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DEEP GLUTEAL PAIN SECONDARY TO PELVIC WALL ENDOMETRIOSIS- A CASE REPORT

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Background: Chronic pelvic pain is a common condition that affects up to 26% of patients according to a recent systematic review. The clinical history and presentation are widely variable. Diagnosis is difficult and often delayed.

Case Report: A 44-year-old woman presented to the pain clinic with a 5-year history of unexplained deep left buttock pain. The pain symptoms initially suggested entrapment of the left pudendal nerve. A subsequent magnetic resonance imaging showed an abnormal mass in the left ischiorectal fossa. Further biopsy confirmed endometriosis of the lateral pelvic wall, a rarely reported finding. Her pain symptoms considerably improved 70% after initiating medical therapy.

Conclusions: The diagnosis of chronic pelvic pain can be difficult and often delayed. The presence of unexplained pain symptoms, with a cyclical pattern in a child-bearing age, should raise the suspicion toward endometriosis.

Key words: Gluteal pain, chronic pelvic pain, endometriosis, central sensitization, pelvic, pudendal, case report

BACKGROUND

Chronic pelvic pain (CPP) is commonly defined as pain symptoms perceived to originate from pelvic organs/structures typically lasting > 6 months (1). An interplay between a number of conditions, as well as psychological and social factors, contribute to the clinical presentation of CPP. A recent updated systematic review (2) estimated that the prevalence of noncyclical CPP is 5.7% to 26.6%. It is associated with considerable utilization of health care resources both directly and indirectly. Patients commonly go through a long series of rehabilitative, diagnostic, and surgical procedures. It is also associated with loss of working time, reduced productivity, and quality of life.

Endometriosis affects 10% of women globally with 71% to 87% of cases reported to have CPP (3). Its pathophysiology remains uncertain. Treatment options are surgical or hormonal, but a recurrence rate as high as 43.5% has been reported. Endometriosis affects pelvic structures, such as ovaries and ligaments and less commonly extrapelvic structures (4). The presentation of endometriosis depends on the site(s) where the endometrial cells were implanted and is often challenging and involves input from multiple specialties.

CASE PRESENTATION

A 44-year-old woman, otherwise healthy presented to the pain clinic with a 5-year history of deep pain in the left buttock area. She mentioned that the pain may have started after getting off a low stool from prolonged sitting. The pain radiated to the vagina, burning in quality, caused difficulty during intercourse and during defecation. Her most disruptive symptom was the sharp left buttock pain and a sensation of a deep mass roughly the size of a small fruit on the inner aspect of the left ischial bone. No motor weakness or

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radicular symptoms were noted. The pain was constant but tended to become more severe during her periods. She had a gynecological procedure earlier. More recent ultrasound of the abdomen did not show a cause of her pain. She had different forms of physiotherapy and rehabilitation with no benefit. She was triaged to musculoskeletal services for evaluation. Initial magnetic resonance imaging (MRI) of the lumbar and sacral area did not show mechanical or neuropathic cause for her symptoms. She had an ultrasound-guided injection in the left ischial bursa with no benefit. She was then seen in the pain management clinic. Her history and symptoms were suggesting pudendal nerve (PN) neuralgia. An MRI of the left ischiorectal fossa and PN was performed. It showed an abnormal mass in the left ischiorectal fossa, which flatten the anal fat and involve the left ischiococcygeus and obturator internus muscles. Colorectal surgical oncology review advised that the mass is not infiltrating the rectal area, but rather involves the left pelvic wall with possible left PN entrapment. She was referred to the Birmingham Sarcoma Service. A further MRI of the pelvis showed an ill-defined 61x22 mm, contrast-enhancing soft tissue abnormality on the left iliococcygeus and obturator internus muscles, which appear to have slightly increased in size since the last scan. No anal fistulas, but some nodularity of the mesorectum. The PN appeared to be intact. A biopsy of the mass revealed endometrial tissue.

A gynecological review of the results recommended starting treatment with tibolone (combined estrogen and progesterone action and protection against osteoporosis), a gonadotropin-releasing hormone analog, and a bone densitometry in 18 months. The long-term plan will be a consideration for surgical excision of the mass. The patient was reviewed in the pain clinic 3 months after initiating her treatment. She reported an ongoing 70% improvement in her symptoms. Informed consent was obtained to present her case.

DISCUSSION

The patient was unable to function well and ultimately had to stop a highly professional job because of the pain. It took > 5 years to diagnose endometriosis as a cause of her nonresolving CPP. This patient had pain symptoms suggesting musculoskeletal pathology within the pelvis or PN. Previous negative gynecological review and abdominal ultrasound were a distraction from considering endometriosis as a potential cause of her symptoms. On the other hand, the presence of alarming signs of a pelvic space-occupying lesion on the second pelvic MRI acted to her benefit and accelerated the diagnosis.

CPP patients commonly endure a long journey of pain and suffering. By the time of diagnosis, they would have seen multiple specialties, undergone repeated tests and imaging studies, and often had one or more unsuccessful surgical procedures.

This long time till diagnosis agrees with evidence from the literature. Agarwal et al (5) quoted a well-established delay in getting a confirmed surgical diagnosis of endometriosis of 4-11 years. The average time to diagnosis using either surgical or nonsurgical means is 4.4 years with women aged 40-49 years having a shorter time till diagnosis compared with under 18-year-old patients (6). Ongoing undiagnosed pain from endometriosis leads to dysregulation between the peripheral and central nervous systems with subsequent central sensitization and pain. The central pain then becomes self-perpetuating and less dependent from peripheral pain.

Higher levels of sensitization can lead to poorer responses to chronic pain management interventions and lower quality of life. These patients are more prone to develop other chronic pain conditions, depression, anxiety (7) and at a higher risk of developing certain types of cancer, autoimmune disease, asthma, and cardiovascular disease (8).

Central sensitization is also a strong predictor to persisting pelvic pain after endometriosis-related hysterectomies (9). Similar findings were reported by Tucker et al (10). They concluded that certain pelvic pain comorbidities, e.g., depression, catastrophizing lead to lower the Endometriosis Health Profile after endometriosis surgery (10). The use of scoring systems, such as the Central Sensitization Inventory scores, has been proposed to help gynecologists to identify and counsel higher-risk patients preoperatively (11). Fortunately, our patient did not have symptoms suggestive of central sensitization or any of the comorbidities described earlier. She is more likely to have a better outcome if surgery is offered in the future.

Extrapelvic endometriosis affecting skeletal muscles is not a common presentation for endometriosis with a reported incidence of 12%. In a recent systematic review (12), the commonest skeletal muscular site for extrapelvic endometriosis was the abdominal wall (50.7%). Pelvic floor endometriosis accounted for 11.6% of cases, but it can affect virtually any muscle or joint in the body (12).

Endometriosis affecting the lateral pelvic wall is rarely reported in the literature (12,13).

CONCLUSIONS

The clinical presentation in patients with CPP is widely variable and often difficult to diagnose. Patients are typically reviewed by multiple health care professionals for long periods before ending up in the pain clinic. The presence of unexplained pain symptoms, with some form of a cyclical pattern in a child-bearing age, should raise the suspicion toward endometriosis. Identification and management of preoperative psychological comorbidities improve the overall outcomes in patients with CPP.

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