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Case Report

A rare case of extrapelvic endometriosis in the right sciatic notch[☆]

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ARTICLE INFO

Article history:

Received 26 September 2024

Revised 30 November 2024

Accepted 6 December 2024

Keywords:

Endometriosis

Right sciatic notch

Extrapelvic endometriosis

ABSTRACT

Endometriosis is described as the proliferation of endometrial tissue outside of the uterus. This most frequently occurs within the pelvis and is a common cause of chronic pelvic pain in women of reproductive age. Rarely, endometriosis can manifest outside of the pelvis and can uncommonly involve the musculoskeletal and peripheral nervous systems. Extrapelvic endometriosis is a difficult radiologic diagnosis due to its rarity and varied magnetic resonance imaging (MRI) appearance. Diagnosis of extrapelvic endometriosis is most frequently made with biopsy and most cases within the literature are treated with surgical resection. Of the cases that were treated medically, there is a paucity of available follow-up imaging to characterize the natural history and treatment response of this entity.

We present a case of extrapelvic endometriosis involving the right sciatic notch, in a 26 year old female who presented with cyclic hip pain and lumbosacral plexopathy. Initial MRI findings were nonspecific, and diagnosis was made via image guided biopsy. The patient underwent medical treatment with a gonadotropin-release hormone agonist and oral contraceptives, with close imaging follow-up including a 6-month post-treatment MRI.

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Introduction

Endometriosis is defined as the presence of endometrial tissue outside of the uterus, most commonly within the adjacent

pelvic and adnexal structures. It is present in approximately 1 in 10 females of reproductive age and commonly presents with pelvic pain and dyspareunia [1]. Up to 1 in 5 women with chronic pelvic pain can carry this diagnosis [1]. Rarely, endometriosis can occur in extrapelvic areas such as the ab-

[☆] Competing Interests: The authors declare that they have no known competing financial interests or personal relationships that could have appeared to influence the work reported in this paper.

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<https://doi.org/10.1016/j.radcr.2024.12.009>

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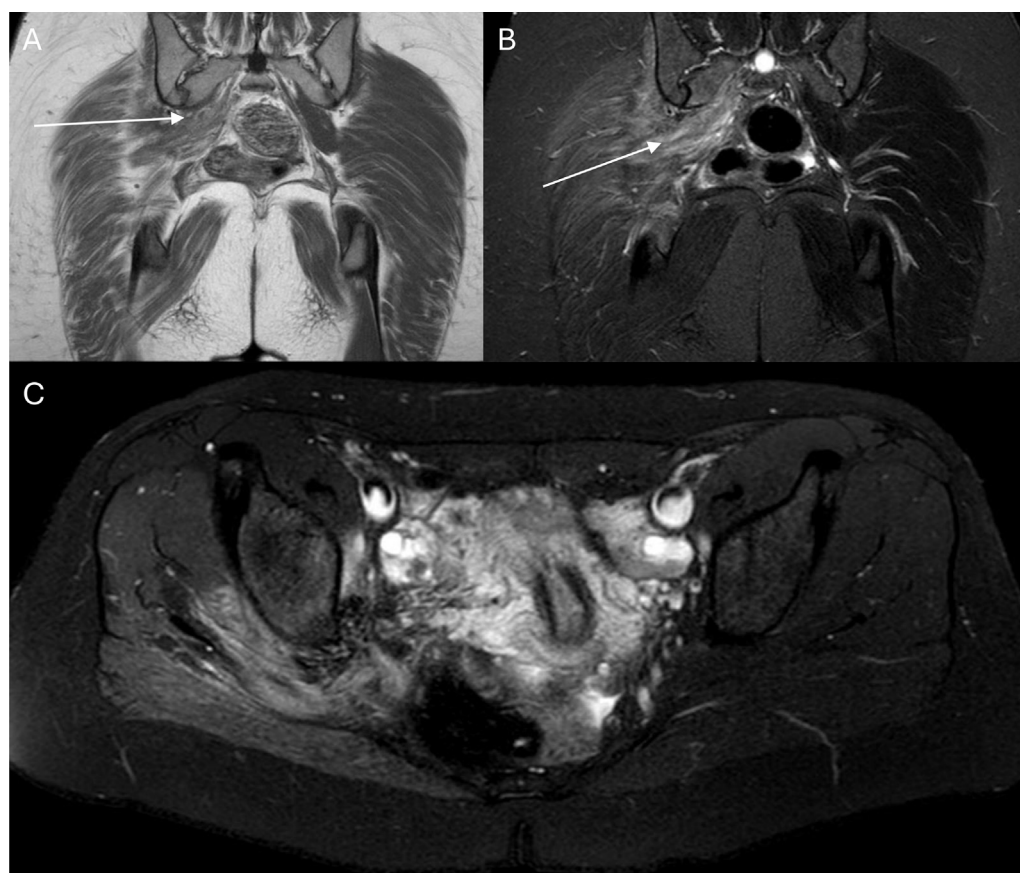


Fig. 1 – Initial MRI: T1 Coronal image demonstrates the presence of an ill-defined isointense enhancing signal involving the right sciatic notch (A). STIR coronal image displays well-defined hyperintense signal within the same region (B). T2-FS axial image demonstrates hyperintense soft tissue inflammatory changes at the level of the right sciatic notch, with hypointense foci thought to represent hemosiderin deposition (C).

domen, thorax, musculoskeletal system, and the peripheral and central nervous systems. Within the musculoskeletal system and peripheral nervous system, the most common presenting symptoms are cyclic localized pain and neuropathic symptoms[2]. MRI findings of endometriosis can vary due to differing degrees of hemorrhagic products within these lesions, and diagnosis is commonly made intra-operatively. The most evidence-based treatment of extrapelvic endometriosis is excision, with or without adjuvant hormonal therapy [2,3]. Notably, there is a lack of follow-up imaging on most reported cases of medically treated extrapelvic endometriosis.

Presented here is a case of extrapelvic endometriosis of the right sciatic notch treated with medical therapy alone and close imaging follow-up.

Case report

A 26-year-old previously healthy female presented with a 2-year history of cyclic right hip pain without a history of trauma. At the time of presentation, physical examination of the pelvis and right lower extremity was unremarkable. Originally the patient was using oral contraceptives; however, 3

years before presentation, the patient had a levonorgestrel-releasing intrauterine device placed. The patient endorsed light and irregular menstrual periods after placement of this device.

Symptoms did not improve despite conservative management with over-the-counter medications, muscle relaxants, or physical therapy over the course of 12 months. MRI of the hip was ordered by the primary provider after the patient began to develop peripheral neuropathic symptoms including right foot and lateral thigh weakness and paresthesia. This study demonstrated an ill-defined, T1 isointense and T2 hypointense lesion in the sciatic notch with gadolinium enhancement and mildly increased signal on fat saturated T1 weighted images (Fig 1). Surrounding this lesion was diffuse intramuscular edema and atrophy of the right piriformis, gluteal, and quadratus femoris musculature. The sciatic nerve was not well visualized within the region of signal abnormality. Foci of T2*-weighted gradient echo hypointense signal within this lesion was felt to represent mineralization. Findings were thought to represent post-traumatic denervation of the sciatic nerve with possible associated myositis ossificans. Additionally, desmoid tumor was included in the differential diagnosis. Computerized tomography (CT) was then performed demonstrating a heterogeneously en-

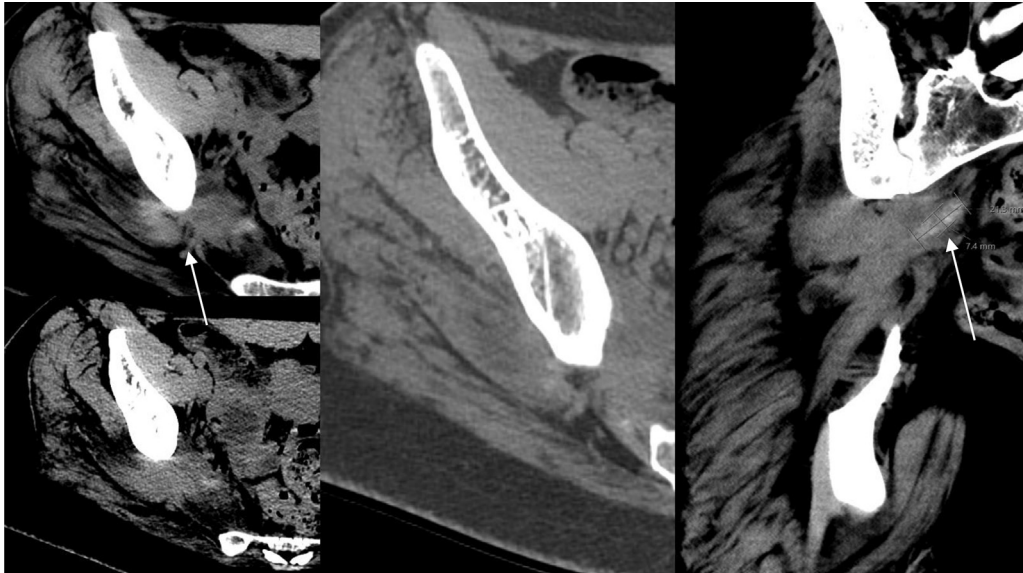


Fig. 2 – Axial and Coronal CT demonstrating a heterogeneous mass-like density involving the right piriformis, gluteal, and quadratus femoris musculature.

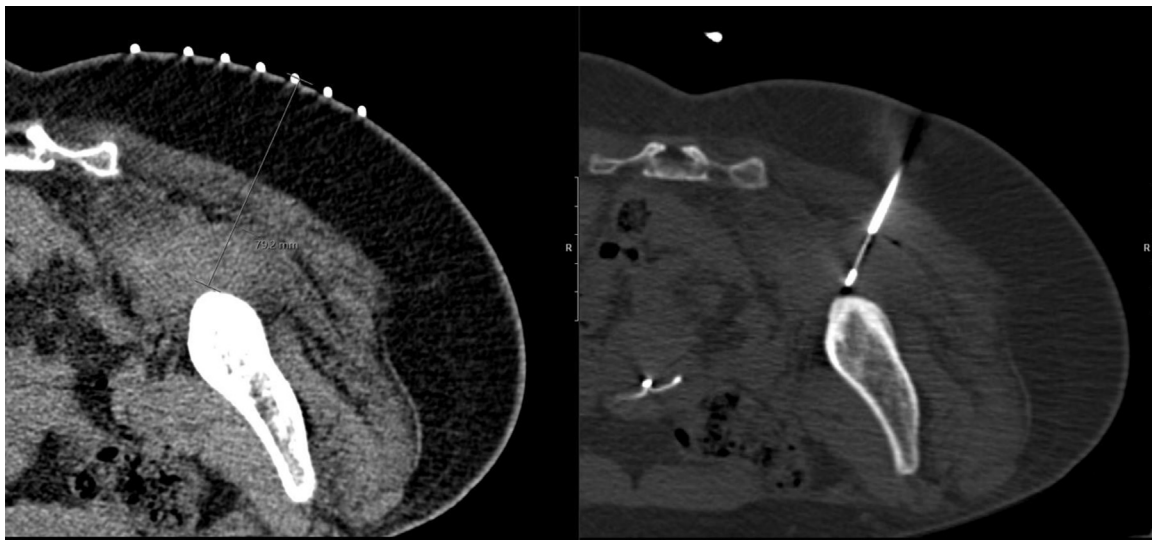


Fig. 3 – CT guided biopsy of the suspected mass located within the right sciatic notch. The biopsy results showed the presence of endometrial cells, stroma, and hemosiderin, confirming the diagnosis of endometriosis.

hancing mass-like density involving the musculature stated above, with faint hypodense foci thought to represent mineralization vs. hemorrhagic products (Fig. 2). Electromyography was also performed, which demonstrated right L5-S1 plexopathy.

CT guided biopsy of the mass within the sciatic notch was performed with pathology demonstrating endometrial cells, stroma, and hemosiderin consistent with endometriosis (Fig. 3). Further examination with pelvic ultrasound did not demonstrate any other sites suspicious for endometriosis. Surgical resection of the lesion was initially considered, however forgone due to risk of sciatic nerve root injury. The patient was medically managed with leuprolide injections for

6 months, with complete resolution of pain and marked improvement of right thigh and foot weakness. At this time, surgical resection was reconsidered in lieu of management with oral contraceptives and follow up MRI was obtained for possible surgical planning 6 months after initiating medical therapy. This showed decreased size of the sciatic notch lesion and decreased mass effect on the sciatic nerve, as well as greatly decreased infiltration and edema of the surrounding musculature (Fig. 4). Muscular atrophy was unchanged compared to the initial MRI. Given these findings and the patient's improving clinical status, the decision was made to continue medical therapy with oral contraceptives. The patient continues to do well clinically at 12 months follow-up.

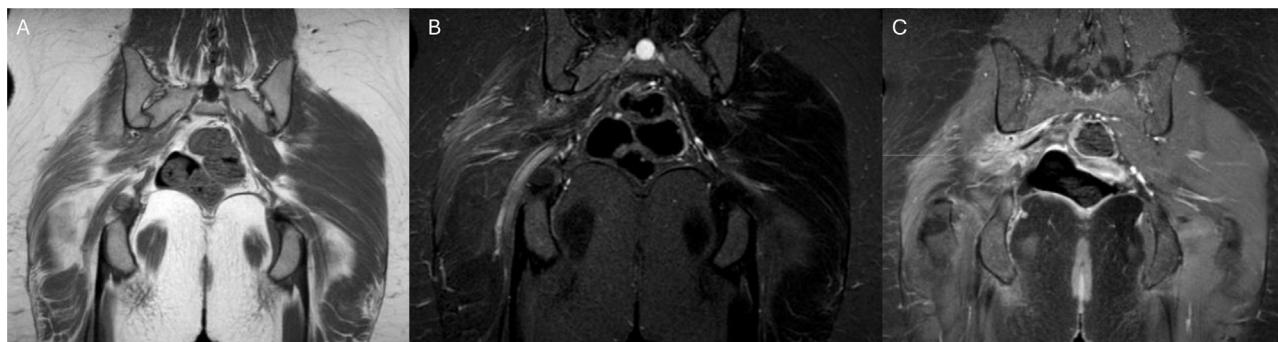


Fig 4 – Coronal T1, STIR, and T1-FS contrast enhanced (A, B, C) MRIs post leuprolide treatment. Images demonstrate decreased size of lesion, decreased mass effect on sciatic nerve, and decreased muscular edema post-treatment.

Discussion

Endometriosis is the proliferation of endometrial tissue outside of the uterus. The exact prevalence of endometriosis is unclear as the definitive diagnosis is made laparoscopically. It is estimated that up to 10% of women of reproductive age, and up to 21% of women with chronic pelvic pain, have endometriosis [1,3,4]. Endometriosis is often asymptomatic, and diagnosis is made at autopsy. When symptomatic, endometriosis classically presents with infertility, dyspareunia, dysuria, and chronic pelvic pain[5]. The strongest associated risk factors for endometriosis include a positive family history, early age at menarche, low BMI, nulliparity, and short menstrual cycles [1,5]. The pathogenesis of endometriosis is not clearly elucidated at this time, with 4 principal hypotheses reported most prominently in the literature. These include a) retrograde menstruation, where the reflux of menstrual debris into the peritoneal cavity via the fallopian tubes, results in ectopic implantation b) metaplasia of coelomic epithelium to endometrial cells, c) vascular or d) lymphatic metastasis of endometrial cells [1,6].

The most common sites of endometriosis are adjacent pelvic structures to the uterus, such as the ovaries, surrounding pelvic ligaments, and the pelvic peritoneum. Outside of the pelvis, the ventral abdominal wall musculature is the most common site of endometriosis and has been associated with open pelvic surgery such as cesarean section or hysterectomy [4]. Less commonly, extrapelvic endometriosis has been identified within the visceral abdomen, thoracic cavity, peripheral nervous system, central nervous system, and within the skeletal muscle [3–10].

Intramuscular and neural endometriosis is a rare and challenging diagnosis which is highly reliant on initial clinical suspicion. The most telling clinical sign is the presence of cyclic and localized musculoskeletal pain associated with the proliferative phase of the menstrual cycle. This presented an additional diagnostic challenge in this case, as the patient had cyclic pain symptoms but also had irregular menstrual cycles due to having an intrauterine device in place. The MRI appearance of endometriosis is variable and depends on the degree of associated hemorrhage. Classically, endometriomas demonstrate hyperintense signal T1-weighted images as well as fat-

saturated images [8,11]. Appearance on T2-weighted images is more variable and largely dependent on the age of the hemorrhagic products deposited within the mass. The lesion in this case had a more nonspecific appearance, with hypointense signal on T1-weighted and only very mildly hyperintense signal on fat saturated images with internal foci of hemosiderin. This led to the differential of post traumatic changes within the sciatic notch vs. desmoid tumor. As was seen in this case, the diagnosis of intramuscular and neural endometriosis is most commonly made at biopsy.

In their systematic review of extrapelvic endometriosis between 1999 and 2019, Andres et al. identified 164 manuscripts describing extrapelvic endometriosis diagnoses, with only 12 case reports describing endometriosis specifically within the musculoskeletal system [3]. In their systematic review of neural involvement of endometriosis, De Sousa et al. identified 140 cases where-in there was extrinsic compression or direct infiltration of the sciatic nerve [6]. The majority of the cases from the above reviews were surgically resected. For example, Andres et al reported that 11 of the 12 cases of intramuscular endometriosis were surgically resected with or without adjuvant suppression with gonadotropin releasing hormone agonist, and 1 case was exclusively treated medically with clinical follow up only. Lomoro et al. also report that 71% of cases in their systematic review of sciatic nerve endometriosis underwent surgical resection at first line treatment [2]. Furthermore, review of the cases which underwent medical therapy reveals a lack of follow up MRI.

Herein presented is a case of extrapelvic endometriosis of the right sciatic notch causing cyclic right hip pain and lumbosacral plexopathy which underwent medical therapy and follow up post-treatment MRI. Review of the available literature demonstrates a paucity of follow-up imaging in patients with extrapelvic endometriosis who are not treated with surgical resection. This case provides an example of the natural history of sciatic notch endometriosis after treatment with gonadotropin release hormone agonists. In this case, 6 month follow up MRI demonstrated decreased size of the lesion within the sciatic notch, decreased mass effect on the sciatic nerve, and improvement of surrounding muscular edema. The patient's hip pain was entirely resolved at the 12-month clinical follow-up with improvement of their right lower extremity weakness and paresthesia. Imaging of extrapelvic en-

dometriosis is challenging due to its rarity and variable appearance. In this particular case, there was a unique challenge as this patient had irregular menstrual cycles given placement of an intrauterine device.

Patient consent

Written consent was obtained from the patient presented in this case and the patient agreed to participate in the case report. The patient understands that this case may be used for educational purposes, training, and medical research. Additionally, the understand that the case may appear in future publications of the journal. The patient was fully informed of the risks and benefits of participating and understands that while their name is removed from the case report, it may be possible that they could be recognized based on the details of the case. The patient understands that their case may be published in the journal.

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