

Rectourethral Fistula Management

Daniel Ramírez-Martín¹ · José Jara-Rascón¹ · Teresa Renedo-Villar¹ ·
Carlos Hernández-Fernández¹ · Enrique Lledó-García¹

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Abstract Rectourethral fistula (RUF) is a rare condition that occurs, in most cases, as a consequence of prostate cancer treatments. Clinical suspicion and proper assessment prior to surgery are essential to adapt and successfully carry out an appropriate treatment plan. There are no randomized trials to guide clinical practice, and therefore, scientific evidence in this respect is limited. Expert recommendations seem to agree on the transperineal approach with flap interposition as the surgical treatment of choice in cases of complex fistulas, especially in those that have undergone prior radiation. Undoubtedly, the key to the successful treatment of the disease is the multidisciplinary and standardized management by physicians with experience in the field.

Keywords Rectourethral fistula · Prostate cancer · Radical prostatectomy complications · Cryotherapy · Brachytherapy · Prostate radiotherapy

Introduction

Rectourethral fistula (RUF) is a connection between the distal part of the rectum and the lower part of the

urinary tract. The pathology was first described by Jones [1] in 1858, although an earlier reference to a colovesical fistula is attributed to Rufus of Ephesus [2] in 200 A.D. RUFs are a devastating but rare complication that represent a surgical challenge due to their anatomical location and the involvement of two important functional structures: the urinary and anal sphincters.

According to etiology, most RUFs are acquired due to trauma and to bowel or urinary pathologies, but most frequently, they are a secondary result of a surgical or an ablative treatment complication. Therefore, most of the cases are iatrogenic [3]. More accurately, prostate cancer treatment is the main cause related to RUFs [4, 5], despite the fact that in the past RUFs have been described as a complication stemming from radical prostatectomies, regardless of the surgical approach was either open, laparoscopic, or robotic. Nowadays, RUFs are more likely to occur as a result of an ablative therapy used for the treatment of prostate cancer such as external radiation therapy (ERT), brachytherapy (BT), cryoablation (CA), or high-intensity focused ultrasound (HIFU). There are several studies published describing all these treatment options as the main cause of RUFs [6–10].

Clinical suspicion is essential to an appropriate management of this type of pathology; therefore, a detailed anamnesis and the administration of a battery of complementary tests are the key to reach an accurate diagnosis [11].

Many treatment options using different surgical approaches and techniques are available, which demonstrates the lack of randomized studies and the absence of standardized action protocols. For this reason, it is important to update and review the available articles regarding this rare but interesting and challenging pathology [12, 13].

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✉ Enrique Lledó-García
elledo.hugm@salud.madrid.org

¹ Andrology and Urethro-Genital Reconstructive Surgery Unit, Urology Dept. Hospital General Universitario Gregorio Marañón, Instituto de Investigación Sanitaria Gregorio Marañón, Universidad Complutense, Doctor Esquerdo, 46, 28007 Madrid, Spain

Incidence and Etiology

RUF is a devastating pathology with a fortunately low incidence and is related to several therapeutic procedures in the management of prostate cancer.

At present, the incidence of RUFs related to prostatectomy is around 0.53 %. Improvements in the technique have allowed for a reduction in the rate of RUF from 11 % in the decade of the 1990s [14] to the present rate. The last step in a prostatectomy procedure consists of a urethrovesical anastomosis. The resulting junction is an especially vulnerable anatomical area where other risk factors are present such as the intra-operative lesion of the rectum, which is described by Thomas [6] as the main factor related to the development of RUF with a correlation of up to 54 % of the cases. Other factors involved include age, previous radiation therapy, previous hormone therapy, or previous transurethral resection of the prostate [15]. There are differences according to surgical approach: the risk of RUF is 3.06 times higher in the perineal approach ($p=0.074$) [6] than in the abdominal one, but no differences are observed between the laparoscopic approach.

The incidence of RUF related to radiation therapy has increased in the last years due to the rise of use of the techniques involving this kind of energy. At present, any type of radiation therapy is related to around 50 % of RUFs [16]. The estimated incidence of RUF after ERT is about 1 % and about 3 % in the case of BT [17]. In cases where a combination of both procedures is used, the incidence rises regardless of the order of the procedures and the radioisotopes used.

Incidence after other ablative procedures such as CA [18] and HIFU [19] is estimated to be about 2 % at the moment. The development of advances in these techniques as well as in the security and control devices, such as the urethral warming catheter, has enhanced the results of these ablative therapies, thereby decreasing the incidence of RUFs [20].

Diagnosis

RUF diagnosis is basically clinical, and for that reason, an appropriate medical interview is imperative. There are some characteristic signs and symptoms for this disease: fecaluria is present in about 43–65 % of the cases, pneumaturia is present in about 67–85 %, and the leakage of urine through the rectum during micturition is present in 40 % of the cases [11]. Fecaluria is considered the most indicative symptom of RUF while pneumaturia, for instance, could be present in other kinds of infections or after a urinary tract catheter insertion.

Some other symptoms that could be found are recurrent urinary tract infections (73 %), abdominal pain (22 %), and dysuria (14.6 %).

In patients with RUF, reported symptoms not only enable an orientation to diagnosis but also an approach to treatment;

thus, cases of considerable fecaluria usually indicate a large defect in the tissue; therefore, it is especially important to consider a fecal diversion before the closure of the fistula tract [6].

Complementary tests not only confirm the clinical suspicion but are also a source of additional information regarding the exact location and size of the fistula and also provide further data regarding concomitant pathologies that must be taken into account (urethral strictures, involvement of the bladder neck, etc.). Cystoscopy and rectoscopy enable the assessment of the vitality and viability of the tissues. Among the different radiological imaging tests, retrograde urethrography, voiding cystourethrography, barium enema, and computed tomography are the most widely used [21, 22].

Treatment

RUF treatment is still a highly debated topic. There are no standardized protocols of action [23] on the subject. Most of the published articles are retrospective studies that share the experience of single institutions in the treatment of RUFs and very often using the same surgical technique. There are a wide range of surgical strategies described in the literature, ranging from transanal endoscopic microsurgery to robotic abdominal surgery, including transsphincteric techniques and transperineal approaches associated with the use of a broad variety of tissue interposition flaps [24–27]. There are few studies based on large studies comparing the results of the different approaches with the objective of analyzing possible factors associated with surgery success or failure, and neither of these studies is prospective [13, 28].

One of the most important points made by most of the authors in the reviewed articles is the importance of distinguishing and classifying RUFs into simple and complex. All of the following are characteristics of RUF complexity, mainly: size larger than 2 cm, the presence of urethral strictures, bladder neck sclerosis, or ischemic damage associated with ablative energies. On the other hand, a fistula is considered simple when it is secondary to surgical trauma on a previously healthy tissue.

Conservative Management [6, 29–32]

The conservative management of RUFs refers to the lack of surgical intervention in the anatomical area of the RUF, but it includes other surgical procedures aimed to accomplish a urinary diversion (suprapubic catheter, nephrostomy) or a fecal diversion (ileostomy or colostomy) [30]. The indications to attempt a conservative management are not well defined. In general terms, complete and spontaneous closure of the RUF is rare but possible within the first 12 weeks in those simple fistulas where the epithelization of the fistulous tract has not

yet occurred. Spontaneous closure of a complex fistula, especially in irradiated tissues, is exceptional; thus, conservative management should not be considered as an alternative in these patients (Mundy et al.) [31]. The decision to perform a temporary colostomy or ileostomy is not standardized, and some groups [6] base this decision on clinical facts. For those patients only suffering from pneumaturia or urine leakage through the anus, resolution through a low-residue diet is first attempted, while those patients suffering from fecaluria or sepsis are considered for colostomy straightaway. The appropriate amount of time needed to achieve a spontaneous resolution of a non-irradiated RUF is 3 months [32]. The literature shows that conservative management in selective cases has a wide range of success rates usually ranging from 14 up to 100 % [6, 29, 32].

Surgical Management [33–35]

The surgical management of RUFs is always challenging. It is a type of reconstructive surgery involving a very high degree of anatomical complexity that often involves fibrotic and dull tissues. The heterogeneity of surgical techniques is due to the low incidence of this pathology and consequently to the lack of action protocol standardization. There are up to 40 different surgical approaches described to treat RUFs including open abdominal techniques as well as laparoscopic and robot-assisted surgery, transperineal, transanal, or transsphincteric, which are the most widespread approaches according to the available literature. Resolution of the fistula can be achieved by primary closure or by graft interposition, generally with a graft from the buccal mucosa. Furthermore, in cases of complex fistulas, the use of flaps that enable an independent blood supply from a healthy tissue that also fills the perineal cavity is recommended. The different flaps that have been used are the gracilis muscle, omentum, abdominal rectus muscle, gluteus maximus muscle and dartos, with the most commonly used muscle being the gracilis. Finally, deciding when to proceed with surgery is also a highly debated topic. According to Grupta et al. [3], for simple fistulas, a 3-month waiting period before surgery is recommended to give time for the lesion to spontaneously close. In the case of complex RUFs, this interval should be increased up to 6 months to improve the quality of the tissues. The closure of a fecal diversion should be accomplished in 1 to 6 months (normally during the first 3 months) after surgery [26].

Different Approaches

As urologist, this is one of the most interesting aspects of this pathology. As listed above, despite the numerous types of surgeries, there are three main approaches that should be discussed:

- The transsphincteric approach, widely known as the York-Mason technique [33] (Fig. 1). York Mason was a British surgeon at the St. Helier University Hospital in London. In 1969, he retrieved the posterior transsphincteric approach described in 1917 for the excision [34] of rectal tumors and employed it in the treatment of RUFs. This technique has become popular since then and is today one of the most common techniques used to repair RUFs [24]. It is still the first choice of approach for simple and small RUFs [35]. However, it has intrinsic limitations; this approach hinders the urethral reparation with grafts and the muscular flaps interposition between the rectum and urethra [36]. The possibility of a transanal fecal fistula (between 9 and 26 %), wound dehiscence, and wound infection are complications that must be taken into account. The possibilities of fecal incontinence, despite the sphincteric section, and urinary incontinence are very low. Renschler et al. [37] describe one of the largest studies of patients treated using the York-Mason technique. They obtained satisfactory results in almost 92 % of treated patients; therefore, it is a safe option with an acceptable success rate in suitable patients.
- The transperineal approach [5, 22, 26, 38–40] is the most used technique in patients with complex fistulas because it enables a larger exposition of the urethral and rectal lesion and allows the use of both onlay mucosa grafts and different flap interpositions. The most commonly used graft flaps according to the literature is taken from the gracilis muscle, a muscle [27] located immediately under the skin of the inner thigh. Its main vascular supply depends on a neurovascular pedicle that reaches the muscle on its medial side. In 95 % of the patients, there are also 1 to 3 perforating arterial branches that reach the

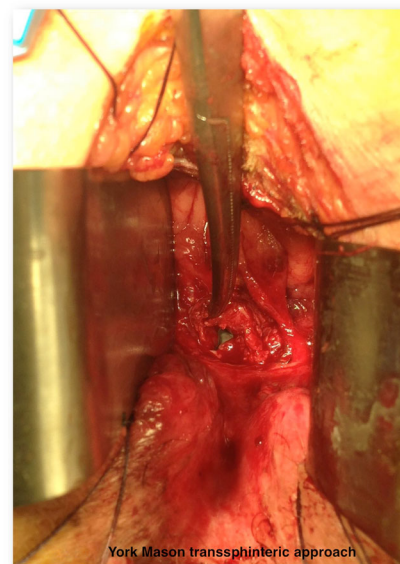


Fig. 1 York-Mason transsphincteric approach

muscle on its lateral side. In 90 % of the cases, ligation of these branches does not compromise the viability of the muscles but does produce a total necrosis of the muscle in 10 % of cases. In obese and small-sized patients, the gracilis muscle has a lower muscle/tendon proportion and could limit the use of this muscle as a flap due to the fact that the tendon is an avascular tissue [34].

There are multiple studies that describe the experience of different groups that have undergone surgery using the transperineal approach and the gracilis muscle flap interposition. Some of those studies are the one carried out by Ghoeiem [26] with 25 patients from a single institution and the one carried out by Vanni [5] with 74 patients which is, at present, the largest published study. Samplaski et al. [39] also support the use of this technique, highlighting the quality of life of the patients. Other studies showing the experience with other types of flaps are less common but can also be mentioned, such as the study carried out by Voelzke [22] using dartos flaps.

- The abdominal approach [41–43] is a technique that is much less commonly used. Laparoscopy-assisted RUF reparation is a direct application of the reconstructive techniques for vesicovaginal fistulas. It is a minimally invasive option that enables a perfect display of the anatomical site involved, guarantees the possibility of instrumental maneuvering, and allows the omentum and peritoneum tissues to be used as interposition flaps. Sotelo's group [41] shared their experience with three patients suffering from simple RUF treated with the laparoscopy-assisted abdominal approach with a 100 % success rate result using the omentum flaps. In robot-assisted approach [42] surgery, patients share the same benefits of the minimal invasiveness of laparoscopic surgery but with a highest economical cost. Omental flap interposition is a flap technique exclusive to abdominal surgery but could be unfeasible due to previous surgeries (Table 1).

Table 1 Rectourethral fistula management orientation

| Management | Possibilities | Indications |
|--------------|-------------------|--|
| Conservative | Antibiotic | Small size |
| | Low-residue diet | Secondary to surgery |
| | Urinary diversion | No ablative energies |
| | Fecal diversion | |
| Surgical | Transphincteric | Simple fistulas |
| | Transanal | Small size |
| | Transabdominal | |
| | - Omental flap | |
| | Transperineal | Complex, large, urethral pathology associated, ablative energies |
| | - Onlay grafts | |
| | - Muscular flaps | |

Latest Available Evidence

One of the most recent reviews regarding RUF management was published in 2013. It was an exhaustive study carried out by Hechenblaikner et al. [30]. Their study analyzed 26 published studies/articles until 2011 with a total of 416 patients. All included studies were retrospective with a wide range of cases (5–74). Without a doubt, this review is one of the most important papers in the meager amount of available literature regarding RUF, because it provides in-depth information with regard to preferences in the management of this rare complication. However, we must highlight two of its limitations: the first one is the etiological diversity of RUF included in the studies; post-radiation; up to 40 %, post-surgical, and post-traumatic. The second one is that all conclusions in that regard are considered level 4 of evidence and, therefore, must be taken as expert opinions.

Since the publication of this comprehensive review, there have been several single-institution studies, such as the study that included 37 patients carried out by the group of J. M Hanna et al. [28] that emphasizes the differences in management and results of RUF repairs in irradiated patients compared to non-irradiated ones.

There are several conclusions to take into account from these articles [28–30]: first, that conservative management is a limited option; only 10 % of the total cases were successfully treated without any surgery to the fistula site (it is important to point out once again that both fecal and urinary diversions, without any kind of intervention to the affected area, are considered as part of RUFs' conservative treatment). They discussed the need for fecal diversion, concluding that apart from selected cases of small surgically induced RUFs [31], fecal diversion should be considered either before surgery (3–6 months earlier in complex fistulas with a history of radiation) or in the same surgical procedure (simple cases).

Furthermore, they noted that the transperineal approach was the preferred technique that was employed in approximately two thirds of cases, especially in those with complicated fistulas or prior radiation and was generally associated with a gracilis muscle flap interposition. It is only if the studies of non-irradiated patients were analyzed that the transphincteric approach, specifically the York-Mason, is used similarly to the transperineal, without a clear preference.

Finally, some data is provided on treatment outcomes; surgical success rate is approximately between 87 and 90 % and is only slightly lower in the cases where the transanal approach was used. However, permanent fecal or urinary diversions, which are not considered as a surgical failure, are required in up to 8 to 10 % globally, but they reach values of 25 and 42 %, respectively, if we only consider irradiated cases.

In addition, the efforts of the Deborah S Keller group are also worthy of mention [13]. They have recently published a study of 30 cases. The main feature of this work is that the

patients were treated according to a preset algorithm treatment. This is important because although it is a retrospective study, there is some standardization in treatment protocol, in contrast to other recently published studies that focus on the results of a specific technique [43–45].

Their therapeutic algorithm is based on five factors at diagnosis: severity of symptoms, size of fistula, previous radiotherapy/cryotherapy, presence of urethral stricture, or sepsis. Depending on these factors, fecal and/or urinary diversion, surgery time and type of approach are decided. They concluded that algorithm-based treatment is useful for the standardization of treatment protocols for a rare clinical pathology such as RUFs.

Conclusions

RUF is a rare condition that occurs, in most cases, as a consequence of prostate cancer treatments. Clinical suspicion and proper assessment prior to surgery are essential to adapt and successfully carry out an appropriate treatment plan. There are no randomized trials to guide clinical practice, and, therefore, scientific evidence in this respect is limited. Expert recommendations seem to agree on the transperineal approach with flap interposition as the surgical treatment of choice in cases of complex fistulas, especially in those that have undergone prior radiation. Undoubtedly, the key to the successful treatment of the disease is the multidisciplinary and standardized management by physicians with experience in the field.

Compliance with Ethical Standards

Conflict of Interest Daniel Ramírez-Martín, José Jara-Rascón, Teresa Renedo-Villar, Carlos Hernández-Fernández, and Enrique Lledó-García each declare no potential conflicts of interest.

Human and Animal Rights and Informed Consent This article does not contain any studies with human or animal subjects performed by any of the authors.

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