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RESEARCH ARTICLE

SPONTANEOUS NEPHROCUTANEOUS FISTULA-A RARE CASE REPORT AND REVIEW OF LITERATURE

***Prof. (Dr) Rajesh Kumar Abbey and Dr. Anantbir Lubana**

Department of Surgery, Giansagar Medical College, Banur, Patiala, Punjab, India

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ABSTRACT

Spontaneous Nephrocutaneous Fistula (SNCF) is a very rare entity. The known causes are: post-operative, calculous pyonephrosis, chronic pyelonephritis, renal infections, perinephric abscess, trauma, xanthogranulomatous pyelonephritis, renal tumours, open surgical procedures, reflux diseases and renal tuberculosis. Till 2013 only 29 cases were published. It is usually associated with obstructing calculus and non functioning kidney. A case of nephrocutaneous fistula in a 67 years old female patient complaining of seropurulent discharge from a fistula over the right lumbar flank for the last eight month, is presented and discussed here, for its rarity and management. CT fistulogram was performed which revealed a fistulous tract from the skin to the pelvicalyceal system of the right kidney, calcification of the renal parenchyma and obstructing renal stones (obstructive uropathy). Patient was investigated for renal tuberculosis, was found positive and was put on anti tubercular treatment to which she responded ,as the fistula healed and closed spontaneously. Open nephrectomy along with excision of the tract was performed. Follow up period remained uneventful. Open surgery, after 3-4 weeks of antitubercular treatment (ATT), is recommended in such cases.

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INTRODUCTION

Spontaneous Nephrocutaneos Fistulae (SNCF) may be internal or external. Internal spontaneous nephrocutaneos fistulae communicating with the adjacent organs (colon, duodenum, jejunum) are not very uncommon (Ansari., 2004), but external spontaneous nephrocutaneos fistulae are quiet rare and only few cases have been reported in the literature (Singer., 2002). The main causes of this condition are post operative, trauma, chronic pyelonephritis and renal tuberculosis (Sharma., 2013), chronic urinary tract infections, stones (Singer., 2002) and xanthogranulomatous pyelonephritis (Rubilotta *et al*., 2014). The occurrence of SNCF due to renal tuberculosis is rare (Singer., 2002) but not uncommon in the Indian scenario where tuberculosis is quite prevalent (Tanwar., 2016, Ansari., 2004). India, China, Indonesia, South Africa and Nigeria are the top five among the so called high-burden countries (HBC) for tuberculosis (WHO., 2008). Until 2013 only 29 cases of SNCF were reported in the literature (Rubilotta *et al*., 2014). The patient usually presents with fistula on the abdominal flank, discharging urine, pus or serous fluid. Physical examination and CT fistulogram are confirmatory of this condition (Rajeev., 2016) and these investigations were done in this patient, which confirmed the diagnosis and were helpful for prognosis as well. The affected kidney of these patients is usually poorly functioning or non functioning (Rubilotta *et al*., 2014, Sharma *et al*., 2013). Open nephrectomy is advocated along with excision of the fistulous tract (Sharma *et al*., 2013, Tanwar., 2016). We recommend that Spontaneous nephrocutaneous fistula as a result of renal tuberculosis should be operated by open surgery preferably after a short course, 3-4 weeks of anti tubercular therapy (ATT), to contain the infection and minimize the morbidity.

Corresponding author: Prof. (Dr) Rajesh Kumar Abbey,
Department of Surgery, SRMS Institute of medical sciences, Bareilly, UP, India.

Case report

A 67 yrs old female patient presented in surgical OPD with painless serous discharge from a chronic sinus on right abdominal flank (Fig.1) for the last eight months. The discharge was seropurulent in nature. She had no fever but had past history of recurrent episodes of UTI. There was no history of previous surgery, trauma, diabetes mellitus or immune compromised status. On clinical examination a ballotable, firm lump of the size of about 6x6 cms was detected on the right lumbar area. The white cell count was 4200/cumm, Serum urea and serum creatinine was 27mg% and .4mg%, respectively.

Fig.1. Discharging sinus [NCF] on right abdominal flank

Fig 2. X Ray KUB showing calculus in pelvis and calcification of right kidney

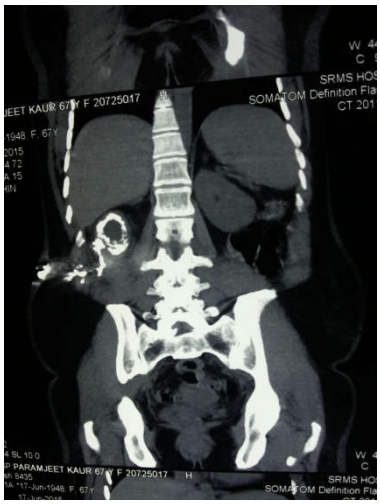

Fig 3. CT fistulogram

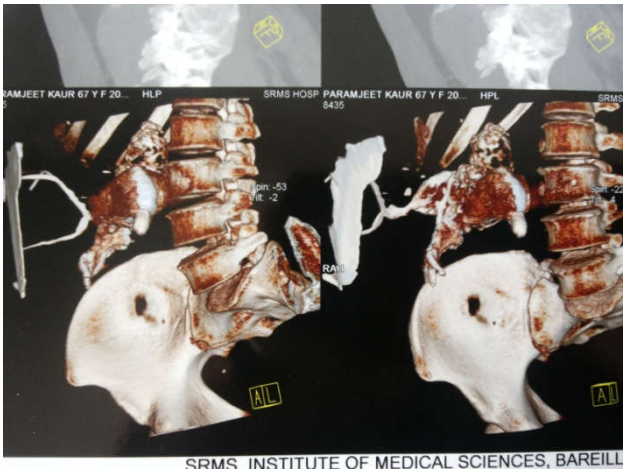

Fig 4. CT fistulogram

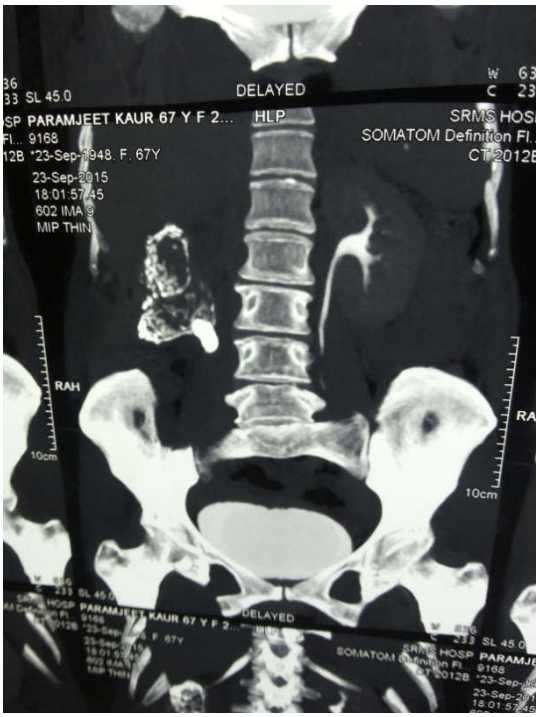

Fig 5. Contrast study showing non-functioning right kidney and normally functioning left kidney

Fig 6. Healed nephrocutaneous fistula after ATT

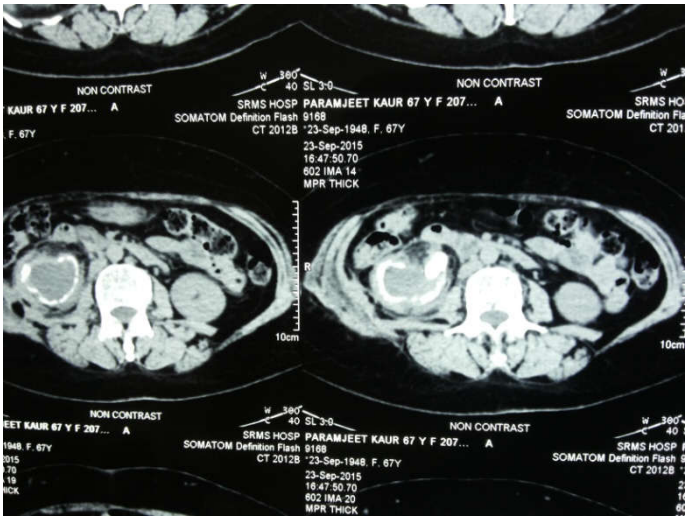

Fig 7. Follow up CT showing obliteration of fistulous tract

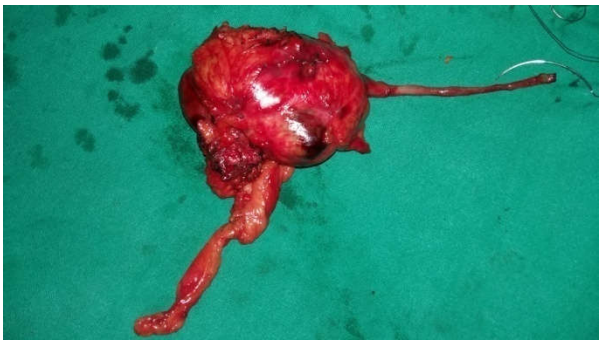

Fig 8. Operated specimen (total nephrectomy with excision of the fistulous tract

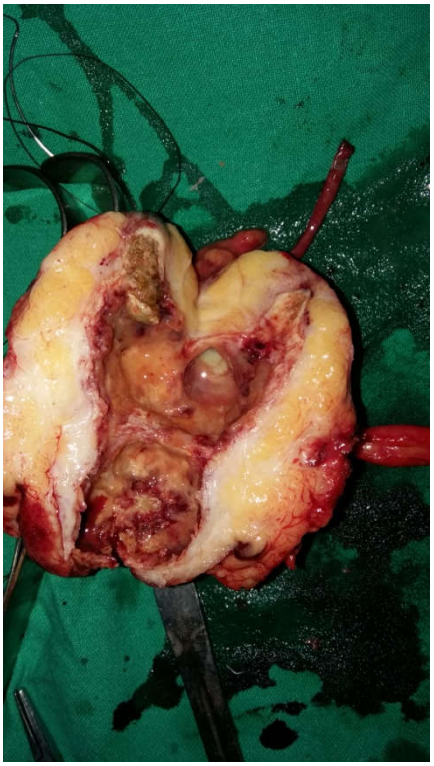

Fig 9. Bisected specimen of the kidney along with ureter and fistula

The culture of the discharge was sterile. PCR test of seropurulent discharge for tuberculosis was positive (24 MTB cells/ml). Urine for AFB was negative. ESR was 36 mm/1st hr and reduced subsequently, significantly, to 9mm/1st hr, after four weeks of ATT, Plain X-Ray KUB showed radio opaque shadow with peripheral calcification of kidney at right lumbar region (Fig.2). On CT fistulogram, a nephrocutaneous fistula was demonstrated in the subcutaneous and muscle planes, leading from the external openings (two in number) of the fistula on the right flank of abdomen, forming common single tract ,6cm in length, to the pelvi calyceal system of right kidney which showed an obstructing large calculus in its pelviureteric junction, alongwith patchy calcification (Fig.3 and Fig.4). This kidney was non functioning and the other kidney was normally functioning (Fig.5). The patient was put on four anti tubercular drugs Isoniazid, Rifampicin, Pyrazinamide and Ethambutol. The patient was advised surgery after a period of four to six weeks of anti tubercular treatment. Patient presented in follow up after six weeks of ATT, with the history of decrease in the discharge from the fistula, patient was not willing for surgery at that time and was advised to continue with the ATT. Patient showed signs of improvement as the discharge from the fistulous tract had decreased. She was shifted to an antitubercular regime of three drugs. She again turned up in the out patient department after about twelve weeks of anti tubercular treatment and on examination the fistula had healed as there was no discharge from its external opening and the fistulous openings had closed for the preceding two weeks (Fig.6). The follow up CT urogram showed the obliteration of fistulous tract but the rest of the findings were same (Fig.7). Patient felt better clinically and was counseled for surgery again for which she consented this time. Total nephrectomy (open) with excision of the ureter and fistulous tract was performed (Fig.8). Meticulous dissection was done as there were very dense adhesions to the surrounding structures like retroperitoneum, peritoneum and inferior vena cava. The cut section of the kidney showed calcification, pus pockets, caseous material, dilated pelvicalyceal system and stone obstructing the pelvis (Fig.9). The post operative period was uneventful. The histopathology report confirmed renal tuberculosis. The patient was advised antitubercular treatment (isoniazid, rifampicin and ethambutol) for a further period of four months and was instructed standard follow up. She was having no complaints till her latest follow up after almost 7 months of surgery.

DISCUSSION

Out of all cases of extra pulmonary tuberculosis, Incidence of urogenital tuberculosis is 4-73% (Perez *et al*., 2006, Langemeier., 2007; Cahill *et al*., 2001). In the present case pulmonary tuberculosis was ruled out by Chest X-ray, which was normal. Kidney is the commonest affected urogenital organ and In cases of renal tuberculosis, Lower Urinary Tract Symptons (LUTS) are seen in 11-88% and haematuria, usually microscopic, in 15-55% of patients (Zarrabi., 2009), constitutional symptoms may be there, in the present case none of these symptoms/signs were present except ballotable right flank mass/pseudotumour on the right lumbar area, which may also be one of the features (Zarrabi., 2009), was noted on clinical examination. Almost In all cases of SCNF an obstructing staghorn calculus is present (Charles., 1990), as was

also the finding in the present case (Fig. 2&5). All patients of SCNF described in literature had nephrolithiasis (Bryniak., 1983, Sarmiento., 1990, Singer.,2002). The present case also falls in this line. Physical examination and CT fistulogram demonstrates the condition best (Rubillota *et al*., 2014), and was done in the present case also confirming the condition vividly (Fig.3&4), preoperative CT showed active fistula (Fig.3&4) whereas the follow up CT after 6 weeks of ATT showed obliteration of the fistula (Fig6&7). The condition is more prevalent in patients of diabetes mellitus, neoplasia and immuno compromised status (Alberto., 2004) but the present patient had none of these conditions. Patient was put on ATT prior to the Surgery on the basis of strong clinical suspicion, substantiated further by positive PCR test for tuberculosis, raised ESR and radiological findings. PCR is more sensitive than mycobacterium culture which takes long time,6-8 weeks. The sensitivity of PCR is 75-94 % (wise., 2008), takes about 24- 48 hrs but is expensive. we were convinced of the tubercular origin of this SNCF when after 10 weeks of ATT patient showed significant improvement and even the fistula had healed (Fig. 6).

Cavity formation and scarring of renal pelvis (Zarrabi., 2009) are other important signs of renal tuberculosis and were also present in the, resected specimen alongwith calcifications and stones (Fig.9), which further supported our diagnosis. Calcification is seen on plain x-ray in 25-50% of cases of renal tuberculosis (amorphous, granular, curvilinear, ring like in papillary necrosis). Peri renal abscesses, one of the causes of SNCF may arise from adjacent structures or from kidney itself. These fistulae can also develop between kidney and pleural cavity, lungs, bronchial tree and gut (Rubilotta *et al*., 2014). Majority of these fistulae drain through the lumbar region i.e. the lumbar triangle (petit), as in this case, and lumbar quadrilateral (grynfeld), these locations being the areas of lowest resistance (Sarmiento., 1990). Timely nephrectomy prevents further morbidity and cure this debilitating condition (Ansari., 2004).Total nephrectomy (open) along with excision of the fistulous tract was done in the present case (Fig.9) being the standard treatment described in literature (Sharma., 2013,Tanwar.,2016). The optimum time for surgery is between 6-12 weeks after the start of ATT (Christopher., 2013), a delay of 4-6 weeks of ATT has been advised in an another study (Mc Aleer., 2007). Nephrectomy with complete debridement of affected peri renal fat, muscles & sub cutaneous tissue, with excision of tract is recommended. There may be dense adhesion with Great vessels, posterior Peritoneum and adjacent viscera, as was also seen in the presenting case. The excision of the fistulous tract in this patient did not involve much additional major surgical procedure or additional morbidity and did not affect the post operative outcome. Other options described in the literature are partial nephrectomy (Singer., 2002), hemi nephrectomy in duplicated cases (Das.,1979) and conservative therapy (Hitter.,1988).

Conclusion

Spontaneous nephro cutaneous fistula is a rare entity arising out of nephrolithiasis and renal infections but in a country like India where tuberculosis is quite common, the possibility of tubercular cause must be ruled out. We suggest that, before surgery is undertaken, in such cases of spontaneous nephrocutaneous fistula of tubercular origin, these patients must be put on anti tubercular treatment for a minimum period of four to twelve weeks, as the fistula heals and general condition improves, thus lessening morbidity, as active tuberculosis hinders healing and produces more morbidity in the form of non healing and recurrence. The active process is subdued and is controlled, thereby surgery becomes easier and less morbid. Open surgery, as undertaken in the present case, is recommended for this condition, rather than minimally invasive techniques. Complete course of anti tubercular treatment should be given to these patients. Clinical diagnosis of SNCF is difficult as there are no specific symptoms, therefore high degree of suspicion of this condition in such patients is important. The clinician should consider the possibility of this condition when the patient presents with a chronic discharging sinus on the lumbar areas.

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