Academic Year 2013-2014

Treatment of rectovaginal fistula : review of literature

Leonie VANDOORNE

Promotor: Prof. Dr. P. Pattyn
Co-promotor : Dr. D. Van de putte

Dissertation presented in the 2nd Master year
in the programme of
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Date 01/04/14

Leonie Vandoorne  
Prof. Dr. Piet Pattyn  
(promotor)

Dr. Dirk Van de Putte  
(co-promotor)
In 2012, we had to choose a subject that would determine our next two years. That’s why I was happy with my subject ‘treatment of rectovaginal fistula’. It contains both a gastrointestinal and gynaecological input, my area of interest. It was an instructive experience and it helped me to understand how to write a scientific report.

To my surprise, this two years flew by very quickly and without any unexpected obstacles. That’s why I would like to thank some persons who helped me realize this.

First, I would like to give special thanks to my supervisor and co-promotor, Dr. Dirk Van de putte, for always giving me valuable advice and support when needed. In the initial phase, Dr. Van de putte helped me to understand and visualize the different techniques, which was quite difficult for a lay person. The cooperation was always very smooth and with straightforward communication.

Second, I want to thank my promotor, Professor Dr. Piet Pattyn, for providing interesting articles and presentations. In the next stage, expert advice helped me to be critical and to see everything as a whole.

Lastly, my family and my friend, Joren Biebuyck, were always present to assist me in difficult moments with good recommendations and positive spirit.
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ABSTRACT

CONTEXT:
A rectovaginal fistula is an abnormal connection between the rectum and the vagina and is often a challenging problem for surgeons. In literature, there is still no consensus regarding the best treatment options for the different types of rectovaginal fistulas.

OBJECTIVE:
To assess which treatment is most suited for each type of fistula according to the etiology, size and location.

RESULTS:
A RVF is a devastating condition for the patient with a major impact on self-esteem and social relationships. Therefore, an individualized treatment approach should be available for the surgeon to treat each patient with the highest success possible. Based on evidence in literature every treatment should start with a local sepsis control by seton drainage. When sepsis is controlled, local repair techniques like an advancement flap can be attempted. If more complex or recurrent fistula present themselves, a muscle interposition with preference for the gracilis muscle is a next option. Lastly, if all treatment fails, more invasive procedures such as a pull-through, proctectomy or definitive colostomy can be necessary. Resulting from analysis of literature, a flow-chart is proposed for simple RVF, complex RVF and RVF complicated by Crohn’s disease.

CONCLUSION:
Applying the most suited management for each RVF patient is still a challenging problem. By reviewing the different surgical techniques in literature, a treatment flow-chart is proposed. By suggesting this flow-chart, the choice of management can be more evident. This decreases the heterogeneity in different treatment options which in turn results in more experienced surgeons and more possibility for high quality studies. There is more need for future research to improve the success rates of described techniques or to develop new techniques to get better results.

Dutch Abstract:
Een rectovaginale fistel is abnormale connectie tussen het rectum en de vagina. Het is eerder een zeldzame conditie maar het gaat gepaard met een grote weerslag op het zelfbeeld, de kwaliteit van het leven en de seksualiteit. Door deze grote psychosociale impact is het belangrijk om deze vrouwen te helpen met de beste mogelijke behandeling. Tot nu toe is er
1. INTRODUCTION

A rectovaginal fistula is an abnormal connection between the rectal and vaginal cavity that results in passage of rectal contents through the vagina. These fistulas are rather uncommon and account for less than 5% of all anorectal fistulas. A RVF is mostly acquired and only rarely caused by a congenital malformation (1, 2). The most common etiological cause of a RVF is obstetrical trauma. Other important etiologies will be discussed below (1, 3).

The vaginal passage of stool and gas causes both physical and psychosocial symptoms. A RVF has a big impact on self-esteem, intimacy and long-term relationships. Furthermore, this condition commonly occurs in the background of other disabling diseases e.g. Crohn’s disease or malignancy (4). Recognizing and acknowledging the problem is therefore very important. A detailed history and physical examination will contribute to the diagnosis (1, 4).

Owing to the major impact on the quality of life of the patient, an appropriate treatment should be implemented. This is where the biggest problem presents itself. Until now, there is no universally accepted treatment option for RVF. Medical literature consist mostly of case reports and case series, due to the small number of patients with rectovaginal fistula. Several treatment possibilities are described, including local repairs, tissue transfer techniques and abdominal operations (1, 3). The success rates after surgery are disappointing and the surgeons often face recurrent fistulas. The success of surgery varies depending on numerous factors.

---

1 RVF = Rectovaginal fistula
factors such as the etiology of the fistula, health of the surrounding tissues, presence of inflammation, infection, scar tissue, patient comorbidities and type of surgical repair (1).

The aim of this paper is to establish the most adequate surgical technique adapted to different types of rectovaginal fistulas. Applying a more individualized approach for each patient results in better success rates and less recurrent fistulas. This will be illustrated by a flow chart that depicts different treatment techniques adapted to each kind of fistula.

2. METHODS

Online databases were consulted to obtain good articles. First Mesh was used to determine which terms are adequate. By selecting different subheadings and by adding the term to the ‘Pubmed search builder’, an extensive and detailed search could be done. Different terms were used, dependent on the content we wanted to deepen. Articles about several subjects were searched: first general articles about rectovaginal fistulas, second the different etiologies (acquired and congenital), thirdly the methods to evaluate and make the diagnosis of RVF and lastly the biggest and most important part, the different treatment options.

The most commonly used databases were Pubmed, Google scholar, Web of science, Web of knowledge, Springer, Ovid, Wiley online library,… To obtain good images or tables, SpringerImages was consulted.


Beside the online searches, useful articles and presentations were received from Prof.Dr. P.Pattyn.
3. RESULTS

3.1 CLASSIFICATION

The majority of classification schemes use etiology, location and size as criteria to classify rectovaginal fistulas as simple or complex (5). A scheme based on the underlying cause of the fistula is the most useful classification for the physician, as it takes into consideration integrity of the local tissue and the health of the patient. Another possibility is based on location, classifying fistulas as being in the low, mid, or high region of the rectum (Fig. 1). Low fistulae are located slightly above the dentate line, high fistulae behind or near the cervix and mid-level is located between the high and low sector. This scheme pays special attention to the location of the fistula in relation to the underlying sphincter muscle. Fistulae are considered small if the size is less than 0.5 cm, medium 0.5-2.5 cm and large when >2.5 cm (6, 7). Simple rectovaginal fistulas mostly consist of small, low fistulas secondary to infection or trauma. These have generally healthy, well-vascularized surrounding tissue. RVF are considered complex if they are large, high, recurrent or caused by inflammatory bowel disease. They are associated with tissue scarring and decreased blood supply (5).

Fig. 1 Low, mid and high RVF (modified from(8)).

3.2 ETIOLOGY

3.2.1 Acquired

3.2.1.1 Obstetrical injuries

In up to 88% of published series, obstetrical injuries are the most common cause of rectovaginal fistulae. RVF occurs in 0.1% of vaginal deliveries in western countries (4, 6). Such fistulas present either immediately postpartum from failed recognition of a fourth-degree injury or in 7 to 10 days following an apparently normal repair. In such instances, the fistula
occurs either from infection of the wound or from breakdown of the repair (6). In Western countries, risk factors associated with severe perineal lacerations are primiparity, anatomical or physiological abnormal perineum shape, midline episiotomies, increasing birth weight, particular presentation and the use of vaginal forceps (4, 9). Approximately 3-5% of vaginal deliveries results in a third- or fourth degree tear, which, when repaired immediately at the time of labor, heals completely in 90-95%. Some fistulas that occur following unsuccessful repair may heal spontaneously. Consequently Saclarides et al. suggests that a period of observation before attempting reoperation is worthwhile. In the end, 1-2% of third- to fourth-degree perineal lacerations following labor will lead to a persistent rectovaginal fistula. A possible associated injury of severe laceration is a sphincter defect which can result in incontinence. Before embarking on a repair of the fistula, a careful continence evaluation should be performed (6).

Other obstetrical cause of RVF is prolonged labor that results in necrosis of the rectovaginal septum (5). In developing nations with limited access to obstetric care, protracted obstructed delivery results in stillbirth with vaginal wall and perineal body pressure necrosis that leads to RVF formation. RVFs are endemic in sub-Saharan Africa and South Asia, with an estimated incidence of 50,000 to 100,000 new cases annually and a suggested prevalence of 2 million (4).

3.2.1.2 Inflammatory bowel disease

IBD, specifically Crohn’s disease, is the second most common cause of rectovaginal fistulas and should be suspected in any instance where a repair has failed (6). The cumulative incidence of all perianal fistulas in Crohn’s disease is 33% after 10 years and 50% after 20 years; up to 9% of these are RVF’s (10). The incidence is thought to be proportionate to the frequency and severity of large-bowel inflammation (4). Several studies reported a 9-10% incidence of rectovaginal fistula in patients with Crohn’s disease (3). In literature, inflammatory bowel disease is estimated to be the cause of RVF in approximately 6% to 23% (1). Because ulcerative colitis is not a transmural disease, it usually does not cause fistulae (6). Other autoimmune disorders, such as Behcet disease, have been associated with RVFs (4).
3.2.1.3 Prior surgery

Previous vaginal, anorectal or pelvic surgery can be the cause (7%) of rectovaginal fistula. Iatrogenic injury during surgery and postoperative infections are probably the main etiologic factors involved in the pathogenesis of fistula formation. Several authors have dated that, within pelvic surgery, abdominal hysterectomy has been associated with the highest risk of pelvic organ fistula (90%). The vaginal approach is less invasive and has a lower incidence of infections and is therefore less associated with fistula formation than the abdominal approach (11). Less frequent, low anterior resection and restorative proctocolectomy with an construction of an ileal J-pouch may also lead to the formation of a fistula. Other procedures, performed perineal, that predispose to RVF include hemorrhoidectomy, local excision of rectal tumors and rectocele repair (6). Overall, the incidence of RVF after operative treatment for low rectal cancer is reported to be 0.9% to 10%. Recently RVF is also a reported complication of new anorectal technique using specialized stapling devices (4).

3.2.1.4 Infections

The majority of the infections in the anorectal region are cryptoglandular or abscesses arising from the Bartholin gland, which may spontaneously drain causing a low rectovaginal fistula (5, 6). These infections are the cause in about 5-10% of the RVF (9). However, other causes must be considered, including tuberculosis, lymphogranuloma venereum, schistosomiasis and diverticulitis. In the last, the fistula is usually located in the upper rectum (high fistula) (6). It may follow perforation and it is usually found in women who have previously undergone a total abdominal hysterectomy (6).

3.2.1.5 Radiation

Radiation therapy is an important component in the therapy for several pelvic malignancies and can result in complex rectovaginal fistula formation. Adjuvant external beam radiotherapy is commonly used for the treatment of high-risk cervical cancer, endometrial cancer with lymph node metastases, rectal cancer and advanced vulvar and vaginal cancers. Radiation is acutely cytotoxic to targeted cells and cause chronic obliterative endarteritis to targeted tissues, resulting in chronic inflammation and ischemia. Interstitial brachytherapy, radioactive seeds implanted in the vaginal canal or intracavitary, deliver more exact doses of radiotherapy and can also cause RVF formation (4). Patients with gynecological malignancies treated with external beam radiotherapy and interstitial brachytherapy have a 3-years risk of
18% to develop a RVF and this was significantly higher in patients who received >76 Gy to the rectum compared to those who received <76Gy (100% vs. 7%) (12). The fistulas usually occur within two years of completion of radiation. Warning signs include the passage of bright red blood per rectum, non-healing rectal ulcerations and anorectal pain. In these situations, the physician first have to rule out a recurrent cancer as the cause of the patients symptoms (6).

3.2.1.6 Cancers
Primary anorectal, perineal and pelvic cancers, with or without surgery or radiation, can cause RVF. Invasive tumor growth can erode into luminal structures, such as the vagina or rectum, allowing fistulization. The added combination of neoadjuvant radiation and surgery to remove cancer in this region predispose toward complex fistula formation (4, 6).

3.2.1.7 Rare causes
Vaginal trauma induced by retained foreign bodies, such as pessaries, sexual object, coitus or sexual assault can result in RVF (4). Other rare causes have been reported, including faecal impaction, vaginal dilatation after radiation of the vaginal cuff and viral and bacterial infections in patients with HIV (5).

Table 1. Etiology RVF

<table>
<thead>
<tr>
<th>Etiology</th>
<th>Percentage</th>
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<tr>
<td>Obstetrical injuries</td>
<td>30-88%</td>
</tr>
<tr>
<td>Inflammatory bowel disease</td>
<td>6-23%</td>
</tr>
<tr>
<td>Prior surgery</td>
<td>7%</td>
</tr>
<tr>
<td>Infections</td>
<td>5-10%</td>
</tr>
<tr>
<td>Others (radiation, cancers, rare causes and unknown)</td>
<td>&lt;5%</td>
</tr>
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3.2.2 Congenital
Congenital causes of fistulas are situated in the area of anorectal malformations with rectovestibular fistulas. As the approach of the repair of this type of congenital fistulas differs from the adult acquired fistulas, it is considered beyond the scope of this writing and should be treated by pediatric surgeons with for instance the PSARP-technique (Posterior sagittal
anorectoplasty). Jozwik et al suggest that there is an association of congenital rectovaginal fistulas with partial vaginal atresia, and this secondary to this atresia (13). Some case reports describe RVF associated with syndromes such as Townes-Brocks syndrome and Currarino syndrome (14).

3.3 EVALUATION

3.3.1 Medical history
During the anamnesis, it’s important to determine the etiology. Prior history of anorectal surgery, complicated vaginal deliveries, radiation therapy or inflammatory bowel disease have to be inquired.

3.3.2 Physical examination
During physical examination the perineum and the anus should be well inspected (6). Stool or signs of vaginitis may be visible. Probing the tract may be very painful and is therefore not recommended (5). A bidigital examination should be performed to palpate the thickness of the perineal body and sometimes a RVF can be perceived. If the location of the fistula is not obvious with the above maneuvers, a vaginal speculum examination or a rigid proctoscopy can be performed (6).

3.3.3 Physical examination anesthetized
If there is pain or difficulty to identify the fistula, physical examination can be performed under anesthesia. This is more comfortable for the patient and easier for the physician. Possible additional techniques: after filling the vagina with water, the site of the fistula can be identified by the escape of air bubbles. If a fistula still cannot be demonstrated, a vaginal tampon can be placed while methylene blue is instilled into the rectum. After 10-15 minutes, the tampon is withdrawn and inspected for any blue staining (5).

3.3.4 Imaging
Imaging is the last possibility to identify fistulas who had not been visible or tangible with physical examination. This can be the case for high located complex fistulas. Possibilities of imaging are vaginography with a water soluble contrast medium (sensitivity : 79-100%) and the use of gastrografin or barium enema. Gastrografin and barium enema are useful, on the one hand to identify the RVF and on the other hand to obtain information about the health of the colon and rectum. But vaginography has been proven to be superior to barium enema because it allows the visualization of multiple fistulas. MR is the modality of choice for disease staging and provides an optimal depiction of fistula (sensitivity 91%)(15).
3.3.5 Additional
To evaluate the associated sphincter function, endoanal ultrasonography, anal manometry and neurophysiological testing can be performed (6). This is not often performed in primary setting.

3.4 TREATMENT
3.4.1 MEDICAL TREATMENT
3.4.1.1 Infliximab (Remicade®): Crohn’s disease
Infliximab is a chimeric monoclonal antibody against tumor necrosis factor alpha. This medicine is used in auto-immune disease like Crohn’s disease by blocking the inflammatory TNF-α. In patients with a symptomatic Crohn’s disease, seton placement and medical treatment like infliximab are the initial steps to control the sepsis before a definitive attempt at closure. The ACCENT II Study, an extensive randomized clinical trial, evaluated the efficacy and safety of infliximab maintenance in patients with fistulizing Crohn’s disease. They assigned 282 (men and women) patients with Crohn’s disease who had a single or multiple draining fistula(s). The study by Sands et al. is a post-hoc analysis of the ACCENT II study to evaluate the efficacy of infliximab therapy in women with RVF. Of the 282 patients in the ACCENT II study, 25 of 138 (18.1%) women had a total of 27 draining rectovaginal fistulas (Fig. 2). All patients received

Fig. 2 Flow-chart Infliximab therapy (10).
intravenous infusions of 5mg/kg of infliximab at 0, 2 and 6 weeks and were examined at week 10 and 14 to determine the (non)responders. The follow-up was 54 weeks. Response was defined as at least a 50% reduction from baseline in the number of all draining fistulas at week 10 and 14. Patients were randomized at week 14 to receive either placebo or infliximab, this in both the responder and non-responder group. Among the responders (n=16/25), nearly 71.4% of RVF were closed after a 3-dose induction regimen of infliximab. The infliximab maintenance treatment was more effective than placebo in maintaining a longer duration of rectovaginal fistula closure. The median duration of fistula closure was more than 3 months longer in responders receiving infliximab maintenance therapy (46 weeks) than in responders receiving placebo maintenance therapy (33 weeks). Among non-responders, additional infliximab therapy did not result in closure of the fistula. About 90% of the fistula initially (week 14) closed, but the long-term closure rates decreased to approximately 54% (week 54).

The most reported complications in the RVF group are abdominal pain and nausea. Others are fatigue, abscess, arthralgia, upper respiratory infection and headache (10).

The aim of the study of Gaertner et al. was to compare the outcome in patients with Crohn’s disease who underwent RVF surgery with or without infliximab infusion. Immunosuppressants such as tumor necrosis factor inhibitors are effective in patients with Crohn’s disease, but their effectiveness in RVF patients is still controversial. Twenty-six of the 51 patients received preoperative infliximab treatment. Even though there was a trend for improved healing associated with preoperative infliximab therapy compared with surgery alone, this was not significant. But the time to fistula healing was significantly shorter with combined infliximab and surgery. Supplementary the preoperative treatment with infliximab did significantly reduce the rate of proctitis from 91% to 69% at the time of surgery. Gaertner et al. mentions the important fact that infliximab has been associated with early cutaneous closure which can result in abscess formation and refistulization (16).

Parsi et al. studied the association between the type of fistula and complete response to infliximab in patients with fistulous Crohn’s disease. The rate of response to infliximab was significantly lower among the patients with RVF (14%) compared to those with other perianal fistulae (78%) (17).

As a conclusion, the ACCENT II Study was very optimistic with excellent outcomes for the infliximab therapy. Though further research and other articles suggest that this therapy is not really suited for RVF but more adequate for other types of fistula complicated by Crohn’s...
disease. The use of infliximab in the preoperative treatment may be beneficial as it reduces the active proctitis. Though, the side-effects of the treatment are not negligible.

3.4.1.2 Other
Other drugs like cyclosporine, azathioprine and 6-mercaptopurine are used in the treatment for Crohn’s disease, but there is no evidence in literature for the treatment of RVF (10).

3.4.1.3 Future for Crohn’s patients : Stem cells?
Recent studies have shown that stem cells, obtained from different tissues such as adipose tissue, can differentiate into muscle cells. This is based on the fact that human adipose tissue contains a population of supposed mesodermal lineage stem cells with adipogenic, myogenic or chondrogenic potential. There is one RVF case, reported by Garcia-olmo et al., that showed a successful healing (follow-up : 3 months) of the RVF after an autologous stem-cell transplantation with a lipoaspirate as source of the stem cells (18). This was the only article that reported on RVF, but more studies are published about the use of this stem-cell transplantation for general Crohn’s fistulas. For example, a recent study in 2013 from Lee et al. reported a 82% healing rate in 33 patients with a follow-up of 1 year. The fistula tract was filled with autologous stem-cells combined with fibrin glue, and if the tract was not closed after 8 weeks, they reinject the tract with a larger amount of cells (19).
In the case of refractory RVF in Crohn’s patients, this new method is promising and simple and can be tried before attempting more invasive procedures.

3.4.2 MINIMAL INVASIVE PROCEDURES
3.4.2.1 Seton drainage
Local sepsis, especially for Crohn’s patients, can be treated with surgical drainage of the abscess and placement of a noncutting seton suture (Fig. 3). This suture may be left in place for circa 3 months and may be all that is required to treat the patient’s symptoms (6). The study by El-Gazzaz et al. did not show a significant benefit, but shows a trend toward increased healing with seton use (p=0.008) (20). The seton will not result in the closure of

Fig. 3 Seton drainage (modified from (9)).
the fistula but contribute to the clearing of inflammation by providing continuous drainage and promotion of fibrosis in the tract (6, 9). At some point removal of the seton should be considered to see if the fistula will close spontaneously. Neal Ellis et al. report recurrent perianal sepsis in approximately 33% of the Crohn’s patients (21). Litza et al. examined the use of seton prior to transanal advancement flap repair. The healing rate in patients without seton drainage was 63% compared to 67% in patients with seton, thus not improving the outcome of TAFR. Important in this study is the exclusion criteria: patients with rectovaginal fistula or a fistula due to Crohn’s disease were excluded (22).

3.4.2.2 Fibrin Glue (Tissucol ®)

Fibrin sealant is a surgical hemostatic/adhesive material that is being utilized in a variety of surgical situations. In practice, it is a two-component system in which a solution of concentrated fibrinogen and factor XIII are combined with a solution of thrombin and calcium in order to form a coagulum. The use of glues and plugs is based on the hypothesis that they act as a matrix or scaffold to allow infiltration of the patient’s native connective tissue (23). Loungnarath et al. describes the technique for the use of fibrin glue treatment in complex anal fistulas. This study includes a total of 42 patients of which 3 patients with rectovaginal fistulas. Both the external and internal openings of the fistula were identified and the tract was slightly debrided. They introduced a flexible catheter into the tract through the external ostium and advanced to the internal opening. Then fibrin glue was injected into the fistula, beginning with a clot at the internal opening and slowly filling the tract as the catheter was withdrawn to form another clot at the external opening (24).

This simple procedure is safe, painless, repeatable and without significant morbidity or complications. Many authors have published their experience with fibrin glue treatment of perianal fistulas but the success rate in this study was not as high as reported by other authors (60-85%): a low overall-healing rate of 31 percent. The reason for these poor results may be related to the fact that only patients with complex and chronic fistulas were included. There was one successful outcome in three patients with rectovaginal fistulas. Loungnarath et al. pose that because of no complication related to its use, fibrin glue treatment should be proposed as a first-line treatment for complex anal fistulas (24). Rivadeneira et al. studied
various reports of treatments with biological agents for anorectal fistulas. In their experience these biological agents as fibrin glue and surgisis anal plug tend to work when the fistula tract is long, which is usually not the case in RVF where the tract is extremely short. Therefore they are not enthusiastic about this approach (3).

3.4.2.3 Bioprosthetic plugs (Surgisis ® anal fistula plug, Cook Surgical, USA)

Gasjek et al. studied the long-term efficacy of the button fistula plug. They used the Surgisis Biodesign button plug (Fig. 4) as a biologically absorbable xenograft made from the submucosa of porcine small intestine. It allows repopulation by the recipient’s cells and remodelling of the tissue. It has inherent resistance to infection, and produces neither foreign body nor giant cell reactions. Two weeks after placement of the button in the fistula tract, the button is expected to expel naturally via the anus, whereas the plug itself would remain in the fistula tract and eventually be fully incorporated by the surrounding tissues. The long-term success rate of the Surgisis biodesign button anal fistula plug for the treatment of RVF (44%) was low (23). Ellis et al. reported the early experience with the bioprosthetic fistula plug and compared the results of advancement flaps repair for complex anal fistulas with these achieved with the bioprosthetic fistula plug. The bioprosthetic plug, made from lyophilized porcine intestinal submucosa, was used for 18 patients of which 5 had a rectovaginal fistula. The control group (advancement flap repair) consist of 51 transsphincteric and 44 rectovaginal fistulas. The porcine fistula plug technique is an ambulatory procedure. A suture is placed through the tapered end of the plug. Both ends of this suture were tied to the end of a fistula probe at the primary opening. The

---

2 A graft obtained from a member of one species and transplanted to a member of another species.
suture is pulled from the primary opening, through the fistula tract, to exit at the secondary opening. With gentle traction on the suture, the porcine plug is pulled into the primary opening of the fistula until ‘wrinkling’ of the superficial layer of the plug was first observed. Excess plug is removed and the plug is secured in the primary opening incorporating a portion of the sphincter mechanism. The results are similar to the advancement flap group, suggesting that porcine fistula plug is useful for the management of complex anal fistulas. The two groups are comparable with 2 exceptions: the follow-up was longer for patients in the control group and patients with Crohn’s disease were excluded from the advancement flap group but comprised 30% of the porcine fistula group. This percentage would be predicted to bias the study in favour of the advancement flap. The results will be validated when the experience with the plug is more widespread. Ellis et al. conclude that the study is underpowered and has a short length of follow-up, which precludes reaching any definite conclusions about the porcine fistula plug. Since the relative high cost, the porcine plug should probably be used only by those who treat a high volume of patients with complex anal fistulas, until more data is available (25). Thekkinkattil et al. studied the efficacy of the Surgisis AFP fistula plug in a wide spectrum of patients with anorectal, rectovaginal and pouch vaginal fistulae. The study shows a moderate (44%) success rate. They suggest that the complex nature of the fistulae selected may be the reason for the low success rate. Only one of the seven RVF patients was healed (26).

3.4.2.4 Electrocautery
Another possible non-surgical repair of RVF is described by Shafik et al. They treated 12 women by electrocauterisation with a success rate of 83%. This technique will not be further discussed as only one article reports this technique. Additionally, the mechanism of this procedure is not clearly explained by the authors (7).

3.4.3 SURGICAL PROCEDURES

3.4.3.1 Fistula excision – direct closure

3.4.3.1.1 Fistulotomy

Fistulotomy is the procedure to open the entire fistula tract from the internal opening to the external opening. For a severe anal sphincter defect, the technique of choice is fistulotomy with immediate repair. If the sphincter is intact, fistulotomy is generally not performed
because it will divide the anal sphincter unnecessarily with a high risk of incontinence. Therefore this technique should be preserved for very superficial tracts (27).

3.4.3.1.2 Fistula excision or fistulectomy

Fistula excision or fistulectomy contains a careful excision of the anal opening and the intersphincteric and transsphincteric parts of the fistula including the fibrotic scarring surrounding the fistula ostium. The next step is preparation and excision of the vaginal fistula ostium and the rest of the fistula tract. The direct multilayer closure technique is mostly used (28).

3.4.3.1.2.1 Transperineal Fistulectomy

Fistulectomy can be performed transperineal, using a transverse perineal incision about 4-5 cm in length through the perineum. Dissection between the anterior rectal wall and posterior vaginal wall is performed laterally around the fistula and continued proximally to several centimetres above the fistula tract. An introduced index finger can be used as a guiding structure which simplifies complete excision of the fistula tract. Thereafter, a lateral dissection of the perineal body was carried out, as well as an exposure of the puborectal muscle on both sides. The level of dissection is wide enough to easily perform a tension-free closure. After excising the fistula tract, closure can be done in two to three layers (28). The transperineal approach has the advantage that an anterior levatorplasty can be performed, interposing healthy tissue between the two repaired internal openings. However, it is technically difficult as it involves the separation of rectum and vagina, either of which may be damaged during the dissection (2). No complications are mentioned.

3.4.3.1.2.2 Transvaginal Fistulectomy

The transvaginal technique provides good exposure and is easy to perform but runs against the principle of repair proposed by Greenwald and Hoexter. Specifically, the repair should be on the high-pressure side, the rectum (2).

3.4.3.2 Advancement flap

3.4.3.2.1 Endorectal advancement flaps

The technical aspects of the procedure depends on the surgeons individual preference. But the critical part of the procedure is performed in a similar way. The transrectal advancement flap technique (Fig. 5) consists of raising a transanal flap, containing mucosa, submucosa and
circular muscle fibers, in a cephalad direction with the apex of the flap incorporating the internal opening. The base of the flap is created broader than the apex and the flap is mobilized sufficiently proximal so that it can be advanced without tension. The next step is performed by excision of the distal portion.

De Paredes et al. adds a transverse plication of the muscular layer which can be the internal anal sphincter and/or rectal muscular layer depending on the height of the fistula. The final step is lowering the flap to cover the internal opening or the muscular plication (27, 29, 30).

Fig. 5 Endorectal advancement flap technique (modified from (30)).

Hyman et al. reports no major complications and two minor complications: urinary tract infection and urinary retention (30). Few other complications are mentioned by the authors but the possible complications after a transanal approach are infection or failure due to necrosis of the flap.

Sonoda et al. (2002) reported the outcomes of primary repair of anorectal and RV fistulae using the endorectal advancement flap technique. They studied several factors that influence the outcome of the repair such as Crohn disease, fistula type, age, body surface area, and others. This will not be discussed any further since this is not RVF specific. They conclude
that the RAF\textsuperscript{3} method is effective but RVF and patients with Crohn disease are associated with a significant lower success rate, respectively 43.2% and 50%. This article is very critic and extensive and therefore very interesting but the studied population is unfortunately mixed (31).

De parades et al. (2010) reported the use of an endorectal advancement flap in 23 patients. Because failures of repair are frequent they modified their technique by adding a muscular plication to create a second layer of well vascularized tissue. They obtained a success rate of 65% (27).

**Sphincteroplasty:**

An advancement flap is frequently carried out simultaneously with a sphincteroplasty. The technique, according to Tsang et al., consists of raising a flap of anoderm, mucosa and submucosa. The retracted healthy ends of the sphincter muscle are identified, mobilized, overlayed and sutured in place in the midline. This is performed via a transperineal approach (32).

If obstetrical trauma is the cause of rectovaginal fistula formation, a traumatic sphincter disruption can be associated. The presence of incontinence can be masked by symptoms of the rectovaginal fistula. Tsang et al. analyzed the outcome of RVF repairs based on the preoperative sphincter status. Fifty-two women participated in the study, whereof 50% complained of varying degrees of faecal incontinence. Success rates were 41% with endorectal advancement flap alone and 80% with sphincteroplasty (96% with and 33% without levatoroplasty). The results were better after sphincteroplasties vs. endorectal advancement flaps in patients with sphincter defects but this could not be proven statistically. Nonetheless, Tsang et al. advise that all patients with RVF due to obstetrical trauma should undergo preoperative evaluation for occult sphincter defects. If a sphincter defect is present, the RVF should be treated with a concomitant sphincteroplasty with levatoroplasty. The success rates in literature of endorectal advancement flap is often high but this is possible because a concomitant sphincteroplasty is performed and this is not mentioned by the authors. In literature the success rates range from 78-100% (3, 32).

\textsuperscript{3} RAF = Rectal advancement flap
3.4.3.2.2 Endovaginal advancement flaps

The **transvaginal** advancement flap technique (Fig. 6), **consists of raising a vaginal flap with the incision made in the posterior vaginal wall around the fistula opening. The vaginal and rectal orifices are identified and repaired with absorbable sutures. The levator ani muscles are approximated in the midline without tension between the rectal and vaginal walls and the vaginal flap is advanced over the repaired area and attached to the perineal skin** (29).

![Fig. 6 A. The vaginal flap is raised. B. The levator ani muscles are approximated. C. The vaginal flap is advanced (modified from (33)).](image)

**Complications** after the transvaginal repair reported by Bauer et al., consists of one incisional hernia and one patient with transient dyspareunia. Dyspareunia is a possible disadvantage after the transvaginal repair because after raising a vaginal flap, the vagina can be shortened (33).

In 1991, Bauer et al. first reported the transvaginal approach for repair of RVF complicating Crohn’s disease. Vaginal repairs have been criticized in literature and shown to be associated with significant recurrence rate. The transanal approach for instance, at that time, did not seem to be superior to any other surgical approach. Bauer et al. described first the use of the vaginal flap with the interposition of the levator ani muscles in the selected group of 13 Crohn patients. The success rate was 92.3% after a follow-up of average of 50 months. They pose that the primary advantage of this technique for this group is the use of non-diseased, intact vaginal tissue with minimal manipulation of the diseased rectum. And by interposing the levator ani muscles between the vaginal and rectal wall, these two are separate and this provides support to the septum (33).

Ruffolo et al. (2009) published a systematic review on advancement flaps for RVF in Crohn’s disease where they compare transrectal versus the transvaginal approach. They included 11
observational studies with a total of 219 flap procedures. The transrectal approach is generally considered to be the best treatment because the repair is undertaken from the high-pressure side and there is no perineal wound to cause postoperative pain and deformity. Additionally, the approach doesn’t require sphincter division that can result in incontinence. However the systematic review suggests that there is no significant difference between the RAF and VAF approach as the primary closure rate was respectively 54.2% and 69.4%. Yet this systematic review has limitations related to the heterogeneity of the different techniques, the low level of clinical evidence of the articles included and the rarity of this type of fistula which makes randomized controlled trials very difficult to perform (29).

As a conclusion, the advantages of the flap procedure are: absence of a perineal wound and keyhole deformity, no deterioration of incontinence and no worsening of the patients symptoms if the procedure fails. The systematic review of Ruffolo et al. suggests that there is no difference in outcome between the RAF (Rectal advancement flap) and VAF(Vaginal advancement flap) procedure. The transrectal approach should be preferred as it approximates the fistula from the high pressure side. But if Crohn’s disease is involved, the transvaginal procedure can be chosen as Crohn’s disease complicates the repair with poor wound healing which can be related to the diseased rectum and the systemic corticosteroids (29, 33).

<table>
<thead>
<tr>
<th>Author</th>
<th>N</th>
<th>VAF success (%)</th>
<th>RAF success (%)</th>
</tr>
</thead>
<tbody>
<tr>
<td>Bauer et al.</td>
<td>13</td>
<td>92.3%</td>
<td></td>
</tr>
<tr>
<td>Tsang et al.</td>
<td>27</td>
<td>41%</td>
<td></td>
</tr>
<tr>
<td>Baig et al.</td>
<td>19</td>
<td>74%</td>
<td></td>
</tr>
<tr>
<td>Watson et al.</td>
<td>12</td>
<td>83%</td>
<td></td>
</tr>
<tr>
<td>Athanasiadis et al.</td>
<td>7</td>
<td>29%</td>
<td></td>
</tr>
<tr>
<td>Kodner et al.</td>
<td>107</td>
<td>93%</td>
<td></td>
</tr>
<tr>
<td>Ozuner et al.</td>
<td>52</td>
<td>94%</td>
<td></td>
</tr>
<tr>
<td>Sonoda et al.</td>
<td>37</td>
<td>63.6%</td>
<td></td>
</tr>
<tr>
<td>Lowry et al.</td>
<td>81</td>
<td>83%</td>
<td></td>
</tr>
<tr>
<td>Rahman et al.</td>
<td>39</td>
<td>100%</td>
<td></td>
</tr>
<tr>
<td>Hyman et al.</td>
<td>33</td>
<td>81%</td>
<td></td>
</tr>
<tr>
<td>Ellis et al.</td>
<td>29</td>
<td>62%</td>
<td></td>
</tr>
<tr>
<td>Ruffalo et al.</td>
<td>23–36</td>
<td>57%</td>
<td>56%</td>
</tr>
<tr>
<td>Pinto et al.</td>
<td>15–71</td>
<td>60%</td>
<td>56%</td>
</tr>
<tr>
<td><strong>Median success rate</strong></td>
<td></td>
<td><strong>57-100%</strong></td>
<td><strong>29-94%</strong></td>
</tr>
</tbody>
</table>
3.4.3.3 Perineoproctotomy

This **technique** includes laying open the fistula, converting it into fourth degree tear and then primarily repairing it as an overlapping sphincteroplasty. The advantage of this method is that a high proportion of women with obstetrically caused RVF also have occult anterior sphincter injuries. However, the obvious **disadvantage** is the potential compromise of the sphincter by mobilization, division, and repair in cases without sphincter injury (2). Rahman et al. choose to use the perineoproctotomy procedure when the diameter of the fistula was large (>1.5cm). The success rate for the 8 RVF patients was 100%, but two patients reported transient postoperative incontinence of flatus (34). Hull et al. compared the results of 50 patients undergoing episiproctotomy with 37 patients having the rectal advancement flap repair. The success rate was respectively 78% and 62.2% and episiproctotomy was associated with significantly better faecal and sexual function than RAF. Perineoproctoctomy offers the advantage of a simultaneous anterior sphincter repair and closure of the fistula. However, the use of this technique has not been embraced, most likely because it is an extensive procedure and requires division of perineal tissue (35).

3.4.3.4 Tissue interposition

Tissue interposition methods intend to put healthy well-vascularized tissue between two sutures. Especially for complex RVF, where fibrosis and chronic inflammatory results in decrease of blood supply to the tissue surrounding the fistula and, in turn, reduces the ability of tissue healing (36). Various muscles were used for flap interposition such as the preferred gracilis muscle or the gluteus, bulbocarvernosus, sartorius and rectus abdominis muscle (37).

3.4.3.4.1 Gracilis muscle interposition

The **technique** described by Nassar et al. **begins with a 10 cm transverse skin incision at the perineal body. Then, the rectovaginal septum is dissected to mobilize the posterior vaginal wall totally from the anorectal area. Fibrotic rectal fistulous edges are excised and the defect is sutured transversally. Wider debridement is done for the posterior vaginal wall around the fistulous opening to reach the vascular edges well. Gracilis muscle on either side may be used and bilateral gracilis is used for defects involving all posterior vaginal width. Three**
transverse 5cm skin incisions are made alongside the position of the gracilis muscle in the inner part of the thigh. This over the junction of lower and middle third and 5 cm below inguinal fold. The middle incision was created around a skin paddle. There is a discrepancy between Nassar et al. and other authors, where the technique for the thigh includes only 2 longitudinal incisions instead of 3 transverse. Vaginal defect is measured and a skin paddle is designed overlying the muscle with similar measurements between the vaginal wall.

Fig. 7 Gracilis muscle interposition (modified from (37)).

defect and the muscle origin. Hereby the mobilization should be performed until 5-6 cm before the root of the thigh with the aim of preserving the vascularisation, based on the obturator artery and vein.. The muscle is then dissected, which creates a tunnel between the incisions. The collateral vessels are ligated and the distal edge of the tendon is released from its tibial insertion. The gracilis is rotated over the neurovascular pedicle and the distal end is brought into the perineal area through a subcutaneous tunnel. Free muscle part (Fig. 7) is placed between the rectum and the vagina to a level of at least 3 cm above the fistula site. The distal end is fixed at the pubic ramus (38). After this, the skin paddle is attached to the vaginal wall defect. After placing 2 suction drains into the thigh and perineum, left in place for 2-3 days, the skin is closed (37).

Nassar and colleagues reports early complications (40 days) such as donor thigh pain, vaginal abscess, partial flap skin necrosis and donor limb DVT. Late morbidity includes dyspareunia, vaginal stricture, local recurrence and anorectal stricture. An important complication, observed at almost every patient who was sexually active after surgery is dyspareunia and decreased libido. This may be related to the flap bulk of tissue in the rectovaginal septum, posterior wall scar and vaginal dryness (37). The quality of life after gracilis transposition is described by Lefèvre et al., where they reported reduction of libido and sexual activity, increased dyspareunia and decreased vaginal lubrication. Recording to the questionnaire, most of the patients felt “less attractive” because of the physical perineal scar (39). Complications described by Wexner and colleagues are similar, including medial
thigh pain, rectovaginal septum abscess, persistent drainage from stoma closure site, wound infection and thigh cellulitis (40).

In literature, they report variable success rates (43-100%). Most of these cases are recurrent fistulas after variable number of attempted repairs. Nassar and others (37) discussed the primary repair of RVF in 11 patients. All patients had a definitive healing of their fistulas (100%). Obrink et al. reported 43% cure rate on 16 recurrent RVF following radiotherapy for cervical cancer. Zmora and others (41) reported on five women with recurrent RVF and 4 other kind of fistulas. The success rate was high (88%). Fürst et al.(38) described 12 cases with Crohn’s disease and recurrent RVF. Only one of the 12 patients, treated by Graciloplasty (Fig. 8), recurred. Wexner and colleagues (40) reported on 15 RVF and 2 pouch vaginal fistula. Graciloplasty was performed, including 2 repeated gracilis muscle interpositions.

Patients with Crohn’s disease (n=9) reported perfect healing in 33%, patients without Crohn had higher success rate (75%). Ulrich et al. (42) discussed gracilis muscle interposition for 35 cases, including 9 RVF. Fistula closure with no recurrence could be achieved in 33 patients (94%). Two cases where the repair failed, were due to Crohn’s disease but no clear data if they were RVF. So far, their study included the largest patient group. Lefèvre and others (39) discussed the results of graciloplasty in 8 patients. The overall healing success rate was 88%.

Fig. 8 Gracilis transposition (lower image) with formation of neosphincter (upper image) (modified from (38)).

Nassar et al. studied the results for primary repair, where most of the other authors studied recurrent fistulas. This could explain the higher success rate as the rate decreases with repeated repair procedures. Ulrich et al (42). concludes that the gracilis muscle flap has an adequate length, can be rotated easily into the perineum and brings well-vascularised muscle to irradiated or traumatised tissue. The advantage of the flap is its close location to the defect and good blood
supply from the branches of the profunda femoralis artery. The complication rate of the repair is low and success rate is high. **This is an excellent option for patients with complicated fistulas for whom other surgical treatments have failed.**

Underlying Crohn’s disease is associated with a higher complication rate, though the success rate is higher compared with other treatments for Crohn’s patients. Both Lefèvre et al. (39) and Wexner and others (40) emphasize the high success rate in especially Crohn’s disease patients and can therefore be considered as a primary option. This because local healthy tissue is not available and previous surgery is repeatedly attempted. Therefore, muscle or other healthy tissue interposition appears to be essential for successful outcome.

### 3.4.3.4.2 Bulbocavernosus graft interposition or Martius flap

The start of the procedure can be described the same way as the start of the gracilis interposition: perineal skin incision, mobilization of vagina and rectum and closure of the rectal and vaginal defect. Instead of using the gracilis muscle of the thigh, they obtained the Martius flap from the labia majora (Fig. 9) by making a 3cm longitudinal incision. The choice of side depends on local conditions, any previous procedures and the surgeon’s preference. The bulbocavernosus muscle and its surrounding fibro adipose tissues are dissected from the labia, preserving its posterior vascular pedicle. A subcutaneous tunnel connecting the labial and perineal incisions is created. The graft is transposed and loosely sutured in place over the rectal closure (36, 43).

The technique described by McNevin et al. includes a transperineal approach with layered rectal closure, anal sphincter reconstruction when indicated and placement of a Martius flap (44).

**Complications** reported by Cui et al. after the surgery included discomfort in the anus in all patients, tenesmus without diarrhea in 3 patients, and feeling of incomplete defecation in one patient. All these symptoms disappeared in 2 weeks after the procedure. Wound swelling occurred in 3 patients, which was caused by hematoma in one patient, and infection in 2 patients. One patient complained of mild dyspareunia (36).
McNevin and colleagues report de novo dyspareunia in 5 patients, one had a labial wound complication and 2 patients complained of faecal incontinence post-operatively. The 2 patients reporting faecal incontinence had pre-existing faecal incontinence resulting from severe obstetric related tissue loss and anal sphincter disruption. All patients became sexually active after repair with increased dyspareunia in all cases, but limited sexual activity in only 1 patient. So persistent or recurrent incontinence and/or dyspareunia are unfortunate sequels of the underlying perineal injury and its repair (44).

Cui et al. studied nine patients with RVF who were treated using an interposition of a vital bulbocavernosus graft. By the time of the last follow-up (6-48 months), the success rate was
still 100%. This excellent outcome can be partially explained by the relative simple aetiology of the patients. The incidence of IBD in China is extremely low and there was no RVF related to tumor. In the discussion of this study they discourse several surgical approaches where they pose that local direct fistula repairs and flap advancements are suitable for simple fistula, while a procedure with interposing viable tissues can be applied to repair complex fistula. Despite the series that’s small, their results demonstrate that the technique is both simple and effective, especially for recurrent RVF without anal sphincter injury. Patients with clinical or anatomic sphincter defects should be treated by sphincteroplasty with levatoroplasty.

Sixteen patients were retrospectively reviewed by McNevin et al. At a mean follow-up of 75 weeks, only one recurrent fistula was identified (success rate 93.8%). McNevin et al. concludes that the Martius flap can be used with minimal morbidity. Cosmetic result is very good and no functional deficits related to the flap was noted. They advise the use of the Martius flap for repair of appropriately selected complex rectovaginal fistulas.

Zimmerman et al. and Dr. Phang wrote a letter to the editor to discuss some critical points about the article of McNevin and colleagues. First, evidence is lacking that would support any recommendation of best management for low RVF because there are no prospective or retrospective studies that compare outcomes between various procedures. Number of patients in most of the series are very small and the quality of evidence is limited. Second, McNevin et al. did not mention several reports concerning Martius flap interposition. The conclusion, to use the Martius flap for fistula repair is mostly based on expert opinion, not on outcomes data that compare various types of procedures. Zimmerman et al. listed the several reports and outcomes of Martius flap treatment in Table 3. Two more recent articles were added. They reported their results of 12 patients after transanal advancement flap repair with additional Martius flap interposition. They suggest because of these data that Martius flap interposition is beneficial for women with a low RVF, except for those who have undergone previous concomitant transanal advancement flap repair. However, the number in each series is small and prospective randomized trials are lacking.
Table 3: Rectovaginal fistula healing rates after Martius flap interposition with or without TAFR4.

<table>
<thead>
<tr>
<th>Author</th>
<th>Year</th>
<th>No. of patients</th>
<th>Success rate (%)</th>
<th>Perineal/TAFR approach</th>
</tr>
</thead>
<tbody>
<tr>
<td>Zacharin</td>
<td>1980</td>
<td>13</td>
<td>85</td>
<td>13/0</td>
</tr>
<tr>
<td>Elkins et al.</td>
<td>1990</td>
<td>6</td>
<td>67</td>
<td>6/0</td>
</tr>
<tr>
<td>Pinedo and Philips et al.</td>
<td>1998</td>
<td>8</td>
<td>75</td>
<td>3/5</td>
</tr>
<tr>
<td>Zimmerman et al.</td>
<td>2002</td>
<td>12</td>
<td>50</td>
<td>0/12</td>
</tr>
<tr>
<td>McNevin et al.</td>
<td>2007</td>
<td>16</td>
<td>94</td>
<td>16/0</td>
</tr>
<tr>
<td>Cui et al.</td>
<td>2009</td>
<td>9</td>
<td>100</td>
<td>9/0</td>
</tr>
<tr>
<td>Pitel et al.</td>
<td>2010</td>
<td>20</td>
<td>65</td>
<td>20/0</td>
</tr>
</tbody>
</table>

3.4.3.4.3 Mesh interposition

- **Biologic mesh interposition**

The technique as described by Pye et al. and Schwandner et al. (Fig. 10) *starts with a combined transvaginal and transrectal approach. Primarily, the posterior vaginal wall is opened and the fistula tract is identified and completely excised in a combined transvaginal and transrectal approach. They performed a transrectal advancement flap repair. After this, the Surgisis™ mesh is cut to size and placed in the rectovaginal space and fixed at the levator muscle laterally and at the posterior vaginal wall superiorly and inferiorly. Finally, the posterior vaginal mucosa is reclosed over the mesh(46, 47). Surgisis™ mesh is a biocompatible mesh obtained from lyophilized porcine small intestinal submucosa. This enables host cells to replace and repair damage or defective tissue. Once extracted from the porcine small intestine, the mesh is processed to enable all cells to be removed. Only the natural matrix molecules remains, including collagen, glycosaminoglycans.

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4 Transanal Flap repair
and glycoproteins. They signal the host cells to repopulate the mesh with host tissue. The mesh is gradually replaced as the host cells rebuild and remodel weakened tissue. The mesh supports the patient’s own connective tissue, vascular ingrowth and smooth muscle growth (46, 47).

Another kind of bioprosthetic mesh described by Shelton et al. is the use of acellular cadaveric dermis graft (AlloDerm®). AlloDerm® is an acellular dermal matrix derived from donated skin tissue. The way this skin is prepared is similar to the Surgisis™ mesh and should achieve the same objective. Studies have shown complete vascular ingrowth into the matrix within 3 weeks of implantation. Because of these properties, Shelton et al. pose that AlloDerm® is less likely than synthetic mesh to become infected and eroded (48).

Postoperatively, four patients in the study of Schwandner et al. reported minor complications. These included urinary tract infection, vaginal sore and partial dehiscence of the rectal advancement flap. The mean follow-up of this study varies between 3-18 months, which is a quite short-term follow-up (46).

In literature, only few cases of Mesh interposition were reported. Schwandner et al. reports the results of a Surgisis™ mesh interposition for a total of 21 patients, prospectively enrolled in the study. Overall success rate after primary mesh procedure was 71%. They performed a reoperation for the patients where the first attempt didn’t succeed, and the success rate for this 6 patients was 75% (46). The success rate of Schwandner et al. is promising. But we should consider some things about the report. The results are based on only 21 procedures and the success rates were only provided from short-term follow-up. Especially, related to the dynamic progress of Crohn’s disease, the healing rates can only be evaluated with long-term follow-up. Schwandner et al. pose that these short-term results indicate that this technique could potentially be an alternative or enrichment to traditional procedures. In the recent series of Ellis and others, they report a lower recurrence rate. However, Schwandner et al state that this could be explained by the lower number of Crohn’s patients who were enrolled in this study (2 of 27 patients).

Pye et al. reported successful closure of one recurrent RVF. They note that the alternative mesh procedures (Prolene® mesh, Ethicon Endo-Surgery, Inc.) formed by synthetic materials do not have the qualities of Surgisis™ mesh, AlloDerm®, Permacol® Mesh, or other kinds of
biological meshes, although they are less expensive. But they suppose that the long-term benefit to the patient makes the product very cost effective (47).

Ellis and colleagues compared the results for advancement flap repair (n=44) and bioprosthetic grafts (n=27). In the advancement group and the bioprosthetic grafts group, 34% and 19% respectively of the repairs failed. Important for these results is that Crohn’s patients were excluded from advancement flap group, which bias the results in favor of the advancement group. Additionally, the bioprosthetic graft group contains most of the patients with a recurrent fistula, which also bias the results against this technique. The results published by Ellis et al. for the use of bioprosthetic grafts are very promising, especially in comparison with the advancement flap repair. But extensive investigation and longer follow-up is necessary for further conclusions (49).

Shelton et al. published two cases with persistent RVF who were treated with AlloDerm® graft interposition. Both of the cases healed completely. This high success rate can be explained by two reasons. First the follow-up period was rather short (3 months) and secondly there were only 2, healthy patients. They recommend more extensive research with bigger series for the use of AlloDerm® or other biological meshes in the setting of chronic infection and inflammation or poorly vascularized tissues. (48)

This technique can be used for the same indications as the muscle interposition grafts. Autologous flaps involve additional surgery with associated morbidity and are therefore more invasive than allografts. Miklos et al. pose that compared to autologous flaps, a cadaveric allograft is available, cost-effective and associated with minimal complications or morbidity (48, 50).
Synthetic mesh interposition

Few articles report the use of synthetic mesh as a graft. Only Walfisch et al. studied the use of a folded polyglycolic acid mesh (Dexon, Tyco, UK) in four patients with RVF. This absorbable mesh separates the suture lines and induces fibrosis and healing. With a long-term follow-up for 3-9 years and a success rate of 100% this synthetic mesh is promising. Noticeable again, the amount of patients is very small and they don’t make a comparison with other techniques. In this way the study is not very reliable and more investigation should be done (51).

Fig. 11 A polyglycolic acid mesh interposition (modified from (51)).

As a conclusion, the biologic mesh interposition can be used for the same indications as autologous graft interposition, when there is need for viable tissue between two suture lines. The disadvantage that are mentioned by several authors is the cost of a mesh. The possible benefit is the cosmetically better result and a less invasive operation. There is no good study that compares biologic mesh and synthetic mesh. Though most of the authors mention the fact that synthetic mesh is more likely to become infected, eroded and has a higher chance for recurrence than the biologic mesh (48, 50). In the comparison between synthetic and bioprosthetic meshes, the price is once again to the detriment of the bioprosthetic meshes: the Surgisis™ mesh is three times the cost of a synthetic Prolene® mesh (47).

As the cost is so high for the use of these biologic meshes ($550 sheet), there should be more research with bigger series and compared with other techniques to prove that they are really cost-effective.
3.4.3.4.4 Flap reconstructions

Nirijan et al. describes the conditions for an ideal flap. The perfect flap for any perineal reconstruction should be tissue that’s not too large, robust and should have protective sensation to maintain normal function. Ideally, it has minimal donor-site morbidity and a good cosmetic outcome. If necessary it provides a skin paddle for perineal reconstruction.

![Fig. 12 Areas of the perineum (modified from (52)).](image)

As demonstrated in figure 12, the perineum can be divided into six areas by a midline and a horizontal line drawn between the ischial tuberosity. This results in a right upper and lower, left upper and lower and central areas. In generally for perineal reconstruction, defects in the upper quadrant can be treated with donor skin flap coming from the groin and/or mons area. For the lower quadrant the donor can be achieved from the gluteal area. For the central area, the donor flap can be obtained from any of the above. The selection of the donor flap depends on the site and size of the defect. The local flaps can be moved in three ways: transposition (pudendal, mons pubis flap), rotation (lotus petal flaps) and V-Y advancement flaps (52).

![Fig. 13 Lotus petal flaps (modified from (52)).](image)

- Pudendal flaps

Flaps used in the perineal reconstruction based on the internal pudendal artery are called pudendal flaps but are often referred to as gluteal-fold flaps, lotus flaps or Singapore flaps (Fig. 14). These perforator flaps are supplied by vessels from the internal pudendal artery, with various types of skin paddle mobility and shape (53).
The operation, described by Onishi et al., begins with a longitudinal incision in the posterior vaginal wall, proceeding from the vaginal introitus toward the rectovaginal fistula. Both the fistula endings are carefully excised and the orifices are closed, with special attention for the anal sphincter. A gluteal-fold flap is designed triangular or diamond-shaped around either the right or the left gluteal fold and after incision, the distal side of the flap was elevated from the subcutaneous fat layer. The proximal subcutaneous flap is made thicker to avoid injury to the pedicle vessel. After securing the pedicle vessel of the flap, a perforator of the internal pudendal artery, dissection is advanced until the flap could be elevated. The distal area of the flap is made thinner up to 5 mm because the flap does not become bulky in the limited space and it can be easily sutured to the surrounding vaginal mucosa. The proximal skin portion of the flap is denuded and after creating a tunnel to the vaginal introitus, the flap is transferred and attached to the posterior vaginal wall. Direct closure of the donor site is performed primarily (55). Kosugi et al. performed the gluteal-fold flap the same way as Onishi et al., except for the transfer of the flap were they just rotated the flap for 180 degrees and advanced it into the vagina instead of creating a subcutaneous tunnel (Fig.14) (54).

Walking is permitted 1 week after surgery, sitting is limited for 3 weeks after surgery to avoid pressure on the donor site of the flap. Discomfort or pain while sitting are possible complaints. Onishi et al., only studied one case, and there were no complications or period of follow-up mentioned (55). No complications such as necrosis of the flap, bleeding, infection or pain was noted by Kosugi et al (54).

In spite of the 100% healing in the study of Onishi et al., the study has many limitations. It’s difficult to make conclusions on results of only one case with not sufficient information of follow-up, possible disadvantages etc (55).
Hashimoto et al. reported an anatomic study and an adjustment of gluteal-fold flap volume. They report the advantages, described in the operation technique above, of the proximal and distal thickness of the flap. They suggest that because the flap can be adjusted to the required volume, the gluteal-fold flap has become useful for perineal reconstruction in the field of plastic and reconstructive surgery (56).

Kosugi et al. performed surgery in 5 patients who suffered from a RVF with the gluteal-fold flap and they obtained good outcomes with no recurrence (54).

Other authors who published case reports, such as Cardon et al. and Gürlek et al., report good outcomes with the reconstruction of a pudendal flap.

The ideal skin-flap reconstruction provides good functional preservation and good cosmetic outcome in both the reconstructed site and donor site. Gluteal-fold flaps are considered good flap donors because the perineum is an area of rich blood supply and arterial anastomoses. It is very easy to adjust the volume of the flap by resecting the fatty tissue. In this way, if the RVF is located higher, the shape and length can be adapted easily. Mostly the flap results in a good aesthetic outcome with no perineal deformities. Because of these advantages the flap can be considered a good option for perineal reconstruction (54-56). It is remarkable that possible disadvantages are not mentioned by any of the authors.

- Omentum flap

The **procedure**, described by van der Hagen et al., *enhances with the placement of four trocars*. The ileum loops are moved out the pelvic cavity and the sigmoid loop is placed in a left cranial position. Possible adhesions are dissected and a peritoneal incision is made over the sacral promontory, extending caudally along the rectum. The denonvilliers fascia is incised and after identifying the fistula, it is resected. The opening on the rectal side is closed in two layers and the vaginal side closed in a single continuous layer. After this, the omentum is dissected free from the transverse colon. The gastroepiploic artery, right or left, is dissected and the omentum is mobilized and brought down to the pelvis either by tunnelling it through the mesocolon or positioning it in the left or right paracolic gutters. Finally, the omentum is placed between the repaired rectum and vagina and sutured to the pelvic floor (57).
Schloericke et al. describes the operative technique more extensively. Point of difference with van der Hagen and colleagues are the types of fistula which were mostly a low or mid RVF, whereas the types of RVF studied by van der Hagen et al. were all high RVF. This required an additional transperineal approach for completion of the mobilization and reconstruction of the rectovaginal space (58).

In the study of van der Hagen et al., two patients could not be treated with an omentoplasty because the omentum was not suitable. Two major complications were the development of a necrotic omentum and an abscess. Minor complications included gastric retention, trocar wound infection and urinary infection. The median follow-up was 28 months (57).
In the study of Schloericke et al. major complications were an anastomotic insufficiency and a persistent fistula within the sphincter that needed re-operation. Minor complications such as gastrointestinal ileus and chest pain were observed in 22%. The median follow-up was 22 months (58).

The success rate reported by van der Hagen et al. is high (95%). In the series of 40 patients, in only 2 patients the omentoplasty was not successful. Several studies reported the use of laparoscopy to repair a RVF, but almost no series of patients with RVF treated by omentoplasty have been published in literature.

Laparoscopic repair has the advantages of minimal access surgery: minimal wound complications, less postoperative pain, and short hospital stay for the patient. Because of these reasons and the high success rate, van der Hagen et al. suggest that laparoscopic fistula excision combined with omentoplasty is a good option for high RVF repair (57).

Schloericke et al. studied the results of transperineal omentum flap repair in 9 patients. The repair was successful in all cases. Schloericke et al. also mentions the advantage of laparoscopic approach which avoids major complications and disturbance of physiological muscle activity by using a omentum flap instead of a muscle flap. They suggest that a muscle flap cannot be divided easily owing to the possible resulting inadequate vascular supply (58).
A major possible complication of the laparoscopic approach is a pelvic abscess and in case of failure, severe peritonitis can arise from a newly formed fistula. To avoid this, diversion with a stoma could be suggested. But both van der Hagen et al. and Schloericke et al. didn’t mention these disadvantages and there are no reports that compares this technique with another standard technique.
Several advantages of the laparoscopic approach is clear. Additional, Schloericke et al. mentions that the omentum flap is possible advantageous over a muscle flap. But with only respectively forty and nine patients, both the studies have not much power and therefore this technique should be investigated further.

- Puborectal sling interposition

![Fig. 15 Puborectal sling interposition (modified from (59)).](image)

As for almost every tissue interposition procedure, this technique begins with a perineal incision. After a while, Oom et al. performed this incision through the posterior fourchette of the vagina because they noted a high incidence of wound complications after the perineal approach. After the incision, the rectovaginal septum is dissected and the fistulous tract excised. The opening in the anterior wall of the anal canal is enlarged by removing all fibrotic tissue and this opening is closed as a first layer. The second layer consists of plication of the anterior wall of the anal canal and the third layer is an approximation of the both limbs of the puborectal muscle (59).

Major complication in the study of Oom et al. is postoperative wound infection in 42%. The second major drawback of this procedure was de novo dyspareunia (57%). The faecal continence was not adversely effected. The mean follow-up was 14 months (59).

Oom et al. studied 26 patients who underwent a puborectal sling interposition. The healing rate was 62% (16/26). But very notable are the poor results for patients with previous repair (31% versus 92% without previous repair) and the patients with a postoperative wound infection (36% versus 80% without wound infection) (59).
In the 1990s, three reports suggested that puborectal muscle interposition might be salutary for the treatment of RVF as the reported healing rates varied between 92%-100% (59).

Based on the poor outcome in patients who had undergone one or more previous repairs and the high incidence of dyspareunia after the procedure, the puborectal muscle interposition should not be considered as the treatment of choice for RVF(59).

3.4.3.5 Rectal sleeve advancement flap or pull-through

The principle of the endorectal pull-through consists of an endorectal mucosectomy (not only the mucosa but also submucosa and muscle layer), closure of the fistula and pulling the stump through the rectum. This can be performed via several approaches: partially via laparotomy or laparoscopy, via posterior sagittal approach (‘Kraske’) or combined with perineal approach to perform an anastomosis.

![Fig. 16 A Stricturectomy is performed in conjunction with rectal sleeve advancement (A+B) (modified from (60))](image)

The posterior ‘Kraske’ technique is described by Schouten et al. An incision is made, starting lateral to the left side of the sacrococcygeal joint ending just outside the external sphincter. They removed the distal part of the coccyx and deepened the incision to expose the pelvic floor after which they incised it to reach the distal partum of the rectum. The rectum was circumferentially mobilized both upwards and downwards as far as possible. Subsequently a circumferential mucosectomy (not only the mucosa but also submucosa and muscle layer) was performed and the entire fistulous tract was excised. Thereafter the defect in the rectovaginal septum was closed in two layers leaving the vaginal mucosa open for drainage. After advancement through the anal canal, the mobilized and transected distal part
of the rectum was anastomosed to the dentate line, which can be performed extraanally. Schouten et al. also describes the **abdominal approach** to reach and mobilize the rectum easily after which the rest of the transanal phase is performed as described in the Kraske procedure (61).

Tang et al. combined the laparoscopic approach to mobilize and mark the normal colon with the posterior sagittal approach to perform the rest of the pull-through as described by Schouten et al (62).

No deterioration of anal sphincter pressures and no de novo dyspareunia was observed. Beside a postoperative wound infection and loss of solid stool in one patient, no **complications** were mentioned by Schouten et al. and Tang et al (61, 62).

In the opinion of Schouten et al. there are two potential benefits for this procedure. First, the anal opening of the fistula is covered by a full-thickness layer of the rectum. Secondly, by not interposing tissue in the rectovaginal space, the risk of de novo dyspareunia is reduced. The healing rate, observed in 8 patients, is 75%. They mentioned that previous studies who also published about the use of rectal sleeve advancement procedure in RVF performed this technique by a transanal approach (61). Simmang et al. reports the use of the transanal approach to perform a stricturectomy in conjunction with rectal sleeve advancement (Fig. 16) in 2 patients with a RVF associated with anorectal stricture secondary to Crohn’s disease (60). While performing the abdominal procedure, because of pelvic adhesions the procedure may be risky because pelvic nerves and adjacent organs can be damaged. For that reason, Tang et al. set that the use of posterior sagittal approach makes the operation more easy and safer because of the good exposure of the surgical field and precise dissection under direct vision. Laparoscopic assistance can offer precise dissection, minimal abdominal injury and spare troublesome mobilization of the fistula. There was no recurrent fistula observed after repair of the RVF caused by Hirschsprung’s disease in a young girl. Tang et al. enumerate the indications of the combined laparascopic assisted endorectal pull-through and posterior sagittal approach: recurrent RVF accompanied with anorectal stenosis, diameter of the fistula more than 5 mm and heavy scar tissue or failure of repair (62).

Few studies are published about this procedure for the repair of a RVF. **As a conclusion**, in most cases, the indications are young girls with anorectal malformations or women with
anorectal stricture secondary to Crohn’s disease. Few complications or effects resulting from this procedure are mentioned by the authors. The advantage of the endorectal pull-through is the avoidance of difficult mobilization of the fistula. Contradictory with Tang et al., overall experience with this procedure learns that there is only a small entrance and little working space. Though further research is essential to properly evaluate the beneficial and side effects of the technique.

3.5 Predictors of outcome following fistula repair

3.5.1 Type of treatment

Pinto et al. published a study about the predictors of outcome following fistula repair. A possible predictor that was studied was the type of fistula repair. The procedures performed in their study included endorectal advancement flaps, gracilis muscle interpositions, seton placements, transperineal and transvaginal repairs. The overall success rate per procedure was 60% with no difference in recurrence rates based on the type of fistula repair. They also note that there was more recurrence in the procedures with postoperative complications (47.6%) such as mucosal bleeding, abscesses, wound dehiscence and others compared with 39.2% in the procedures without complications, but this wasn’t a significant difference (1).

3.5.2 Repeated fistula repair and interval between subsequent repair

Also studied by Pinto et al. was the difference in failure after treatment regarding to the time interval between a recurrent episode and a subsequent repair. The fistula recurrence after repeated surgical intervention within or after 3 months after initial surgery was exactly the same (44%). If the interval is longer, for example 6 months, the outcome was slightly different: 37.5% fistula recurrence within 6 months, 46% after 6 months, but this result is not statistically significant (1). Contradictory with this finding, Halverson et al. made the observation that patients who underwent reoperation within 3 months of their prior operation had a lower success rate compared with those whose repeat operation was performed at least 3 months after the prior repair (71% vs 45%, p= 0.58). They observed a correlation between successful outcome and increased time interval between repairs (63).

Management of recurrent RVF is even more challenging than the initial treatment. Decreased success with subsequent repairs may be attributed to unresolved inflammation, tension,
haematoma or underlying disease, eg Crohn’s disease. Pinto et al. also analysed the influence of the number of prior repairs on recurrence rates. They found that the greater the number of prior repair attempts, the higher the recurrence rate. But this could not be proven statistically (p=0.06). Lowry et al. reported a 88% success in initial RVF repairs. However, the success rate decreased to 85% after one failed attempt and to 55% after 2 prior attempts. Conversely, Halverson and colleagues failed to identify any correlation between the number of prior repairs and success rates (1). Ruffulo et al. studied the outcome of surgery for RVF due to Crohn’s disease in 52 patients. The overall cumulative closure rates, obtained with different surgical techniques, were 56, 75, 78 and 81 per cent. So, half of the patients whose initial operation has failed can expect eventual fistula healing if further surgery is attempted (29). In conclusion, after 2 fistula repair attempts, the success rate decreases. But as there is still a chance for successful healing, a new fistula repair attempt is worthwhile.

3.5.3 Etiology

- Obstetric fistulas

As these fistulas are mostly simple and non-complicated, the prognosis is better. Beside the small risk to develop a RVF after vaginal delivery, the risk to have a concomitant sphincter defect if a RVF is present is bigger. Tsang et al. emphasize the importance of preoperative sphincter evaluations before attempting for surgery. Patients with clinical or anatomical sphincter defects have inadequate local tissues to perform an endorectal advancement flap without sphincteroplasty. Due to the small number of patients in their study, they were unable to show statistical significance. Addition of levatoroplasty to the sphincteroplasty improves the success rates even more, this can be explained by interposing healthy tissue between the rectum and vagina (32). Rivadeneira et al. and other authors supports these findings (2, 3).

- Fistulas related to Crohn’s disease

Fistulas related to Crohn’s disease have a higher recurrence rate. This may be related to the treatment with systemic corticosteroids on the one hand, and the poor healing qualities of the diseased rectum on the other hand. Additional diarrhea makes it even more difficult to cope with the RVF for the patient. Due to the transmural nature of the disease, the fistula recurs more easily or may form anew (33). Pinto et al. found that the presence of Crohn’s disease is strongly associated with RVF recurrence (p<0.01). The success rate per procedure was 44.2%, significantly lower than other etiologies. Although, 78% of the Crohn’s patients eventually
healed after an average of 1.8 procedures (1). These findings were also observed by Halverson et al. who found that only 50% (6/12) of the Crohn’s patients healed compared with 90% (21/23) of the patients without Crohn’s disease (63). Contraindications to fistula repair in Crohn’s disease include stricture, indurated perianal tags of Crohn’s disease, macroscopic evidence of proctitis and undrained sepsis (16, 63).

Four alternatives in the treatment of a RVF in Crohn’s disease were proposed by Tuxen and Castro in 1979. They include 1. medical treatment with adequate drainage to control the infection, 2. proximal diversion of faecal stream with or without intestinal resection, 3 abdominoperineal resection of the rectum and closure of the vaginal fistula and the creation of a diverting stoma and 4. local closure of the fistula. But we should remark that there is a trend toward rectal preservation and the study of Tuxen et al. date from 1979, so this advice should be interpret with caution (33).

Ruffulo et al. remark that although the results of combined medical and surgical treatment in perianal Crohn’s disease are promising, RVF seems to be a negative prognostic factor in the therapy with anti-TNF-α. Sequential, surgery remains the main treatment for these fistulas. They also conducted research into the correlation between the duration of Crohn’s disease and previous extended colonic resection with failure of the first surgery attempt. Only the previous extended colonic resection turned out to be an independent risk factor (p<0.001) (29).

If fistula repair in patients with Crohn’s disease fail and severe proctitis is still present, proctectomy is the most definitive option (4).

3.5.4 Diverting stoma

Faecal diversion serves three purposes. First it diverts the faecal stream away from the healing perineum. Secondly it may have a beneficial effect on the local disease by temporally defunctioning the colon. Thirdly, if the repair fails and the resulting defect is even larger, no faeces but mucus will be discharged through the vagina. A diverting stoma is mostly used in the presence of infection or in complicated recurrent cases where surgical repair would require extensive dissection and eventually tissue interposition. If the rectum is not affected, the RVF should better be treated without faecal diversion (33).

In the study of Pinto et al., they consider whether the use of a diverting stoma influences the outcome of fistula repair in 43 patients. The fistula recurrence rate for patients with a diverting stoma was 48.8% when compared with 35.5% for patients without diverting stomas.
But 67.4% of these patients had prior recurrent fistulas, after a median of 3 previous procedures. There was no significant association between stoma creation before or at the time of the RVF repair with subsequent recurrence (p=0.18). This may be secondary to stoma creations being performed only in patients with complex fistulas (1). Other authors mostly support these findings (16, 29, 63). Halverson et al. even observed an increase in recurrence rate of 67% with diverting stoma versus 50% without stoma, which is statistically significant (p<0.001) (63).

Bauer et al. remark in their discussion that faecal diversion alone may lead to reduction of the symptoms but won’t result in the healing of the fistula (33). The general trend concerning a diverting stoma, is that this is only performed in complicated cases, but should not be advised for every fistula repair (63).

As a final note, faecal diversion is performed if all treatment fails to reduce the bothersome symptoms caused by RVF.

3.5.5 Others

Pinto et al. studied the influence of patient factors such as age, BMI, smoking history, comorbid condition of diabetes, use of steroid and immunosuppressive medications and number of prior vaginal deliveries. Only tobacco smoking seemed to be a significant factor (p=0.02) related to poor outcome of surgery. Chemicals present in cigarettes cause vasoconstriction as well as greater platelet adhesiveness, subsequently increasing the risk of microvascular occlusion and tissue ischemia. Nicotine use also promotes catecholamine release which stimulates vasoconstriction thereby decreasing tissue perfusion (1).

4. DISCUSSION

4.1 Etiology

The observation that obstetrical trauma is the most common cause of RVF and IBD the second most prevalent is generally accepted in literature. Other conditions that can result in RVF are radiation therapy, colorectal or gynecological surgery, infections, cancers,… However, the prevalence of each cause varies strongly in literature. This depends on the population characteristics on the one hand whereby obstetrical injuries for example are endemic in some areas in Africa and South-Asia or diseases like IBD has an extremely low incidence in China and other Asian countries (36). It’s very difficult to properly estimate the
correct prevalence of each etiology because the authors either don’t mention the percentages or they vary greatly.

4.2 Evaluation
Sometimes, extensive techniques are mentioned to diagnose the fistula but it should be noted that history and physical examination remain the principal means to identify the fistula.

4.3 Treatment
Several treatment options are described in literature. The non-surgical option mentioned by some authors is the infliximab therapy used for Crohn’s patients. The new, rising method of the autologous stem-cell transplantation should not be neglected and can be promising in the future for Crohn’s patients. But, more studies solely focused on the RVF, should be conducted. The surgical approach consists of local repair, muscle transposition or laparotomy. Local repair techniques consists of temporizing measures such as seton placement, fibrin glue instillation and bioprosthetic plugs. Seton placement should always be the first step in the repair of RVF since local sepsis has to be controlled before embarking to surgical repair. Fibrin glue instillation and bioprosthetic plugs are also minimal invasive but success rates reported in literature are disappointing. More advanced local repair techniques are fistulectomy and advancement flap repair. The advancement flap repair is the most implemented technique in the repair of RVF. The surgical approach for these local procedures may be transrectal, transvaginal or transperineal. The choice for either the transrectal or transvaginal approach depends on the individual education of the surgeons. The transrectal approach is the procedure most implemented by colorectal surgeons and conversely a gynecologist will typically favor the transvaginal approach with local muscle interposition. Results of the advancement flaps are better than the other less invasive procedures but still there’s a lot of discrepancy between different studies. In literature, the transvaginal approach tend to be preferable to the transrectal approach.

For recurrent RVF or fistulas with larger defects, muscle interposition with use of the gracilis, bulbocavernosus muscle and rarely the rectus abdominus is performed most commonly. Most of the colorectal surgeons have already experience with use of the gracilis muscle in the case of sphincter repair with Graciloplasty (Fig. 8). Therefore there will be a tendency to use this muscle in the rectovaginal fistula repair for more complex fistula or recurrent fistula. If muscle is not available, mesh using bioprosthetic grafts or flap
reconstructions for RVF repair has shown a worthy alternative but should be studied more extensively in the future.

When all treatment fails, a laparotomy is required for more complex fistulas that are not accessible or amenable to local repair. The remaining options then are a pull-through, proctectomy, permanent colostomy or even colpocleisis for high-risk elderly (1). The individual case complexity and surgeons preference may be the most important determinants for a successful repair.

**Limitations:**

General limitations are related to the heterogeneity of the overall study population, the different etiologies and a very broad spectrum of different techniques resulting from this heterogeneity. The low level of clinical evidence of the articles included and the rarity of the fistula type makes randomized controlled trials very difficult to perform. Selecting only the most qualitative articles is very hard due to the small number of patients with this type of fistula. This forces you to compile articles which are not high quality, for example very old articles, only small number of patients, case reports, no randomized clinical trials, little comparison between different surgical techniques, newer techniques not yet frequently studied,… Now there is not enough information to decide whether or not a technique should be rejected.

5. **CONCLUSION**

Due to the rarity of this type of fistula and the lack of adequate evidence, no appropriate treatment is available. As it is a devastating condition for the patient, an individual adapted treatment should be the main priority for the surgeon. This can be improved by proposing a flow-chart that makes the choice of treatment more adapted to each individual. Additionally, the quality of articles in literature should increase with the result of adequate conclusions about surgical techniques.
6. FLOW-CHARTS

Flow-chart 1
7. REFERENCES


