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# SPONTANEOUS PROSTATO-CUTANEOUS FISTULA: AN INTERESTING AND RARE CASE

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#### ABSTRACT

Urinary fistulae from the prostate are very rare; they may occur as a result of surgery, trauma, malignancy or chronic inflammation. The prostato-cutaneous fistula is a rare entity and a very few cases have been reported. We report an interesting case of a spontaneous prostato-cutaneous fistula with emphasis on the management.

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#### INTRODUCTION

Urinary fistulae from the prostate are very rare; they may occur as a result of surgery, trauma, malignancy or chronic inflammation. The most common sites for fistulae include the vagina, rectum, bowel, abdominal wall, scrotum or perineum. The prostato-cutaneous fistula (PCF) is a rare entity and a very few cases have been reported. The spontaneous PCF is even more rare and has not been reported in the past to the best of our knowledge.

## **CASE DISCUSSION**

A 44-year-old male presented with the history of dysuria, increased frequency of micturition (Day: 8-10, Night: 3-4) and intermittent, low grade fever for the last 2 months. After one month of the initiation of the above mentioned symptoms, he noticed a swelling in the perineum. The swelling was associated with mild pain, for which he took analgesics without any proper consultation. After 1 week, the swelling burst spontaneously leading to multiple openings in the perineum. These openings were associated with discharge of pus and urine during micturition. There was no history of tuberculosis, sexually transmitted disease, trauma, GI tract symptoms or previous endourological procedures. He was a known case of de-compensated liver disease on treatment. He was also having diabetes mellitus, which was poorly controlled. He was on steroid inhalers for the bronchial asthma and oral low dose steroid for the Idiopathic thrombocytopenic purpura.

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The general physical examination and the vital statistics were in normal range. On local examination, there were multiple fistulous openings in the perineum with urine and pus discharge. On per-rectal examination, there was grade II benign prostate with no tenderness. The external genitalia were normal. The serum prostatic specific antigen (PSA) and routine blood investigations were in normal range. Urine analysis revealed pyuria (WBC: 20-50/ High power field). The urine culture was positive for Escherichia coli sensitive to multiple antibiotics. Montoux test, Chest x-ray and Urine for acid fast bacilli were negative for tuberculosis. The ultrasound showed normal upper urinary tract and mildly enlarged prostate (28 gm) with 20 ml of residual urine. The retrograde urethrogram (RGU) / micturating cystourethrogram (MCU) showed a fistulous tract between prostatic urethra and perineal skin with collection of contrast medium in the prostate (figure 1). A diagnosis of PCF was made and suprapubic catheterization (SPC) was done for the diversion of urine. The intravenous antibiotics (Ceftriaxone and Amikacin) were started according to the culture/sensitivity report and continued for 2 weeks. After that, oral levofloxacin (500 mg, once a day) was given for another 2 weeks. Strict glycemic control was achieved with the help of an endocrinologist. The adequate nutritional support was provided to the patient with optimum energy and protein supplementation. Multiple biopsies taken from the margins of the fistulous openings were suggestive of chronic non-specific inflammation. After 1 month of above mentioned conservative measures, urine discharge gradually stopped, but minimal pus discharge persisted. We planned for endoscopic evaluation, which revealed normal urethra with the elevated bladder neck and the prostate with small lateral lobes. There was no endoscopic evidence of any fistulous opening or abscess in the prostate.



Figure 1 RGU/ MCU showing prostato-cutaneous fistula

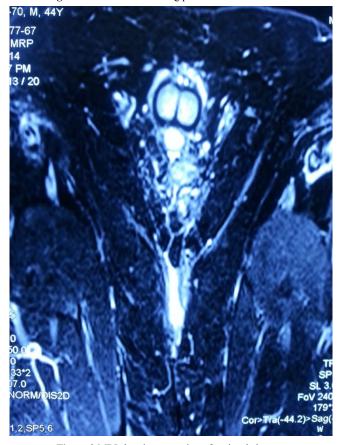


Figure 2 MRI showing extension of perineal abscess

In view of RGU finding, the prostate was resected superficially to de-roof any residual prostatic abscess. As discharge persisted even after 1 month of the trans-urethral resection (TUR), we planned for magnetic resonance imaging (MRI) to evaluate for any residual disease. The MRI showed perineal abscess with inflammatory changes in the para-urethral region as well as an extension towards the anterior aspect of anorectal junction with multiple sinuses in the perineum (figure 2). We planned for the excision of all sinus tracts in view of the MRI findings. The phlegmon including the sinus tracts was completely excised and the wound was closed primarily with a drain. RGU was done intra-operatively which revealed normal urethra with a wide prostatic fossa without any leak. The drain was removed on the third day and the wound healed well. At the end of 6 months, the patient is doing well without any urinary symptoms or any sexual dysfunction.

## **DISCUSSION**

The PCF is a rare diagnosis with only few cases reported.<sup>1, 3</sup> Two cases of the PCF were reported after the transurethral resection of the prostate in men who had previously undergone external radiotherapy for localized prostate cancer.<sup>3</sup> Palit *et al.* presented a case of PCF following injection of oily phenol for the internal hemorrhoids.<sup>4</sup> Garg *et al.* reported a rare case of the prostato-ano-cutaneous fistula after a prostatic abscess.<sup>5</sup> Kumar *et al.* reported a case of PCF in patient who underwent the anastomotic urethroplasty for a pelvic fracture and urethral distraction defect.<sup>6</sup>

The PCF may occur after a neglected prostatic abscess. The prostatic abscess is now uncommon due to the widespread use of broad spectrum antibiotics. Prostatic abscess is usually the sequelae of acute bacterial prostatitis due to Escherichia coli and other gram negative rods, although the tubercular and fungal prostatic abscesses are also known to occur.7, 8 The predisposing factors for development of prostatic abscess diabetes mellitus, include indwelling catheters, immunosuppressive therapy, instrumentation and chronic renal failure requiring maintenance haemodialysis. 7, 8 The exact aetiology in our case is unclear but it may have resulted from a missed peripherally located abscess which may have ruptured in the prostatic urethra inside and the perineum outside. Fever and dysuria as initial presentation and after that appearance of a painful swelling indicate toward a missed prostatic abscess. In addition, the presence of immunosuppressive therapy, uncontrolled diabetes and other co-morbidities were various predisposing factors for the prostatic abscess in our patient.

The PCF may present with a single or multiple openings with persistent or intermittent discharge through the scrotal wall or the perineum or the medial aspect of thigh.<sup>3-6</sup> There may be an associated "bubbling" at the openings on forceful micturition. The PCF may also present with leg or groin pain, dysuria, pus discharge per urethra and low backache.<sup>3-6</sup> In our case, the patient presented with dysuria, increased frequency of micturition (Day: 8-10, Night: 3-4) and intermittent, low grade fever. After that, a perineal swelling appeared which burst spontaneously leading to multiple openings.

Routine blood investigations, urine routine examination, urine microscopic examination and urine culture should be performed. The creatinine levels in urine are higher than the serum levels. Therefore, in the condition of a suspected urinary fistula, testing the creatinine level in the extravasated fluid and comparing this value with the serum creatinine levels will confirm urinary leakage. The pelvic imaging is helpful for reaching at a diagnosis and for making an optimal treatment plan. The RGU/ MCU and fistulogram are helpful for the

delineation of a fistulous tract. 8-10 Ultrasound is a non-invasive and an easily available study for the initial evaluation of kidneys, bladder and prostate. The transrectal ultrasound may show any prostatic and peri-prostatic collection with an associated fistulous tract.<sup>3-6</sup> MRI is better in comparison to the computated tomography for demonstration of a fistulous tract and any associated prostatic abscess.3-6 The findings on cystourethroscopy are often nonspecific and include localized erythema, papillary, or bullous changes. Immature fistulae may appear as an area of localized bullous edema without any distinct ostia. Mature fistulae may have smooth margins with a variably sized ostia.<sup>8-10</sup> The patient with a prior history of pelvic malignancy or in the condition of any abnormal appearing tissue, the biopsy of the fistula should be performed to evaluate any malignant fistula. 9, 10 There is no general consensus regarding the investigations to be performed due to rarity of cases. Imaging options can be chosen according to the case scenario and available options. In our case, RGU/MCU was helpful in making the diagnosis and cystourethroscopy was inconclusive. MRI was helpful in our case for making a definitive treatment plan.

The PCF is a serious complication and can be very difficult to treat.9 Immediate management of the urinary leakage is vital. This includes temporary urinary diversion usually in the form of suprapubic cystostomy or rarely bilateral nephrostomy or Ileal conduit urinary diversion.<sup>3-6, 9</sup> Addressing this quickly will reduce psychological distress, skin breakdown and related complications. 8-10 The appropriate use of pads, catheters and appliances can be very helpful in this regard. Adequate care of skin and control of odour are also adjunctive measures in minimizing patient-related distress until definitive therapy can be undertaken. 8-10 The nutritional status, infection, malignancy, urinary obstruction, and the presence of a foreign body are important considerations during the evaluation of a urinary fistula.<sup>8-10</sup> Ensuring adequate nutrition, eliminating infection and achieving an unobstructed urinary drainage is integral to the healing of a urinary fistula.<sup>8-10</sup> Appropriate antibiotic therapy should be started according to the available guidelines and preferably according to the culture/sensitivity report. Tuberculosis, fungal infection, uncommon bacteria and resistant species should be ruled out in case of no response or worsening of the symptoms with commonly prescribed antibiotics. As tuberculosis is prevalent in India, we ruled out tuberculosis as a possible cause by appropriate tests. Obstruction distal to the diseased site and the malignancy should be also ruled out.<sup>10</sup> In general, conservative measures are useful for the small fistulae (usually less than 2 to 3 mm in diameter). 8-10 Suprapubic cystostomy, eliminating infection by appropriate antibiotics, strict glycemic control and optimum nutrition provided in our case led to significant improvement of the symptoms. There was a significant reduction in the size of phlegmon with the resolution of the prostatic portion of the abscess. This helped us to completely excise all of the infected tissue without injuring urethra.

Minimally invasive treatment involving disruption of the epithelial layer of the fistula tract has been mentioned in the small epithelialized fistulae. Prolonged bladder catheterization combined with the endoscopic fulguration of the fistula tract may be successful in small, immature fistulae. The fibrin sealant may also be injected directly into the fistula tract after fulguration. Exploration with adequate excision of the fistulous tract and debridement of devitalized and ischemic tissue is necessary if the conservative or endoscopic management failed.<sup>3-6, 9</sup> Any foreign bodies or synthetic materials present in the region of fistula should be removed.8-10 A careful dissection with maintenance of the hemostasis is necessary for good results. <sup>10</sup> The suture line should be tension-free and nonoverlapping. <sup>10</sup> An adequate urinary tract drainage and/or stenting after repair is must. <sup>10</sup> Our patient responded very well by exploration and excision of all diseased tissue. We continued the adequate urinary drainage in form of SPC for 2 more weeks after excision of phlegmon. There was a complete wound healing and the absence of any urinary symptoms. Our patient regained his normal sexual life after 1 month of surgery without the need of any intervention.

To the best of our knowledge, this is the first report of a spontaneous PCF extending into the perineum. There was a partial response to the conservative management and complete response to the definitive management.

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