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Case Report

Case Report of Pelvic Xanthogranulomatous Inflammation- Mimicking A Relapsed Malignancy

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Abstract

Xanthogranulomatous inflammation is a chronic inflammatory process characterised by lipid-filled histiocytes. It is indistinguishable from other infiltrative processes such as malignancy radiologically, but definitive diagnosis hinges on histopathology with immunohistochemistry. While some cases resolve spontaneously, some lead to strictures or visceral perforation. As a result, only some need treatment by either antibiotics or surgery. We present a self-resolving case of pelvic xanthogranulomatous inflammation post-endometrial cancer and adjuvant chemoradiotherapy.

Keywords: Xanthogranulomatous Inflammation; Endometrial Cancer; Radiotherapy

Introduction

Xanthogranulomatous inflammation is an under-recognized entity that may mimics conditions such as malignancy or other infiltrative processes. This can affect any organ but is most commonly reported in the gallbladder and the kidneys. We present a self-resolving case of pelvic xanthogranulomatous inflammation diagnosed on histopathology, which was initially thought to be a recurrence of endometrial cancer post adjuvant chemoradiotherapy.

Case Presentation

A 66-year-old ethnic Chinese female presented with intermittent lower back pain for 2 weeks.

She had a history of endometrial cancer and had undergone transabdominal hysterectomy, bilateral salpingo-oophorectomy and pelvic lymph node dissection with adjuvant chemoradiotherapy 6 years prior and was on regular surveillance. The cancer was in remission but she had longstanding bilateral lower limb lymphedema post-surgery.

She presented with diffuse lower back pain at rest for 2 weeks. There was no weakness or numbness in bilateral lower limbs and no bladder or bowel symptoms. There was no prior trauma. She did not have any fever, night sweats, loss of appetite or loss of weight. However, she did have bilateral flank swelling in addition to her chronic lymphedema for the same period. She had no history of tuberculosis contacts.

On examination, she was afebrile and her vitals were stable. Her abdomen was soft and nontender though there was some lower abdominal distension. There was no costovertebral angle tenderness. Examination of the spine was normal, range of movement of bilateral hip joints was full, and lower limb lymphedema was stable. Her neurological examination was intact and the rest of physical examination was unremarkable.

Her peripheral white cell count was 5.80X10^9/L (4.30-10.40 X10^9/L), haemoglobin 10.2 g/dL (11.5-14.5 g/dL), albumin 30g/L (32-46 g/L), renal and liver function were normal,. Xray of the lumbosacral spine was normal. Contrast enhanced CT abdomen and pelvis (Figure 1) revealed abnormal enhancing soft tissue in the left psoas, left iliacus and gluteal musculature (white arrows) with erosions in the left sacro-iliac joint (white star), suggestive

of an aggressive etiology. A FDG PET study (Figure 2) showed areas of increased FDG uptake in the left psoas, left iliacus and gluteal musculature (white arrows) corresponding to the areas of soft tissue enhancement on the CT scan.

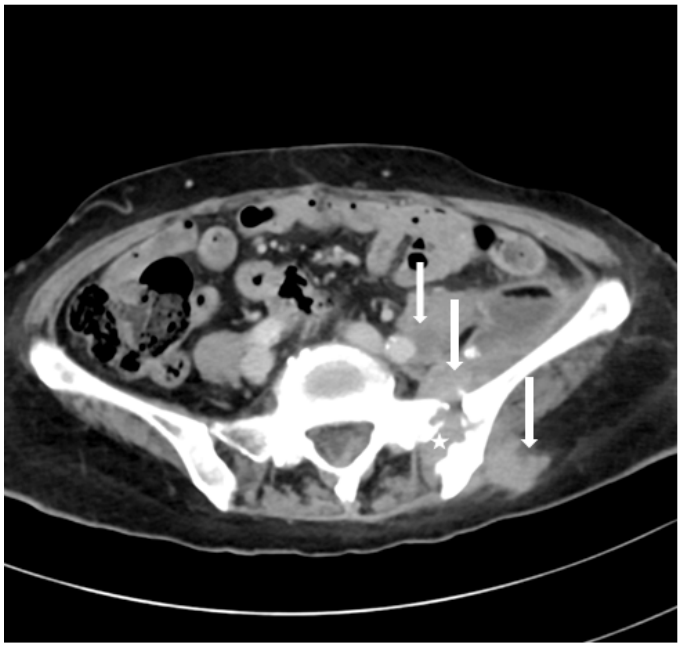

Figure 1: Contrast enhanced CT study shows abnormal enhancing soft tissue in the left psoas, left iliacus and gluteal musculature (white arrows) with erosions in the left sacro-iliac joint (white star), suggestive of an aggressive etiology.

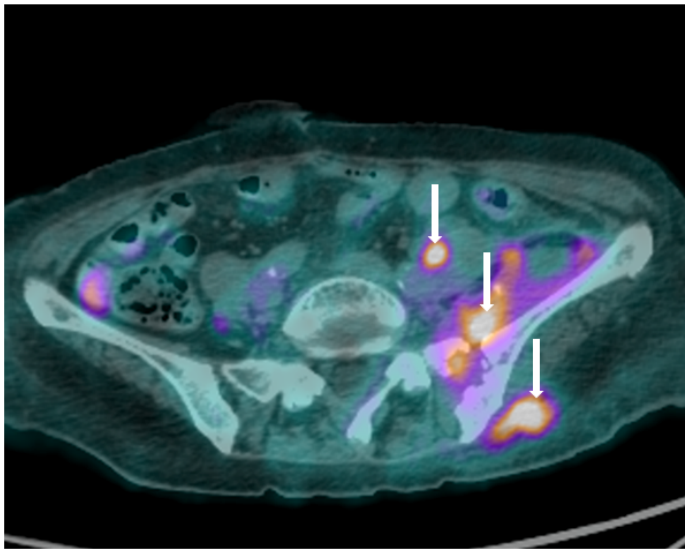

Figure 2: FDG PET study showing areas of increased FDG uptake in the left psoas, left iliacus and gluteal musculature (white arrows) corresponding to the areas of soft tissue enhancement.

A biopsy of the left gluteal mass was performed in view of concerns for relapse of cancer. Histopathology revealed foamy histiocytes admixed with chronic inflammatory cells including lymphocytes, plasma cells, and intervening fibrous stroma. Immunohistochemical stains for CD 116 show diffuse positivity of the histiocytes. CD1a staining was negative which excluded an abnormal proliferation of Langerhans cells. AFB smear, cultures, and TB PCR were negative as were the fungal smears and cultures. Whole genome sequencing on the biopsy samples did not yield any significant infective pathogens. Overall features were consistent with xanthogranulomatous inflammation.

The patient was followed up closely and her symptoms and pelvic xanthogranulomatous inflammation resolved spontaneously without any surgical intervention or antimicrobial treatment. A repeat CT pelvis 6 weeks later (Figure 3) showed almost complete resolution of the enhancing soft tissue in the left psoas, iliacus and gluteal musculature. Residual soft tissue thickening was noted with stable erosions in the left sacroiliac joint. There was no interval progression in another repeat CT pelvis 6 months later and the patient remained clinically well.

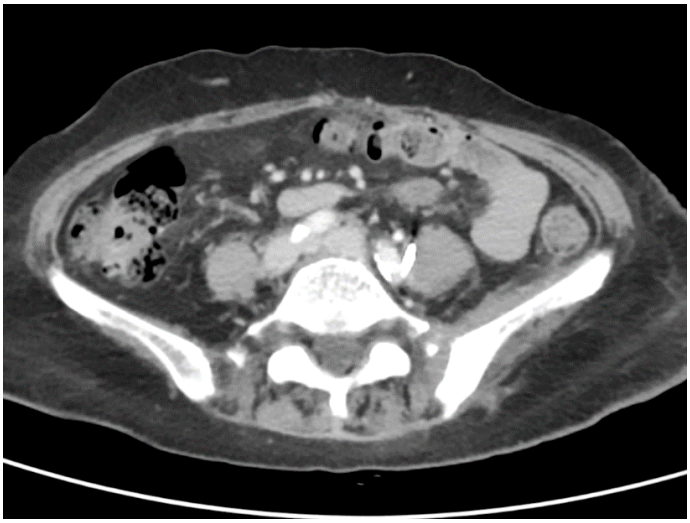

Figure 3: Contrast enhanced CT scan of the pelvis shows almost complete resolution of the enhancing soft tissue in the left psoas, iliacus, and gluteal musculature.

Discussion

Xanthogranulomatous (XG) inflammation is a chronic inflammatory process that results in surrounding tissue destruction characterized by lipid-filled macrophages or histiocytes and is most commonly reported in cholecystitis and pyelonephritis [1]. However, other abdominopelvic organs can be involved, including the stomach, pancreas, terminal ileum, appendix, colon, spleen, mesentery, bladder, ovaries, fallopian tubes, uterus, prostate, testes and adrenals [2].

The chronic inflammation in xanthogranulomatous inflammation is postulated to be due to activation of immune cascade causing formation of abscess, haemorrhage or necrosis, contributed by defective lipid transport, disorders of neutrophilic chemotaxis, abnormal lymphatic flow and immune response to recurrent infections by Proteus and *E. coli* which are commonly encountered in xanthogranulomatous pyelonephritis and cholecystitis associated with anatomical strictures or obstruction.

Similarly, pelvic xanthogranulomatous inflammation may be a consequence of cervical stenosis and resultant pyometria particularly in post-menopausal females with endometrial hyperplasia or carcinoma, presence of intrauterine devices or manipulation such as endometrial biopsy, or receipt of radiotherapy or chemotherapy [4,5] Superimposed infection with *E coli* and *Proteu*s, *Salmonella typhi*, *Bacteroides fragilis*, *Staphylococcus aureus* and *Mycoplasma hominis* have all been reported [6,7]. In a few cases, xanthogranulomatous inflammation can lead to catastrophic complications including uterine perforation [8]. Xanthogranulomatous salpingo-oophoritis has also been associated with endometriosis, previous inadequately treated pelvic inflammatory disease and infertility [9,10].

Xanthogranulomatous inflammation of abdominal pelvic organs frequently poses a diagnostic challenge. Radiologically, the diffuse inflammatory and fibrotic changes may appear aggressive and cannot be distinguished from a malignant process, chronic infections that cross fascial planes easily such as actinomycetes, endometriosis or malakoplakia. There have been cases of coexisting cancers particularly in patients with history of malignancy [11]. Interestingly, tumour markers such as carcinoembryonic antigen (CEA), carbohydrate antigen 19-9 (CA19-9), and alpha fetoprotein (αFP) may be elevated in xanthogranulomatous cholecystitis. [12] Our patient did have a history of endometrial cancer but no evidence of recurrence either histologically or biochemically.

Typical histological features of xanthogranulomatous inflammation include foamy macrophages (lipid laden histiocytes), multinucleated giant cells, and cholesterol clefts amidst a mixture of mononuclear cells, lymphocytes, plasma cells and neutrophils [13]. These foamy histiocytes may sometimes be mistaken for neoplastic cells on hematoxylin and eosin stain [14]. Thus, the use of immunohistochemistry is crucial: apart from CD116 staining, histiocytes are CD68 positive and cytokeratin negative, with vimentin-positive and cytokeratin-negative reactive fibrous tissue [13].

Fortunately, in our patient, while recurrence of endometrial cancer was certainly a concern, she had biopsy-proven pelvic xanthogranulomatous inflammation with no histological evidence of malignancy. Management of this condition has included surgical excision, antibiotic treatment or a watch-and-wait strategy whereby some patients experience spontaneous resolution, similar to our patient, although there is the possibility of a relapse [4,15,16].

Conclusions

Xanthogranulomatous inflammation is a great mimicker of malignancy or an infiltrative process. Histopathology including immunohistochemistry is crucial in diagnosis. While its pathogenesis is not well understood and treatment is not well studied, our case illustrates the possibility of a conservative strategy with spontaneous resolution in patients without compressive symptoms. Larger observational studies are probably needed to better define the role of interventions in this unusual condition.

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Ethical Guidelines: We have complied with relevant ethical guidelines. Patient consent has been obtained

Conflict of Interest: Nil

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