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Case Report
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A Rare Case of Malakoplakia Masquerading as a Recurrence of Surgically Treated Renal Cell Carcinoma

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Abstract

A 57 year old woman presented with ongoing pain and a mass in her left loin approximately four months after undergoing laparoscopic radical nephrectomy because of a left renal mass. She had a large mass at the site of her resection which continued to grow and cause destruction of bone, in this case her left sacral wing. However, the histology showed that this was not a recurrence but Malakoplakia secondary to a wound infection. To the best of our knowledge, Malakoplakia stemming from a urinary tract source this extensive and including bone destruction, has never been reported before.

Keywords: Malakoplakia; Multi-disciplinary team; Renal cell carcinoma

Introduction

The current report represents a rare case of malakoplakia which presented after a left-sided laparoscopic radical nephrectomy. Malakoplakia represents a differential diagnosis which needs to be considered in these cases, since despite an appropriate source control with drainage of collections and antibiotics; this type of infection might increase fast and significantly. Finally, the presented woman underwent a surgically open exploration of the left retroperitoneum to exclude recurrent disease (which might not have been necessary if the patient would have been treated with antibiotics adequately, that are highly effective in malakoplakia).

Case Presentation

A 57 year old female initially presented to the Department of Urology of a general district hospital with visible haematuria and a CT revealed a left renal tumour. Her past medical history included non-insulin dependent diabetes and hypertension, which were both controlled with medications.

After Multi-Disciplinary Team (MDT) discussion she underwent a left laparoscopic radical nephrectomy. Histology showed a clear cell renal cell carcinoma, Grade 3 (pT2a N0M0). Histopathology confirmed there were negative surgical margins. Postoperatively she made an uneventful recovery and was discharged three days after the operation. Nevertheless, she complained of ongoing discomfort around her wound and also left sided abdominal pain at the time of discharge.

Almost four months later she presented with severe anaemia and considerably worsened abdominal pain. On examination a left iliac fossa mass was palpated. CT revealed a 10 cm left psoas abscess and CT guided drainage was done. After twelve days, she was discharged with the drain-*in-situ*. The drain was removed after two days, when it was draining sufficiently small amounts.

She re-presented two months after this with worsening pain in the left loin and left iliac fossa and there was a recurrence of the left iliac fossa mass. She had another CT (Figure 1) that showed extensive para-aortic lymphadenopathy extending into iliopsoas and left Sacroiliac joint with abscess formation. Changes were reported to have increased compared to the previous scan. A drain was inserted on the ward and 200 mls of pus was drained. CT guided biopsy of retroperitoneal mass was undertaken and histologically a diagnosis of malakoplakia was made. This was the same pathologist who diagnosed the original cancer after the patient's nephrectomy. After an inpatient stay for two weeks, the patient was discharged.

Within a couple of weeks, the patient was readmitted with collapse, general malaise, lethargy and worsening left loin pain. Despite histologically confirming a diagnosis of malakoplakia, there was mounting concerns that this was a recurrent tumour. After an MDT discussion the general consensus was that a proper histological diagnosis was needed before labelling the findings as a recurrent tumour as it could still be an ongoing inflammatory/infectious process. A repeat CT

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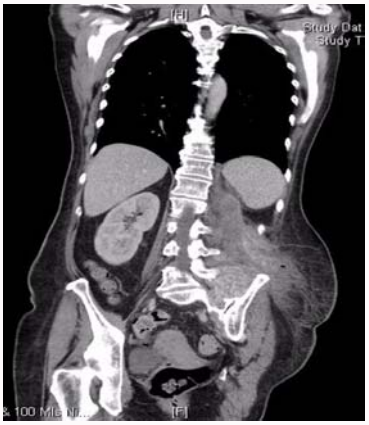

Figure 1: This is a CT slice showing extensive inflammation with para-aortic lymphadenopathy extending into iliopsoas and left Sacroiliac joint with abscess formation.

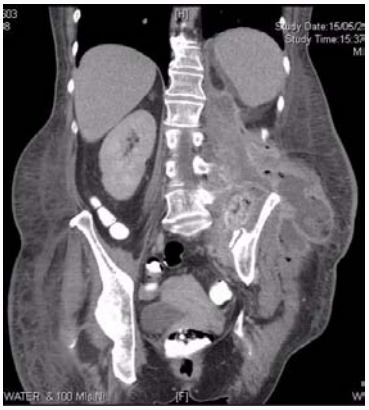

Figure 2: This is a CT slice showing there is an irregular enhancing soft tissue with extension into left groin pushing iliac vessels medially. This CT also showed the mass was also extending into the posterior abdominal wall and was causing destruction of the left sacral wing extending into left sacral iliac joint and left iliac crest. Additionally, the medial margin of this mass was abutting the aorta over a long length.

biopsy was done which again confirmed malakoplakia. An additional detailed CT was conducted (Figure 2). This stated that the entire left rectoperineal area was abnormal with irregular enhancing soft tissue with extension into left groin pushing iliac vessels medially. This mass was also extending into the posterior abdominal wall and was causing destruction of the left sacral wing extending into left sacral iliac joint and left iliac crest. Additionally, the medial margin of this mass was abutting the aorta over a long length. From these images it was felt that this mass represented a huge incurable retroperitoneal renal cell tumour recurrence complicated by secondary infection.

An open exploration of the left retroperitoneum was done and findings were of a hard, inflammatory area with no collection to drain. Extensive biopsies were taken and 200 mls of pus were drained from a separate gluteal incision. No fungi or acid fast bacilli were seen on special staining.

The sarcoma unit (a tertiary referral specialist centre unit specialising in sarcoma) was involved as it was felt she may have possible osteomyelitis complicating an extensive, invasive retroperitoneal and a left retro-pelvic mass that was confirmed to be malakoplakia. Additionally there was continuing evidence of chronic local sepsis. The sarcoma unit recommended a Contrast CT and

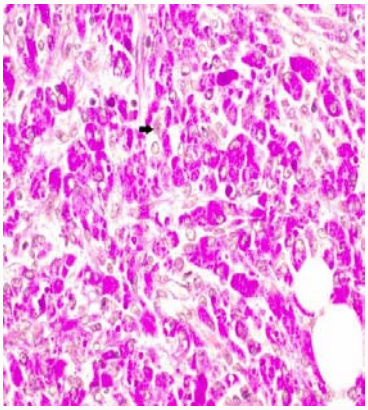

Figure 3: This is a Histology slide of the specimen using the periodic acid-Schiff stain. This shows Michaelis-Gutmann bodies (histiocytes with basophilic inclusions with concentric laminations) which are diagnostic.

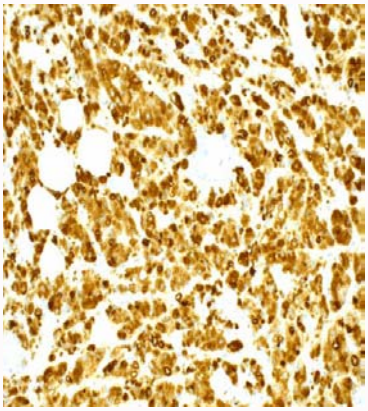

Figure 4: This is an immunohistochemical study of the specimen using CD68 antibodies. This positive stain for CD68 antibodies suggests that the Michaelis-Gutmann bodies (which consist of lysosomes filled with partially digested bacteria) contain gram negative bacteria.

defunctioning colostomy as they had considered the possibility of bowel leak that was persisting to account for the chronic sepsis. The CT did not show any colonic leak and therefore no further operations were undertaken.

Investigations

The patient's haemoglobin continued to be low and she required several blood transfusions. Urine cultures had always shown a coliform infection and multiple CTs demonstrated that the mass was increasing in size and causing bone destruction (Figures 1 and 2). Blood tests were all normal apart from raised inflammatory marker (CRP), which settled after appropriate treatment.

Histology showed classic Michaelis-Gutmann bodies consistent with malakoplakia (Figures 3 and 4). There was no positivity for pan cytokeratin, CK7 or CK20 further confirming there was no histological evidence to suggest carcinoma. Her cultures from the biopsies showed coliforms resistant to most oral antibiotics but sensitive to Ciprofloxacin.

Outcome

During the course of the disease this patient has been treated with multiple antibiotics, including IV Tazocin and IV Teicoplanin. However, she did make a good recovery when she was switched to ciprofloxacin.

The patient had initially reduced mobility and chronic pain from the bony retroperitoneal involvement. These symptoms all resolved with minimal sequelae after the appropriate antibiotics and physiotherapy.

Her follow-up after the Malakoplakia was initially based around her infection. However, this then returned to her long-term follow-up which was based on the guidelines for her intermediate risk renal cell carcinoma, so no follow-up images were taken for her Malakoplakia.

Discussion

Malakoplakia is a chronic granulomatous inflammatory disorder associated with an infectious etiology [1], usually involving the urinary bladder, though it has been reported affecting the kidneys, respiratory system and digestive system [2]. It is often associated with those patients who are immunocompromised, either iatrogenically by immunosuppressant drugs (e.g. post-transplant) or secondary to their co morbidities (e.g. Diabetes, malignancy, immune deficiency states, and alcohol abuse) [3]. Clinically, as well as radiologically malakoplakia produces tumour-like nodules that are able to mimic malignant neoplasms, thus a confirmation of the diagnosis can only be achieved histologically [4].

Since its original description in 1901 by von Hansemann [5], over 400 cases of Malakoplakia have been reported. The reason an infective source has been linked to malakoplakia is twofold: first, within the cytoplasm of macrophages large calcified structures called Michaelis-Gutmann bodies are present, which are believed to develop as a result of ineffective digestion of bacteria [3]. Second, when examined with an electron microscope you can find coliform bacteria within phagolysosomes of these macrophages we have described above [4]. This is obviously not the whole story, as although having a UTI's in the UK with *E. coli* is common, malakoplakia is rare, therefore there has to be another reason for this condition to occur.

There is still uncertainty about the pathogenesis of the disease, however one theory is that there are defective killing and impaired digestion of phagocytosed bacteria [6]. This is suggested by the characteristic intracellular abnormalities within the macrophages [4]. Ciprofloxacin is more effective in this condition as unlike other antibiotics which are effective on *E. coli*, ciprofloxacin has shown to penetrate well into macrophages [4].

Malakoplakia that involves the urinary tract has a 4:1 female predominance with a peak incidence of over 40 years [7,8]. Relevant common laboratory findings include anaemia. The clinical presentation is variable and non-specific, however most commonly these patients present with flank pain, fever and a palpable abdominal mass [3].

Due to the clinical and radiological appearance mimicking neoplasia then it is important to get a histological diagnosis. Malakoplakia managed medically with antibiotics, therefore avoiding the complications and morbidities, as well as the risk of mortality, which are associated with a nephrectomy.

Learning Points

1. Malakoplakia can masquerade as cancer on radiological appearances and early biopsy to confirm the diagnosis is important to ensure early treatment like antibiotics, esp. ciprofloxacin, which is very efficacious in Malakoplakia as in the case above.

2. Awareness that surgical site infection could cause Malakoplakia, which is a rare, unreported post-operative infective complication of a laparoscopic nephrectomy for cancer.

3. To be aware that malakoplakia of urinary tract origin can result in severe, extensive retro-peritoneal malakoplakia involving bone. This has not been reported before.

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