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Renal Actinomycosis: An Unexpected Diagnosis for Unusual Symptoms

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Author's contribution

The sole author designed, analysed, interpreted and prepared the manuscript.

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Case Study

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ABSTRACT

Actinomycosis is an uncommon chronic granulomatous infection caused by gram-positive anaerobic bacterium, Actinomyces israelii (A. Israelli).There are a very few cases of renal actinomycosis (RA) as clinical findings of genitourinary actinomycosis are usually non-specific. Here we present two cases of RA which presented with urinary tract infection and one of them was suspected to have neoplastic lesion on radiology.

Keywords: Renal; actinomycosis; sulphur granules.

1. INTRODUCTION

Actinomycosis is an uncommon chronic granulomatous infection caused by gram-positive anaerobic bacterium. The disease has a worldwide distribution with greater prevalence internationally in populations with low socioeconomic status [1,2]. Actinomyces israelii (A.Israelli) is the most common human pathogen amongst all pathogenic Actinomyces species [3,4,5,6]. The most common clinical forms of actinomycosis are cervicofacial (ie, lumpy jaw), thoracic, and abdominal with very few cases of renal actinomycosis (RA) [1,3,7]. Clinical findings of genitourinary actinomycosis are usually non-specific and may include abdo`minal pain, urinary frequency or repetitive cystitis [1,3]. Microbiologic identification of the etiological agents causing actinomycosis is done in only a minority of cases. Actinomycosis is primarily

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diagnosed by the presence of sulfur granules in pus or histologic sections of a surgical specimen [4,6,7]. Here we present two cases of RA which presented with urinary tract infection and one of them was suspected to have neoplastic lesion on radiology. Both the patients underwent simple nephrectomy and histology revealed presence of basophilic colonies surrounded by inflammatory reaction.

2. CASE PRESENTATION

Case 1: A 62 year old man, with type-2 DM for last 10 years on insulin for last two years. He presented with pain over the right flank region since 1 month, low grade intermittent fever since 20 days and burning micturition since 4 days. On routine urine examination, urine appeared hazy, had acidic pH and specific gravity of 1.040. Microscopic examination revealed plenty of pus cells, RBCs (40-50/HPF-20% crenated) and bacteria. On culture studies, E.coli was isolated. Glycosylated hemoglobin was 9.6%, fasting glucose level 140 mg/dL and post-meal glucose levels were 235 mg/dL. Serum creatinine (SCr) was 1.6 mg/dL, serum Na+ was 144.0 mEq/L and K+ was 4.5 mEq/L. The total leucocyte count (TLC) was raised (15.8 x 10^3/ µl) with rise of neutrophils (absolute neutrophil count: 11.8 x 10^3/ µl). CT scan revealed grossly hydronephrotic right kidney (size 15.9 x 9.4 x 5.8 ccm) with thinning of the parenchyma and walls of upper and middle calyces showing irregular enhancing thickening with multiple filling defects. The left kidney measured 12.5 x 4.5 x 4.0 ccm with preserved corticomedullary differentiation. The radiological features were suggestive of neoplastic lesion. Laparoscopic guided nephrectomy was performed and was sent for histopathological examination.

Case 2: A 35 year old non-diabetic man with history of recurrent renal calculi over the last 5 years presented with high grade fever and dysuria for last 5 days. The patient had undergone lithotripsy twice for renal calculi. On routine urine examination, it appeared hazy, with specific gravity of 1.010, acidic pH and proteinuria of +1 (on scale of 0 to +4). Microscopic examination revealed plenty of pus cells, RBCs (60-70/ HPF-10% crenated) and bacteria. On culture E.coli was isolated. SCr was 1.8 mg/dL. Hemoglobin was 15.2 gm/dL, TLC was raised (25.8 x 10^3 /µl) with rise of neutrophils (absolute neutrophil count: 20.6 x 10^3/ µl). Ultrasonography of the abdomen and pelvis showed shrunken left kidney (size: 10.5 x 3.5 x 3.0 ccm), with thinned out cortex, dilated pelvi-calyceal system and a staghorn calculus measuring 22 x 11 x 6 mm. Laparoscopic guided nephrectomy was performed and was sent for histopathological examination.

2.1 Pathological Examination

2.1.1 Gross findings

Case 1: The right nephrectomy specimen weighed approximately 340 grams, measured 16.5 x 9.5 x 6 ccm, and was pale brown in colour with irregular scars on external examination. The renal capsule was focally adherent. On sectioning, the cortico-medullary junction (CMJ) was ill-defined and pelvi-calyceal system appeared dilated measuring 6.5 x 4.5 cm. Multiple abscesses were noted involving the cortex and calyceal walls, yellowish in colour with ragged outlines, having granular appearance. Stump of ureter and renal vessel identified were unremarkable.

Case 2: The left nephrectomy specimen was received in multiple irregular pieces of varying sizes, altogether weighing 85 grams, measuring 10.0 x 4.5 x 3.5 ccm. The external surface appeared pale brown with irregular 'U-shaped' scars. CMJ was indiscernible. Calyces identified in few of the pieces appeared dilated. Scattered yellowish areas were identified on the wall of calyces studded with yellow granules. One of the preserved calyx showed a calculus, measuring 2.0 x 1.5 x 0.8 ccm, brown in colour having irregular surface. Segment of ureter identified measured 5.5 cm in length.

2.1.2 Microscopic findings

Multiple sections were studied from both the specimens from various parts of cortex and pelvi-calyceal system were stained with hematoxylin and eosin, periodic acid Schiff's, Jone's methaneamine silver, Gram's and modified Ziehl Neelsen stains. About 80-85% glomeruli of the first case and 90% of the second case were globally sclerosed. The scattered viable glomeruli revealed mild mesangial prominence with partially obliterated capillary lumina lined by thickened and wrinkled membranes. Bowman capsules were thickened and revealed variable degree of periglomerular fibrosis. Majority of the tubules revealed classical atrophy with thickened and irregular basement membranes and tubular lumina were filled with proteinaceous casts. Arteries revealed moderate fibrointimal

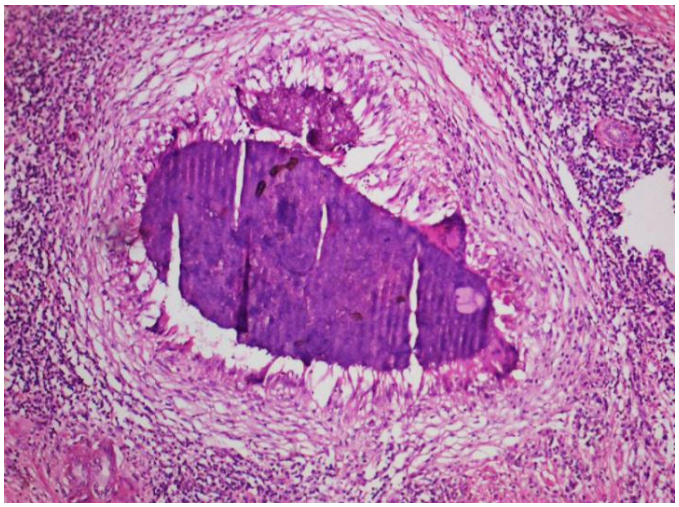

Fig. 1. Section from the left nephrectomy specimen showing granulomatous inflammation surrounding the basophilic actinomycotic colony. The colony is surrounded by neutrophils along with eosinophils, plasma cells and foreign body type of giant cells (Hematoxylin and eosin; X 100)

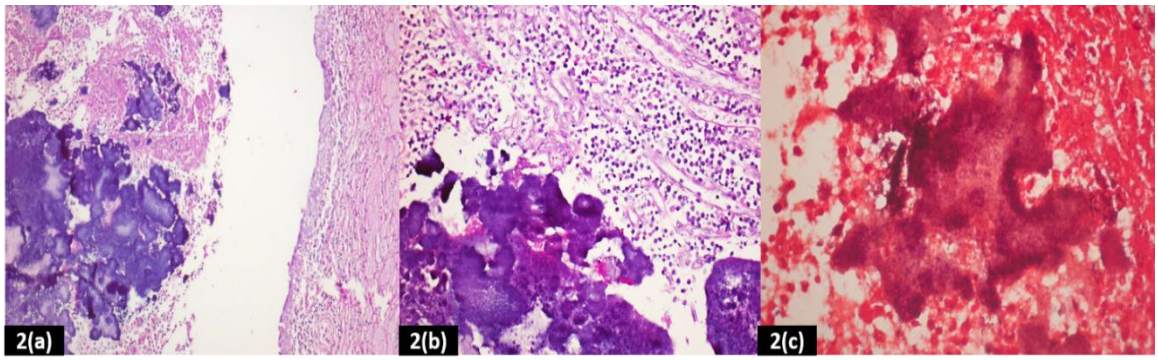

Fig. 2. Section showing basophilic colonies of actinomycosis, surrounded by necrotic material and mixed inflammatory cells adjacent to the urothelial lining of the pelvis. (b) Renal tubules interspersed between the basophilic actinomycotic colonies and inflammatory cells (PAS, X 200); (c) Gram positive organisms (Gram stain, x 1000)

proliferation. The interstitium revealed scattered basophilic actinomycotic colonies surrounded by neutrophils, eosinophils, lymphoplasmacytic cells and foreign body type of giant cells. The basophilic colonies seen on H&E stains appeared PAS positive and argyrophillic on silver stain. These appeared as blue filamentous structures on ZN stain and gram positive on Gram's stain (Figs. 1 & 2).

Both the patients were treated with intravenous antibiotics along and supportive medications were given as medical management for first seven days. The total count on follow-up was 6.5 x 10^6 µl and 4.8 x 10^6 µl 15 days after surgery. Urine routine was unremarkable and culture studies did not yield any organisms.

3. DISCUSSION AND CONCLUSION

Actinomycosis is a subacute-to-chronic bacterial infection caused by filamentous, gram-positive, non–acid-fast, anaerobic-to-microaerophilic bacteria and is characterized by suppurative and granulomatous inflammation, with formation of multiple abscesses and sinus tracts that may discharge sulfur granules [8,9]. These organisms belong to the order of Actinomycetales, family Actinomycetaceae, genus *Actinomyces (A.).* The most common isolated species are *A. israeli, A. gerencseriae, A. turicensis, A. radingae* and *A. europaeus,* followed by *A. naeslundii, A. odontolyticus, A. viscosus, A. meyeri,* and *Propionibacterium propionicum* [8-10]. In the humans, actinomyces occur as a

harmless saprophyte in the mouth, tonsils and gastrointestinal tract, causing disease under special circumstances, during trauma, viscous perforation or surgery [1,3]. Infection can occur at all ages with a peak incidence reported in the mid-decades (20-60 years) with a male predominance. [2,7]. Actinomycosis is often called as “the most misdiagnosed disease” as it can be often missed by an experienced clinician [11]. The most common presentation is cervico-facial (50-70%), followed by thoracic (15-20%) and less commonly the abdomino-pelvic form (10-20%) [8]. Genitourinary involvement appears only sporadically and mostly manifests as renal or perirenal masses [10]. Various predisposing factors include diabetes mellitus, immunosuppressed state, long term use of contraceptive intrauterine devices (pelvic disease), previous surgery (post-appendectomy abdominal actinomycosis), dental extraction, trauma and poor dental hygiene (cervico-facial disease). Rarely Actinomyces can present as retroperitoneal or scrotal mass or cause ureteral stenosis, prostatitis, penile pilonidal sinus and cystitis. A few case reports have reported RA associated with nephrolithiasis [12,13]. The route of infection in RA is still controversial. It is hypothesized to occur as direct spread from contiguous structures such as lungs or colon and aspiration of infected material, and also secondary to hematogenous dissemination. Four type of renal lesions have been described: chronic suppurative lesion (carbuncle), renal mass, pyelonephritis and pyonephrosis [13,14]. Radiologically the disease can appear as an aggressive infiltrative process with indistiguishable anatomic boundaries giving a suspicion of an invasive renal tumor [2] Intravenous urography (IVU), angiography, and ultrasonography, are not diagnostic. The diagnosis is suspected with histological findings since cultures are often unsuccessful, being positive in less than 50% of cases [14].

A definitive diagnosis of actinomycosis is made after histologic identification of sulfur granules in the biopsy specimen or aspirated pus and is confirmed with Gram-stained smears and anaerobic cultures [5]. Histologically, lesions produced by A. israelii, lesions produced by A. israelii, tend to develop multiple loculations at three weeks and contain islands of bacterial masses surrounded by neutrophils and enveloped by a well-organized capsule composed of mature collagen fibers with accompanying fibroblasts and some inflammatory cells. Lesion in the later stages shows plasma cells. Gram-stained sections from these resolving lesions contain scattered, beaded, gram-positive bacteria filaments surrounded by neutrophils [5]. Traditionally the treatment of RA has been urological extirpation of the kidney followed by prolonged antibiotic therapy. However, in early stages, solitary RA may be successfully treated using prolonged antibiotic therapy (8 weeks-1 year) [3,15].

Both our patients presented with UTI, with one of them having a lesion of neoplastic origin on radiology and other a case of nephrolithiasis leading to non-functioning kidney. Both the patients had to undergo nephrectomy. Gross examination of the kidneys from both the patients revealed yellowish granular abscesses (sulphur granules) which corresponded to actinomycotic colonies on microscopy. Both the patients responded to antibiotic therapy post-nephrectomy.

Thus diagnosis of RA remains a challenge for the clinician, radiologist and microbiologist; histopathology still remains the only diagnostic modality in confirming the diagnosis.

CONSENT

As per international standard or university standard, patients’ written consent has been collected and preserved by the author(s).

ETHICAL APPROVAL

As per international standard or university standard written ethical approval has been collected and preserved by the author(s).

COMPETING INTERESTS

Author has declared that no competing interests exist.

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