was reported in two-thirds of patients, with only 60% reporting satisfactory intercourse without dyspareunia, 9% reported dyspareunia and 5% required either repeat surgery or dilation under anaesthetic following index surgery. Sexual function was assessed using a validated questionnaire in Fedele's study [2008] in a subset of 27 patients and compared to a group of controls 12 months after surgery. The scores obtained were similar in the surgery and the control groups.

Overall, long-term sexual function results are disappointing in the studies that report it, particularly considering that these women were largely patients with MRKH in whom dilator therapy is highly successful. It was also noted in Fedele's study [2008] that those patients who had lower sexual satisfaction scores also had lower scores in the domains of desire and arousal highlighting a major issue in this subset of patients that adequate sexual psychology is integral in management of this unique group of patients, and that surgery alone is not the only aspect in sexual function that requires treatment [Liao, 2006].

Balloon Vaginoplasty

This new and novel approach involves a retropubic Foley catheter to create a neovagina [El Saman, 2009]. A Foley's catheter is inserted laparoscopically or by laparotomy in the space between the urethra and rectum. The balloon is then inflated allowing traction to carry up the vaginal dimple above the perineal plane, and the catheter is fixed to a metal plate placed above the dressing on the abdominal wall. Progressive balloon distension is performed in the following days [El Saman et al., 2007; El Saman, 2009]. A neovagina depth of 8–12 cm has been reported on day 7 following the procedure. Authors claim that the main advantage of this technique is the neovaginal width, which can be manipulated according to balloon distension. Although the reported results are interesting, only 6 cases have been reported and there is no long-term follow-up data on this group of patients.

Peritoneal Flap

The peritoneal vaginoplasty, often referred to as the Davydov procedure, adopts the use of peritoneum to cover a newly created space. It was initially described via laparotomy [Davydov, 1969; Davydov and Zhvitiashvili, 1974], but it is now more frequently performed laparo-

scopically [Giannesi et al., 2005]. After separation of the bladder from the rectum, the pelvic peritoneum is opened at the bottom of the pouch of Douglas and the 2 borders of the incision are drawn downwards and stitched to the edges of the vestibulotomy. Then the vault of the neovagina is created performing a purse string stitch involving the peritoneum of bladder, right sided round ligament, right sided utero-ovarian ligament and the peritoneum between right ovary and rectum. To avoid collapse of the neovagina a mould is inserted until complete epithelisation has occurred or patient is asked to perform self-dilation. Complications of this technique include bladder and ureteric injuries, and vesico-vaginal fistula [Giannesi et al., 2005]. Validated sexual function outcomes were reported in Giannesi's study where 25 women were compared to controls. Six of these patients reported poor scores which were mainly related to psychological aspects of intercourse, and interestingly it was found that the scores in the group of women who had surgery were not significantly different to age matched controls undergoing the same questionnaire, highlighting the complex and supratentorial nature of intercourse and sexual satisfaction which encompasses more than simply an adequate vaginal length.

Amnion Graft

This technique has now essentially fallen into disuse although gynaecologists may come across women who have had this procedure performed and so should be aware of it. In 1934, Brindeau [1934] described a novel approach using human amnion to construct the vagina for a patient with müllerian agenesis. It has subsequently been reported by a number of other authors [Nisolle and Donnez, 1992; Mizia et al., 2006]. Amnion is collected a few days prior to the planned procedure and is stripped from the chorion and cleaned appropriately. The collected amnion is then placed around a vaginal mould, which is inserted into an incision made in the vaginal plate. The amnion and mould are left in situ for a number of days to allow the graft to take hold. Patients are required to perform ongoing dilation after surgery, and there is a theoretical risk of viral transmission through the allograft. Numbers are small in these series, and there remains no long-term functional data reported in this group of patients.

Skin Flaps

Williams [1964] described a technique of a skin flap to form a vagina in 1964, which has been modified most recently by Creatsas et al. [2001]. Essentially, the technique

involves a U-shaped incision made in the vulva, extending across the perineum and up to the medial side of the labia to the level of the external urethral meatus. The tissues are then mobilised and sutured in layers to form a pocket in the perineum to allow coitus. Patients require long-term dilation following the procedure to prevent stenosis in the neovagina. Creatsas et al. [2001] reported non-validated follow-up results of 72 patients in whom 94% reported having a satisfactory sexual life, and none of the women in this study complained of pain or dryness during postoperative intercourse. The neovagina was reported to be able to produce lubrication for intercourse [Creatsas and Deligeorgiou, 2007]. This could only be achieved if the flap was made of vaginal epithelium. Therefore, it is likely that subsequent intercourse has provided dilation of the vaginal epithelium, and, in fact, these neovaginas are actually dilated vaginal epithelium rather than the perineal skin, which has been formed into a flap. Once again, the patients in this study are largely MRKH patients, who did not have a trial of dilator therapy prior to surgery; therefore, it is likely that a majority of these women would have achieved similar results without the need for surgery.

Skin Grafts

McIndoe and Banister [1938] introduced a technique of vaginal reconstruction using a split-thickness skin graft tubularised over a plastic stent and inserted in the cavity obtained with dissection between the urethra and the rectum. A mould is then secured to the labia allowing a variable period of time for the graft to take. The major benefit of this procedure is the lower operative risk associated with avoidance of abdominal incision. Over the years, this technique has been slightly modified and remains a relatively common procedure. Different types of grafts have been used; split thickness skin graft harvested from various sites and rotational flaps (fasciocutaneous and musculocutaneous). The main disadvantages of this procedure are the visible scar, including keloids and infection [Seccia et al., 2002; Klingele et al., 2003], inadequate lubrication and high rate of graft stenosis necessitating frequent and potentially life long dilation. Long-term sexual function results have not been validated; however, sexual satisfaction rates have been reported to be between 80 and 90% [Mobus et al., 1996; Keser et al., 2005].

Autografts

Recently, a vaginal reconstruction performed with autologous in vitro cultured vaginal tissue has been described on one woman with Rokitansky syndrome [Pa-

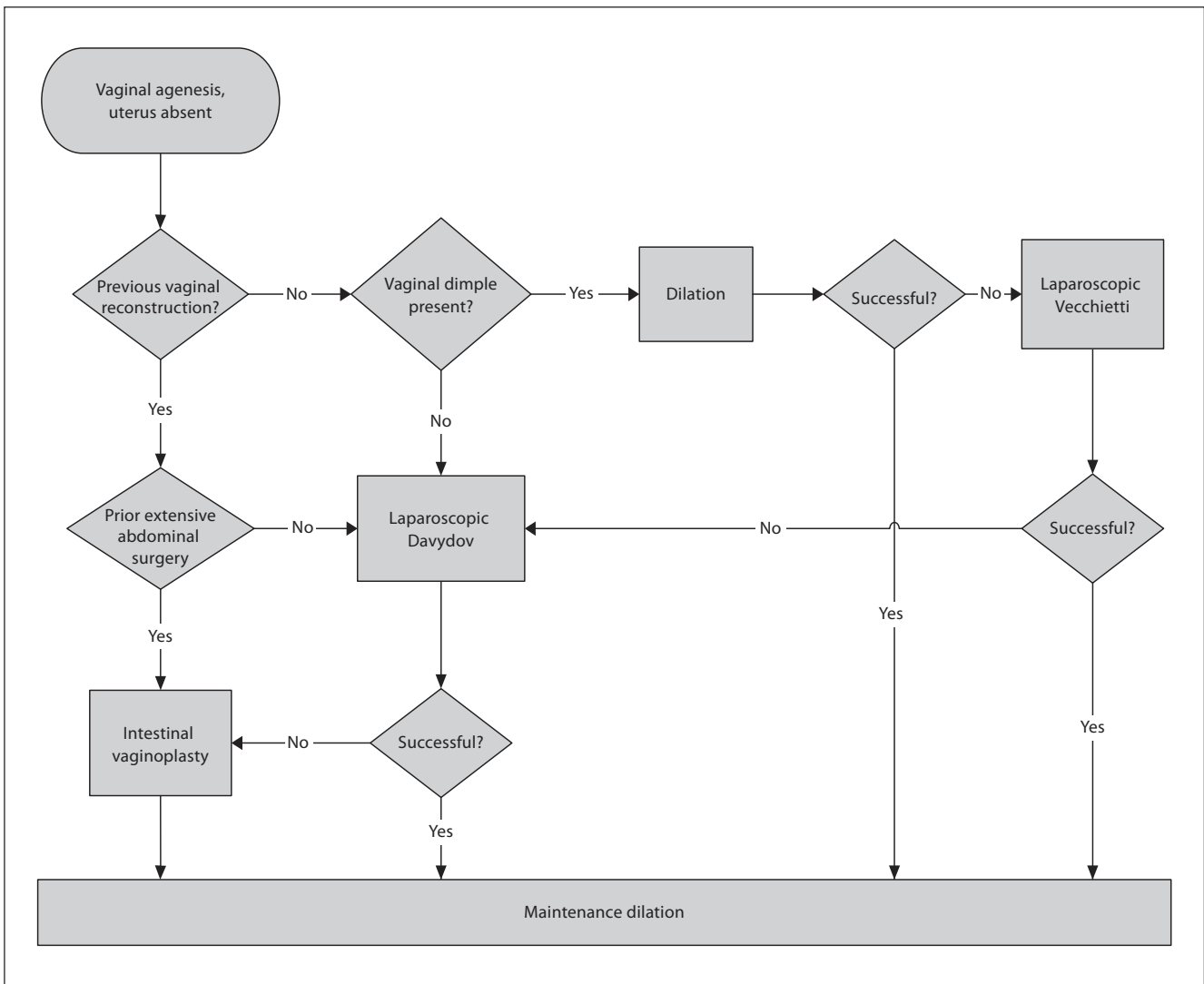


Fig. 1. London University College Hospital vaginal agenesis treatment algorithm.

nici et al., 2007]. Good vaginal function was reported at 4-month follow-up but long-term results and larger numbers are required before considering it a replicable technique.

Discussion

When comparing vaginal reconstruction approaches, it is impossible to compare the results of such a heterogeneous group of patients and surgical techniques. Surgeons will always be biased toward an operative technique that they have developed or have achieved a high level of experience in, and therefore it is likely that a large number of the patients who have been reported as having successful surgery are likely to have achieved this same goal with graduated dilator techniques. Long-term data, particularly with regards to sexual function, is lacking in the literature, and more research and follow-up is required in this field. Patients with DSD requiring creation of a neovagina are often a complex group, with physical and psychological symptoms. Therefore, it is important to individualise both operative and non-operative care in this group of patients. To aid with this process the Uni-

versity of London has developed a treatment algorithm for vaginal agenesis. This algorithm is based on the experience of the authors and is intended to provide a structured approach to the management of these patients. The algorithm starts with a decision on whether there has been previous vaginal reconstruction. If there has not, the next step is to check for a vaginal dimple. If present, dilation is attempted. If successful, a laparoscopic Vecchietti procedure is performed. If unsuccessful, a laparoscopic Davydov procedure is performed. If there has been previous vaginal reconstruction, the next step is to check for prior extensive abdominal surgery. If present, an intestinal vaginoplasty is performed. If not, a laparoscopic Davydov procedure is performed. The success of these procedures is determined, and if successful, the patient proceeds to maintenance dilation. If unsuccessful, the patient proceeds to the next step in the algorithm.

versity College Hospital Algorithm (fig. 1) has been established. This takes into consideration all relevant factors which may impact on final decision-making for clinicians contemplating vaginoplasty [Michala et al., 2007]. Dilation should always be the first-line treatment for vaginal agenesis unless there is extensive scarring or when there is no vaginal dimple.

Conclusions

Achieving satisfactory vaginal reconstructive results in women with DSD depends on many factors, including the underlying condition and previous pelvic surgery as well as the surgeon's skills and technique chosen. With high success rates reported, vaginal dilators remain the

cornerstone of treatment of women with vaginal hypoplasia and atresia, and should be used as the first-line technique, particularly in women with MRKH. Surgical vaginoplasty has a role in those women who are poorly compliant and in complex patients with previous failed dilation and surgical intervention, particularly those cases where there is significant scarring from previous surgery. Efficacies of the various surgical techniques vary, and studies are difficult to compare due to the heterogeneity of reports and patient selection. Long-term follow-up data are lacking in all studies, particularly with regard to sexual function. In the majority of cases undergoing surgery, ongoing dilator therapy is required. Regardless of the vaginal reconstruction technique, patients should be managed in a multidisciplinary team where there is adequate emotional and psychological support available.

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