



Rectourethral Fistulas: A Review

Amato A*

Department of Surgery, Hospital of Sanremo, Italy

Abstract

Rectourethral fistula is an uncommon but challenging disease that may have a congenital or acquired origin. The most frequent etiologies are traumatic and iatrogenic. In the last decades the widespread use of radiation therapy for prostate cancer led to an increase of complex cases. The diagnostic work up is usually not demanding. Spontaneous closure can be achieved after single or double diversion in small, non irradiated fistulas. Many patients will ultimately require surgery and several surgical procedures have been described. The available literature shows retrospective studies of small series on single techniques and does not allow an objective comparison because patients and disease characteristics are markedly different among reports. The optimal therapeutic choice for every stage of the disease still remains controversial and no single procedure has been proven to be the best treatment.

Introduction

Rectourethral fistulas (RUF) in adults are a rare but potentially devastating condition requiring complex and demanding surgery. Most of the studies that have been published on this subject are retrospective, report small series and some of them are no more than case-reports. Due to the rarity and complexity of this condition, epidemiologic data are lacking and no standardized algorithm exists to guide the surgeon in the management of these patients.

Classification and epidemiology

Culp & Calhoon et al. [1] described five groups of RUF according to their etiology: congenital due to malformation of the anus and the urinary tract; iatrogenic following surgery, radiotherapy or other treatment modalities; traumatic following fracture of the pelvis, gunshots or war wounds; neoplastic and inflammatory.

In a series of 23 male patients, Tiptaft et al. [2] found that the most common single causes of RUF were fracture of the pelvis and iatrogenic causes (prostatic surgery, radiotherapy and urethral instrumentation). Nyam et al. [3] reviewed a consecutive series of patients diagnosed in a 15 years period. Out of 15 male patients treated for a prostatic cancer, 7 have had a radical retropubic prostatectomy, 2 have had a radical retropubic prostatectomy after external beam radiation, 2 have had brachytherapy, and 3 were treated by a combination of external beam radiation and brachytherapy. One patient showed a fistula after dilation of a urethral stricture. A systematic review collected 416 patients from 26 papers [4]. A history of pelvic irradiation was found in 40% of the cases, a figure that grew in the study period from 4% before 1997 to 50% after 1998. Combination therapy was the most frequent radiation modality. In non-irradiated patients, etiology was iatrogenic in 65%, traumatic in 22% and inflammatory in 6% of the cases respectively.

RUFs were also reported after prostate cryosurgery, high-intensity focused ultrasound therapy (HIFU) and transrectal hyperthermia [5-8]. Barisic et al. [9] described 6 war wounds related RUF in Bosnia-Serbia war, successfully treated by York Mason technique. Anecdotally a RUF was found following rectal biopsy in irradiated patient or after sclerotherapy for piles [10,11]. RUF represents one of the most devastating complications of prostate radiotherapy, graded as "IV" according to the "Modified Radiation Therapy Oncology Group Lower Gastrointestinal Toxicity Scale" [12,13]. After prostate brachytherapy it occurred in 0.2-3 % of patients submitted to monotherapy, in 2.9 % treated by combined modality and in 8.8% who received salvage brachytherapy. Shah et al. [14] found that the onset of symptoms occurs at a mean interval of 27.3 months after treatment.

After radical retropubic prostatectomy, most RUFs are due to neglected rectal injury occurring during dissection of the posterior prostate. This anatomic area is particularly critical for the surgeon because previous biopsies or episodes of prostatitis or locally advanced tumors increase the risk of adhesion. The prostatic urethra being adjacent to the rectal wall is the commonest site for fistulation.

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*Correspondence:

Antonio Amato, Department of Surgery,
Hospital of Sanremo, Via Borea
56-18038 Sanremo, Italy, Tel: +39
0184 536017-536021; Fax: +39 0184
536215;
E-mail: ab.amato@libero.it

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Less commonly, the bulbous and membranous urethra may be involved. If the wound can be directly visualized intraoperatively, a direct suture of the rectal wound is performed, avoiding systematic colostomy. Failure to notice the wound intraoperatively can lead to fistula formation. A review on 3834 radical prostatectomies found a mean incidence of rectal injuries of 0.7% (range 0.2–2.9%), regardless of open or laparoscopic surgical approach [15]. Smith & Veenema et al. [16] reported their 20-year experience with 160 patients who had undergone radical retropubic prostatectomy with an incidence of 9.4% rectal injuries. In this group only 4 fistulas developed. Noldus et al. [17] reported 23 rectal injuries (3.9%) over 589 radical prostatectomies and cystoprostatectomy, with 12 RUF.

Case Presentation

Diagnosis

The diagnosis may be strongly suspected from the patient's history. The patients complain of the passage of gas or fecal particles through the urethra or abnormal urethral discharge, passage of urine through the rectum during micturition, recurrent urinary tract infections. At the rectal examination a hard area in the anterior wall of the low-middle rectum could be detected. The most important diagnostic step is voiding cystourethrography that can demonstrate the passage of the radiopaque contrast agent into the rectum through the fistula tract. Proctoscopy and urethroscopy could be useful to localize the enteric and urinary openings of the fistula. The distance of the rectal opening from the anal verge is a significant parameter for the subsequent therapeutic strategy.

Treatment

Especially in small fistulas, conservative treatment with urinary catheter diversion and fully absorbable diet, alone or combined with temporary colostomy, can achieve healing. Therefore, an initial attempt with conservative treatment is reasonable. Diversion of urine as well as correction of any stricture of the urethra distal to the fistula and antibiotic therapy is the first-line therapy. In 1997, Noldus et al. [17] reported that 7 out of 12 RUF after radical prostatectomy healed spontaneously by long-term transurethral catheter.

While urinary diversion is mandatory, fecal diversion by means of a temporary colostomy is controversial: it is mandatory as part of a double diversion for some authors or selectively indicated by others. It can prevent rectal wall distension and intraluminal pressure, minimizing infection. Currently, colostomy could be recommended when antibiotics alone cannot control fistula-associated sepsis and severity of symptoms, when the fistula lies on irradiated tissues, in recurrent fistulas, in large RUF (diameter >2 cm) and in immunosuppressed patients. A bridge-to-surgery diverting colostomy with a 3–4 months interval before surgical treatment is advocated in order to reduce the local inflammation. Hechenbleikner et al. [4] reported that a double diversion was performed before surgery for fistula in 89% of patients. In 1997, Al-Ali et al. [18] reported 47% of spontaneous healing after double diversion alone in a series of patients affected by war wounds, while healing after diversion was noted by Chun in 60% of patients affected by RUF following laparoscopic radical prostatectomy [19]. On the other side, Shah et al. [14] found that diversion of fecal stream does not heal RUF after brachytherapy for prostate cancer.

Many patients will ultimately require surgery and it is often technically demanding. Several approaches have been described and more than 40 surgical techniques have been proposed with

variable rate of success [20–22]. However, no comparative study exists; the published series are usually retrospective and show a wide heterogeneity in terms of etiology, location, morphology of the track and quality of the surrounding tissues. So, till now the optimal operative management still remains controversial and there is no consensus regarding the ideal treatment.

In 1958, Goodwin et al. [23] reported a series of 22 rectourethral fistulas treated through a wide perineal approach. The procedure requires an extensive mobilization of the rectum posteriorly and the bladder anteriorly allowing the interposition of the levator ani muscles between the urinary tract and rectum.

In 1885, Kraske described a posterior approach to the rectum that required coccygectomy and partial sacrectomy, in order to perform a rectal resection avoiding the challenge of a laparotomy. Some decades later, Kilpatrick & Thompson proposed the same approach for the treatment of RUF. After a fully circumferential mobilization of the rectum proximally and distally to the track, the fistula was divided sparing as much as possible on the urethral side. The rectal opening was excised and closed, the urethra was repaired and stented with a catheter.

The York-Mason procedure [24] requires the division of the rectal sphincters to give direct access to the RUF. It allows direct visualization of the fistula via para-coccygeal trans-sphincteric incision (from the tip of the coccyx to the anal canal). The patient is placed in prone jackknife position with abduction of both lower limbs and buttocks separated with adhesive tape. After incision and sphincter division, the mucocutaneous junction and both internal and external anal sphincter are marked by color-coded sutures to provide a proper alignment and reconstruction at closure. The fistula is excised exposing the catheter in the prostatic urethra and the rectal wall is separated from the urinary tract by sharp dissection to allow a sufficient mobilization. After closing the urethra, the rectum is sutured paying attention that the suture lines do not overlap each other with a "vest over pants" technique. Some authors suggest that suture should be, if possible, covered with soft, healthy tissue, e.g. omentum, gracilis muscle, scrotal flap, while others believe that it is not an essential part of the procedure. The York-Mason procedure appears to be effective and provides a superb exposure through unscarred tissue planes. Published papers show it is a safe and reproducible technique with a success rate of 91.7–100%, minimal morbidity and no impairment of fecal continence [22,25–30]. Thus, for a long time it was considered as first-line treatment of RUF after radical prostatectomy and has been the most widely popular technique in the last decades.

Subsequently Parks et al. [31] described a fistula's repair by means of a full thickness flap of the anterior rectal wall through a transanal approach, with the aim to avoid any division of the sphincter mechanism. The rectal mucosa is excised laterally and distally to the rectal opening of the track, denuding the circular muscular layer of the rectum. Then a flap of about four centimeters in length is performed. Dreznik et al. [32] described a similar procedure excising the fistulous track to leave a transverse defect of 1–2 cm in the anterior rectal wall. A longitudinal incision was made in the rectum from each lateral edge of the defect for 3–4 cm proximally, performing a U-shaped rectal flap. The width of the flap should be at least twice than length. The defect in the urethra was closed using interrupted absorbable sutures over the urethral catheter to prevent stenosis. The rectal flap was advanced over the fistula and sutured to the rectal wall with interrupted absorbable sutures ensuring the absence

of tension. Transanal flap repair showed a healing rate from 67 to 100% and the procedure is repeatable with the same effectiveness [32-34]. We proposed a sphincter-saving procedure through an anterior intersphincteric approach. The anterior rectal wall is sharply dissected from the urethra until at least 2 cm above the fistula level. The track is isolated and excised and the surrounding tissues are mobilized to allow a tension-free repair. The urethral defect is closed using a single row of 3-0 polyglactin interrupted sutures. Transanally a U-shaped full-thickness rectal flap is advanced over the rectal opening and sutured with a 3-0 monofilament interrupted sutures, being careful that the two suture lines lie in different planes. Five patients were successfully treated with no fistula recurrence or impaired continence after a median follow up of 2 years [35].

Direct repair is best suited for surgically acquired fistulas that are relatively small and surrounded by healthy tissue. In the last decades the number of patients submitted to multimodalities treatment for prostate cancer has risen and consensually the risk to develop a RUF has increased [12,13]. As a consequence, surgeons more frequently have to deal with more complex situation than in the past. Radiation fistulas are often large and necrotic, with poor tissue quality and impaired wound healing and they cannot be safely repaired with simple division and closure. Tissue interposition is almost always required and it can be accomplished using a dartos or gracilis flap via a perineal incision. Furthermore, tissue interposition can be combined with other technique to provide a more versatile surgical armamentarium. Youssef et al. [36] successfully treated 12 male patients by means of a perineal subcutaneous dartos pedicled flap procedure. RUFs were traumatic in 8 cases, or were developed after prostatectomy in 4 cases. Through an inverted U-shaped perineoscrotal incision a dartos flap was interposed as a vascularized tissue flap between the repaired rectum and urethra. In the follow up period ranging from 9 to 42 months no recurrence was registered.

The gracilis muscle transfer (unilateral or bilateral flap) can be used universally regardless of patient age, gender or body type, for closure of vesicovaginal, urethrovaginal and prostatocutaneous fistulas as well as RUFs. It can reliably be harvested, and is rarely lost if the dominant proximal vascular pedicle is carefully preserved and an adequate subcutaneous tunnel is created to avoid flap compression. A small comparative study including five other techniques demonstrated the clear-cut advantage of transperineal gracilis muscle interposition flap in a heterogeneous group of patients [3]. Besides the well-known technical aspects related to the proper dissection and repositioning of the muscle, the peculiar key-points of the procedure include: a perineal incision that allows a complete dissection of the urethra from the rectum proximally and distally to the track as well as a urethroplasty if a concomitant urethral stricture is present; the fixation of the tip of the gracilis muscle to the apex of the prostate and anterior rectal wall above the level of the closed fistula, so that the bulk of the muscle rests between the sutured rectal and urethral defects. In a retrospective study, Ghoniem et al. [37] reported a 100% healing rate at a mean follow up of 28 months in a series of 25 patients with complex RUFs, 15 of which underwent radiotherapy. Vanni et al. [38] performed 74 transperineal repair using gracilis muscle flap in 68 cases and other interposition flaps in 6 cases, with a 100% healing rate in 35 non irradiated patients vs. 84% in 39 irradiated patients. The greater omentum is an option but this procedure involves laparotomy and deep anterior pelvic dissection and it may not be feasible in patients who have undergone previous abdominal surgery.

High-tech mini-invasive procedures were also proposed in the last years. Felipetto et al. [39] closed a prostatic-cutaneous fistula which complicated a Pseudomonas prostatitis by means of human fibrin sealant (Tissucol[®]). Etienney et al. [40] successfully treated a recto-urethrocutaneous track in Crohn's disease. The biological glue passively occludes the fistula, promoting the process of endogenous fibrin deposit and cicatrization by stimulating fibroblast proliferation and migration, neovascularization and reepithelialization. Autologous fibrin glue was used by Venkatesh et al. [41] in the treatment of a group of patients including complex anorectal fistulas, rectovaginal fistulas and urethro-vesico-rectal fistulas. A success rate of 60% was reported without major complications but patients with urinary tract fistulas and acquired immunodeficiency syndrome failed to respond. Synthetic biological glue (Glubran[®]) was successfully injected to close a prostatic-perineal fistula complicating an abdomino-perineal resection of the rectum and a neobladder-ileal fistula [42]. These small experiences suggest that the therapeutic use of biological glue is a minimally invasive technique, without any significant complication and side effect. The procedure is easily repeatable and does not affect further surgery suggesting a role as first-line therapy before venturing on complex and high-risk surgical procedures. A combined approach was suggested by Verriello et al. [43] who obliterated a RUF following radical prostatectomy injecting a fibrin sealant before performing a rectal mucosal flap to close the rectal opening.

In 2007, Rivera et al. [44] developed a RUF staging system combining the fistula location and caliber with the presence or absence of an irradiated field. They aimed to achieve a rational-though not evidence-based - repair algorithm allowing the selection of the most appropriate technique for each patient. Stages I and II include non-irradiated patients with low or high RUF respectively. Transanal and transperineal (trans-sphincteric) procedures with rectal flap seem to be appropriate in these conditions. The latter techniques are suggested also in stage III irradiated patients with small (diameter < 2 cm) RUF. Large (stage IV) and complex (stage V) RUF in an irradiated field should be managed with a muscle interposition flap procedure. In stages from III to V a diverting colostomy is performed 4-6 weeks before definitive surgery.

Conclusion

RUF is an uncommon but challenging disease, and several surgical procedure with different approaches have been described. Surgery is technically demanding with a cumulative healing rate of 87.5% and with an overall permanent fecal diversion rate of 10.6% (25% in irradiated vs. 3.8% in non-irradiated cases) [4]. Retrospective studies, often from single institutions, highlight the single repair techniques, but until now no comparative prospective trial has been carried out due to the rarity and heterogeneity of this condition. Pertinent literature shows poor quality data that makes difficult objective comparison, and no consensus on the best treatment option can be defined. Most of the therapeutic choice lies on personal surgeon preference and technical expertise.

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