

Peroneal Nerve Endometriosis: A Case Report

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Abstract

Introduction: Infiltrating endometriosis in the extra pelvic part of the sciatic nerve is extremely rare. This report details a case of separate infiltration of the peroneal nerve in high division of the sciatic nerve without any evidence of intrapelvic disease.

Case presentation: A 23-year-old woman presented with a three year history of debilitating right-sided cyclical pain in her right buttock radiating into the leg, and eventually an inability to walk due to weakness of right ankle dorsiflexion. She was otherwise asymptomatic and pelvic examination was normal. A magnetic resonance scan (MRI) demonstrated endometriosis in the peroneal division of the extra-pelvic sciatic nerve. This affected segment was excised using a trans-gluteal approach and the nerve was reconstructed with sural nerve grafts. Pain symptoms immediately improved and she was still free of pain 3 years after the surgery unfortunately there was no recovery of dorsiflexion function.

Discussion: Endometriosis of the extra-pelvic sciatic nerve is very rare. As our case shows early diagnosis is extremely important because delayed diagnosis may lead to severe pain and even nerve damage. Excision of the affected peroneal nerve segment in this case led to complete pain relief, but unfortunately no recovery of function.

Conclusion: Extra-pelvic infiltration of the sciatic nerve by endometriosis is extremely rare although traction on the sacral plexus by severe rectal disease is relatively common. A cyclical pattern of sciatic pain in a menstruating woman should give rise to the suspicion of underlying endometriotic neural involvement. Pain relief is good but may be incomplete and long-term neurological sequelae common.

Keywords: Endometriosis; Sciatic nerve; Sacral plexus; Case report

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Introduction

Endometriosis commonly presents with pain related to intra-abdominal organ involvement: common symptoms include dysmenorrhea, chronic pelvic pain, right iliac fossa pain, dysphasia, dysuria and dyspareunia; back pain, pain on sitting, shoulder pain and on breathing have also been well described and are dependent upon the site of disease [1].

Endometriosis involving the sacral plexus is uncommon and is usually seen in association with extension of severe rectal disease producing traction on the sacral hypogastric fascia. More rarely endometriosis may infiltrate about the sacral ventral nerve roots and invade them sometimes even forming small 'chocolate cysts' within the nerves. It is very unusual to find an isolated lesion

involving the sciatic nerve and even more unusual for such disease to be outside of the pelvis [2]. Sacral nerve disease will typically lead to sciatic pain (S2), pain in the buttock (S3) and perianal pain (S4). Otherwise involvement may result in pudendal pain (S2, S3, S4). It is more unusual to develop gluteal atrophy due to involvement of the superior gluteal nerve (L4, S5, S1) supplying the gluteus medius and minimus and the inferior gluteal nerve (L5, S1, S2) supplying gluteus maximus as these branches arise more proximally and laterally than the usual site of disease. There are many theories regarding the origin and pathogenesis of such endometriosis but most fail to achieve an adequate explanation. Many theories exist regarding the potential pathogenesis of endometriosis, but few can explain the existence of extra pelvic sciatic disease.

Diagnosis may be difficult, but magnetic resonance imaging appears to be most useful [3]. Treatment of endometriosis may be by surgical excision of the disease or medically by means of hormonal suppression using GnRH analogues or progesterone.

Case Presentation

A 23-year-old woman presented to a tertiary endometriosis clinic with a three-year history of pain in the buttock region radiating into the lateral side of her right upper and lower leg. Pain symptoms initially worsened during her period, but at the moment of presentation she had continuous pain symptoms (scored 7 on scale of 0-10) despite pain medication consisting of fentanyl patches, pregabalin 300 mg BD and oxycodone. The patient also had increasingly difficult walking, because of dorsiflexion weakness, pelvic muscle instability and pain. She had completed a three-month course of the GnRH agonist leuproreline, which had rendered her amenorrhoeic but which had not helped with the pain; at presentation she had used Orgametril for about four months and remained amenorrhoeic.

The patient had minimal dysmenorrhoea and lower back pain associated with her periods but no dyspareunia, dyschezia or dysuria. Pelvic examination was normal with no evidence of any tenderness. At neurologic examination she had weakness of dorsiflexion (MRC 4) and slight weakness of gluteal muscles (Trendelenburg).

A magnetic resonance scan was obtained which demonstrated the presence of a likely endometriosis nodule inside the peroneal division of the sciatic nerve, just outside the greater sciatic foramen. There was no evidence of any pelvic disease (**Figure 1**).

We pre-operatively discussed different options with the patient, including the possibility that it would be necessary to transect the peroneal division of the sciatic nerve to be able to remove the endometriosis. She was aware of the possibility of completely losing dorsiflexion function of her right foot postoperatively. Despite this potential consequence she decided to opt for surgery, because the severity of her pain symptoms. She was operated under general anesthesia in prone position using the transgluteal modified Stookey approach (**Figure 2**).

The ischial tuberosity was exposed and the sciatic nerve was identified that at this level had already split into separate divisions of the tibial and common peroneal nerve (**Figure 2**) verified by separate stimulation using intra-operative monitoring. The peroneal division of the sciatic nerve had a clearly tapered aspect for 2-3 cm due to an infiltrating nodule of endometriosis, while the tibial nerve had a normal appearance. Initially the epineurium of the peroneal nerve was opened, but since there was no internal fluid collection that could be drained and the nerve was largely fibrosed it was decided to resect the affected part of the nerve. The defect was subsequently reconstructed with multiple grafts obtained from the sural nerve that was harvested from the right lower leg. The post-operative course was uncomplicated. Pain symptoms had immediately improved, which lasted until the last follow-up in the outpatient clinic three years after the surgery showed that the patient's pain had improved to a very large extent

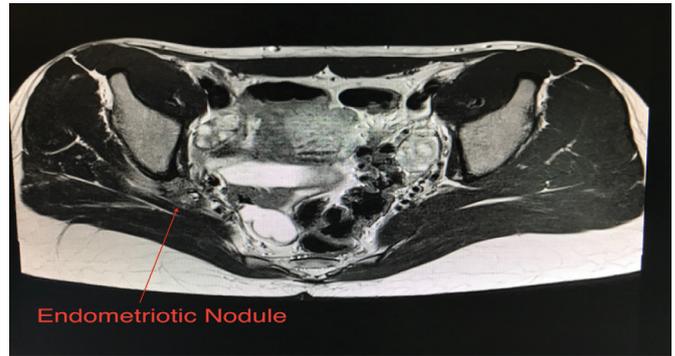


Figure 1 Magnetic resonance scan of pelvic bones.

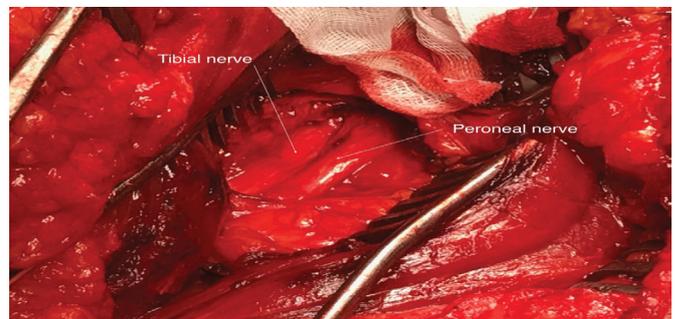


Figure 2 Transgluteal modified stookey approach.

and that she was able to function normally. Unfortunately, she had no recovery of dorsiflexion function and there were no signs of reinnervation of the tibialis anterior muscle on electromyography performed 18 months after the reconstruction.

Discussion

Severe rectal endometriosis not uncommonly causes traction on the sacral hypogastric fascia resulting in involvement of the underlying nerves. It rarely occurs that the disease is found posterior to the sacral hypogastric fascia and even less frequently it is found that the disease infiltrates the nerves. The existence of extra pelvic peroneal nerve endometriosis described in this case appears to be the rarest form and diagnosis is made all the more difficult by the lack of any gynaecological symptoms which can point to the aetiology. This led to a substantial delay in referral and severe symptoms at presentation that not only occurred during menstruation, but were continuously present. As showed by this case this delay resulted in severe intraneural fibrosis that probably was the result of multiple episodes of hemorrhage in the past during the menstrual cycles, causing considerable inflammatory reactions and fibrosis during the healing process, eventually resulting in permanent nerve damage. In our opinion therefore it is important that a careful neurological assessment is carried out in patients with episodes of sciatica related to the menstrual cycle. In addition it is important to establish the level of the lesion and to see what nerves are involved; in the

case described above the lesion appeared to involve the fibres originating from the ventral roots of S2 and S3 between the origin of the pudendal nerve and the superior and inferior gluteal nerves due to the lack of pudendal neuropathy and wasting of the gluteal muscles. Difficulty in this case thereby was that there was a separate involvement of the peroneal nerve due to a high split of the sciatic nerve into the tibial and peroneal nerve division. This anatomical variation is found in about 20% in cadavers.

Diagnosis is difficult unless there is intrapelvic disease in which case a fixed pararectal nodule may be palpated on the curve of the sacrum. In this case reliance had to be placed on MRI which is of proven value due to the haemorrhage nature of the lesions [3] in the determination of pelvic endometriosis and examination confirmed the present of an extra-pelvic endometriotic nodule (Figure 1).

It has been shown that nerve pain may temporarily worsen after neurolysis and so the use of amitriptyline for three months following neurolysis is worth consideration. Some authors recommend the use of GnRH agonists or other hormones [4,5] to suppress endometriosis but the evidence to support this is poor and there is evidence as in this case that such therapy does not provide any relief and may even leave the patient open to the long term deterioration of symptoms. Disease within or around the nerves of the sacral plexus and sciatic nerve may well be progressive [6,7] and delay in treatment may result in permanent damage making it necessary to treat such disease promptly and effectively [8].

It is difficult to explain the pathogenesis of isolated extra pelvic sciatic endometriosis. Retrograde menstruation, lymphatic spread, coelomic metaplasia and transfer along pelvic gutters can in no way provide an explanation. Haematogenous spread has been demonstrated [9] but in this case there would appear to be no intrapelvic primary lesion from which spread might have occurred. Perineural spread as suggested by Siquara de Sousa et al. [10] may explain the passage of cells from intra to extra-pelvic. Endometriosis has been found in the fetus [11] and the newborn [12] and there is strong evidence that deep endometriosis may well be a congenital disease resulting from abnormalities of the Homeobox genes 9, 10, 11 and 13 responsible for the development of the genital and lower alimentary tracts [13]. It is certainly possible that endometriosis may have developed in this patient between 7 and 11 weeks of fetal life and that perineural migration occurred during growth in childhood and adolescence. This is also consistent with the findings of Kinugasa [14] which show that the sacral hypogastric fascia only develops after 12 weeks gestation and thus isolated disease of the nerves may occur external to the fascial layer if laid down before that time.

When endometriosis envelops the nerves of the pelvis or infiltrates them, it is likely that excision of the disease will result in significant injury including hypoaesthesia and muscle paralysis depending on the nerves involved and level of transection; With laparoscopic dissection of the sacral plexus/sciatic nerve care must be taken to avoid injury to the rectal parasympathetic supply arising from the ventral nerve roots of S2-S4. Patients must be made aware of such risks pre-operatively. In our case we expected preoperatively that transection might be necessary due to the long duration and continuous presence of symptoms. It is rare however that the nerve is fibrosed in endometriosis. In a recent systematic review on extrapelvic sciatic nerve endometriosis [15] involvement inside the sciatic nerve was found in 20% of the cases (8 out of 40). Surgery in most of these cases consisted of external or internal neurolysis with drainage of a chocolate-like cyst [16], probably caused by bleeding inside the nerve during the menstrual cyclus. In our case the peroneal nerve at the effected side had completely fibrosed, which led us to decide for resection instead of neurolysis. Although this resulted in dorsiflexion paralysis,

It might have been possible to have effected laparoscopic excision of this nodule by following the intra-pelvic sciatic nerve through its exit from the pelvic in the greater sciatic foramen although the provision of adequate exposure would have been uncertain. What the trans-gluteal approach avoided was the significant risk of injury to the gluteal veins which at laparoscopy can result in considerable blood loss, difficult to control.

Conclusion

This case demonstrates that severe endometriosis may present in very young women as a cause of catamenial sciatic pain without the presence of any specific gynaecological symptoms or evidence of intrapelvic endometriosis. A high level of suspicion for endometriosis should arise when a woman presents with catamenial sciatica and diagnosis is likely to be best established by means of MRI. Surgery should be undertaken as soon as possible as medical treatment does not provide relief and may permit deterioration of the disease and a worsening of the long-term outcome. Surgical treatment is difficult and should only be undertaken by those with a thorough knowledge of pelvic and gluteal neuroanatomy. In most cases surgery will require to be undertaken by a team as severe endometriosis normally shows multi-organ involvement. Clearly, patients must be counselled beforehand regarding potential complications such as muscle paresis or hypoaesthesia. Consideration should be given to the use of amitriptyline post-operatively for three months as nerve-related pain may worsen after surgery.

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