



Pain in the enthesis of levator ani muscle: A novel source of chronic pelvic pain

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Abstract

Background: Managing chronic pelvic pain (CPP) remains a challenge due to its diverse range of causes. A newly identified anatomical entity known as the enthesis of the levator ani muscle (LAM) and its associated disorders might play a role. This paper describes a novel insight into CPP's origin, aiming to improve accurate diagnosis and treatment.

Methods: Data were collected from medical records (paper or electronic) retrospectively. The study included 112 patients meeting the criteria, divided into CPP and non-CPP groups. Clinical symptoms, including location of LAM enthesis, referred pain from pain in LAM enthesis, and related lower urinary tract symptoms (LUTSs) were discussed. To identify differences in symptoms between the groups, a Chi-squared test and descriptive analyses were conducted.

Results: Bimanual examination revealed tender sites in the attachment of the LAM to the pubic bone. LAM enthesis pain presumably caused referred pain in at least 10 areas, primarily in the lower abdominal quadrante (40.2%-47.3%) followed by the inguinal area (8.9%-15.1%). Multiple LUTSs were observed, including urinary frequency (72.3%), urgency (42.9%), nocturia (53.6%), residual urine sensation (64.3%), urinary incontinence (30.3%), painful bladder (34.8%), and weak urine stream (47.9%). Patients in the CPP groups experienced significant residual urine sensation (53.6%) and bearing-down sensation (42%) compared to the non-CPP group.

Conclusion: Pain in LAM enthesis is a novel cause of pelvic pain and LUTSs that warrants attention for the evaluation and management of CPP.

Keywords: Enthesopathy; Nocturia; Pelvic pain; Pubic bone; Referred pain

1. INTRODUCTION

Chronic pelvic pain (CPP) is defined as continuous or recurrent episodic pain lasting at least 6 months, which is unrelated to the menstrual cycle, infection, or other identifiable pelvic pathologies.^{1,2} It affects up to 24% of women globally and substantially impacts their quality of life.³ CPP is a complex condition with multiple potential causes, including both physical and psychosocial causes.³⁻⁵ Among the causes, myofascial pelvic pain

syndrome (MPPS) is a common contributor to CPP, affecting 14% to 22% of women with CPP.⁶

MPPS is characterized by tight and tender pelvic floor muscles, which may include palpable muscle bands or trigger points.⁷ Enthesopathy refers to pain resulting from a disorder involving an enthesis.⁸ Enthesis is the attachment site of a tendon or ligament to a bone,⁹ which in this case extra-articular entheses, which often span a large portion of the diaphysis, comprise fibrous attachments where a tendon is attached either to the periosteum or directly to the bone.¹⁰

However, we proposed that not only does MPPS as a cause of CPP, but also pain defined as enthesopathy which serves as a major source of musculoskeletal pain, can be involved in causing pelvic pain. These two conditions have distinct pain locations, pathological mechanisms, and treatment approaches. The pain from enthesis of levator ani muscle (LAM) was located in the attachment of the aponeurosis of the LAM, extending beyond the ATLA to the inferior border of the superior ramus of the pubic bone.¹¹ Conversely, pain associated with MPPS is characterized by tense and sensitive pelvic floor muscles.⁷ In general, treatment for enthesopathy depends on the underlying etiology such as muscle overuse, metabolic conditions, or autoimmune diseases. It might require the injection of steroids to the enthesis

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pain site to reduce the inflammatory process. Moreover, when an autoimmune disease is identified as the underlying cause, the use of a disease-modifying antirheumatic drug (DMARD) can effectively address immune responses. Additional medications, including methotrexate and sulfasalazine (Azulfidine), may also provide relief from symptoms.¹² In contrast, the treatment for MPPS involves inserting a needle into the trigger point or tense muscle band. Unlike the use of medications like steroids or DMARDs, MPPS typically results in a less painful or shorter duration of the disease.¹³

The arcus tendinous levator ani (ATLA) is conventionally regarded as the origin of the LAM. However, according to our previous study, the attachment of the aponeurosis of the LAM extends beyond the ATLA to the inferior border of the superior ramus of the pubic bone.¹¹ Kim et al¹⁴ proposed a connection between the aponeurosis of pubovisceral muscle (a variable part of the LAM) and pubic bone. Because aponeuroses are sheet-like elastic fibrous structures that partially cover muscles and anchor them to the bone, the attachment of the aponeurosis of the LAM to the pubic bone qualifies as an enthesism in medical terminology.¹⁵ Consequently, the possibility of pain related to a disorder in the enthesism of the LAM cannot be overlooked.

Both the recently discovered attachment of the LAM to the superior ramus of the pubic bone (enthesism of the LAM) and the consequent pain originating from this site may provide a reference for the clinical diagnosis and treatment of CPP. In this article, we present novel insights into the etiology of CPP, with the ultimate goal of facilitating its accurate diagnosis and treatment. To the best of our knowledge, this is the first article to describe the clinical manifestations of pain arising from the attachment of the LAM to the superior ramus of the pubic bone (enthesism of the LAM).

2. METHODS

2.1. Study design and population

The data collection process was approved by the Taipei Medical University-Joint Institutional Review Board and the Ethics Committee of Taipei Medical University Hospital in Taiwan (approval no. N202202006). Data were retrospectively collected from either paper or electronic medical records from January 2020 to December 2021. A total of 112 patients who met the inclusion criteria were included in the study. All methods and procedures were conducted in accordance with the relevant guidelines and regulations.

2.2. Inclusion and exclusion criteria

All patients included in this study presented with pain localized to the pelvic or lower abdominal area, which was their primary complaint upon presenting to our outpatient clinic. Eligible patients were those who had painful sites originating from the enthesism of the LAM, which were confirmed by a bimanual pelvic examination.

Patients with the following pathological conditions were excluded from the study: evidence of urinary tract infection, urogenital tract malignancy, pelvic mass or malignancy, pelvic organ prolapse, active urinary tract stones, intravesical lesions, and enlarged uterine size that is, more than 8 cm in length, 5 cm in width, and 4 cm in thickness of dimensions of the uterus or there was more than 5 cm in size in the anterior wall of the uterus.^{16,17} Patients who had a history of medications known to affect voiding dysfunction and those who had prior urogenital and anal surgery, diabetes mellitus, or neurogenic lower urinary tract dysfunction were also excluded. In addition, patients aged <20 years were excluded from this study.

2.3. Bimanual examination

According to our previous study, the origin of the LAM (specifically the iliococcygeus part) is a type of aponeurosis that connects with the pubic bone. This aponeurosis involves attachment to the inferior border of the superior ramus of the pubic bone, posterior attachment to the anterior border of the greater sciatic notch, and inferior attachment to the ischial spine and arcus tendinous fasciae pelvis. In this study, we evaluated painful sites originating from the enthesism of the LAM through digital palpation during regular vaginal examinations. To ensure the precise analysis of LAM enthesism pain, we divided these sites into three locations, corresponding to an area measuring approximately 2 to 3 cm rostral to the ischial spine before the great sciatic notch (location A), the cranial part of the inferior border of the superior ramus of the pubic bone (location B), and the caudal part of the inferior border of the superior ramus of the pubic bone (location C; Fig. 1). We also identified various locations of referred pain that developed during the palpation of the enthesism of the LAM, including the pelvic crest, inguinal area, labia majora, labia minora, vaginal outlet, urethral area, inner thigh, lumbosacral area, anus, and lower quadrant of the abdomen. Through the aforementioned approaches, the common locations of pain associated with these conditions were identified. All of these patients had pelvic examinations by the same physician which applied a uniform pressure while palpating areas in this newly identified anatomy. The severity of pain was not evaluated using pain analog scores nor questionnaires; however, patients reported pain that considerably affected their quality of life.

CPP is defined as intermittent or constant lower abdominal or pelvic pain lasting at least 3 months. Cyclical pelvic pain during a period exceeding 6 months is regarded as CPP.² Acute pelvic pain (APP) is defined as lower abdominal or pelvic pain lasting <3 months.

During their visit, all outpatients in our clinic were routinely instructed to maintain a 3-day urinary log. Using this log, the clinician could identify patients who voided more than eight times per day (frequency), those who woke up to urinate at least twice during the night (nocturia), and those who did not experience fluid overload during the day. Other symptoms, such as bearing-down sensation, painful bladder, morning stiffness, residual urine sensation, and voiding difficulty (weak or interrupted urine stream), were identified through personal physician-patient interactions and were documented in the patients' medical records. Irritable bowel syndrome (IBS), dysmenorrhea, and myofascial pain syndrome were diagnosed on the basis of patients' symptoms, medical history, and physical examination results.

2.4. Statistical analysis

The collected data were exported into IBM SPSS Statistics version 26 (IBM, Armonk, NY) for further analysis. The Chi-squared test was used to determine the differences in all variables between the acute and CPP groups, and a $p \leq 0.05$ was considered statistically significant. All values are reported as means (SDs). Descriptive statistics, including frequencies and percentages, were calculated to examine referred pain originating from each of the painful sites of the enthesism of the LAM.

3. RESULTS

3.1. Demographics and clinical features of patients with pain in enthesism of LAM

All patients in this study experienced LAM enthesism pain. Table 1 presents the demographic and clinical characteristics of these patients. The mean age and body mass index of patients were 50.05 ± 14.36 years and 22.73 ± 3.67 kg/m², respectively.

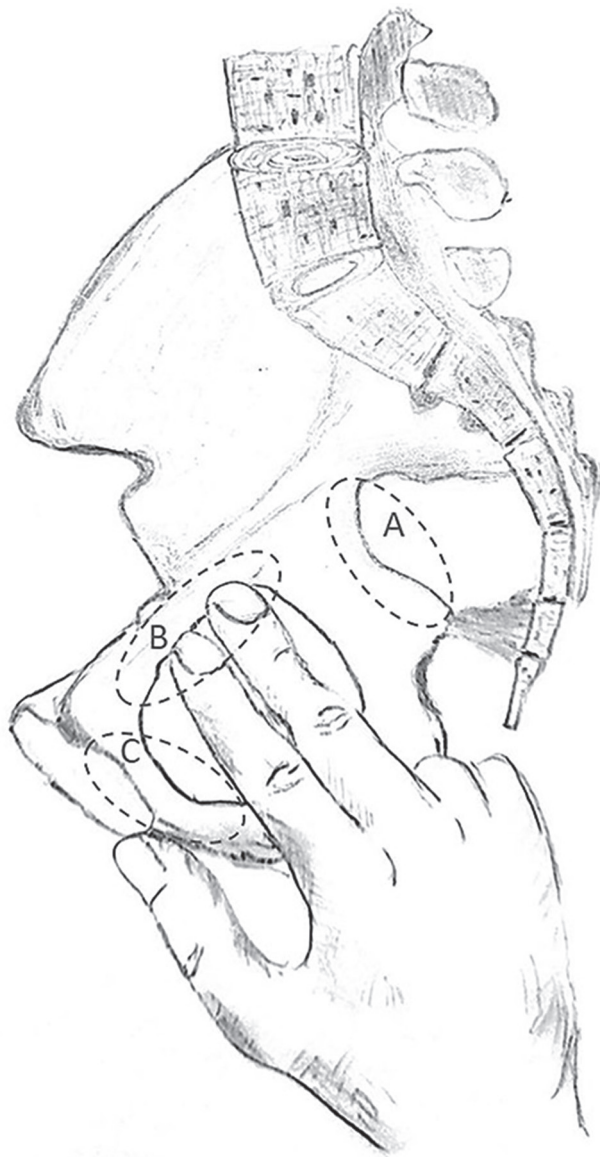


Fig. 1 Enthesis of the LAM in locations A, B, and C. A, Location A of entheses LAM in an area measuring approximately 2 to 3 cm rostral to the ischial spine before the great sciatic notch. B, Location B of entheses LAM, which is the cranial part of the inferior border of the superior ramus of the pubic bone. C, Location C of entheses LAM is in the caudal part of the inferior border of the superior ramus of the pubic bone. LAM = levator ani muscle.

The majority of the patients with LAM entheses pain had given birth, with 66.1% delivering normally and spontaneously, 10.7% delivering through cesarean section, and 4.5% delivering through both methods.

Patients with LAM entheses pain experienced various urinary symptoms, including an increased frequency of urination (72.3%), nocturia (53.6%), urgency (42.9%), and urinary incontinence (31.3%). They also reported additional lower urinary tract symptoms (LUTSs), such as residual urine sensation (64.3%), bearing-down sensation (42.0%), weak urine stream (49.1%), and painful bladder (34.8%). In addition, they reported other pain-related conditions, including IBS (50%), myofascial pain from other body parts (68.8%), and dysmenorrhea (33.9%).

Patients with pain in the entheses of the LAM were divided into two groups: a CPP group, comprising 74 patients (66.07%) with CPP, and a non-CPP group, comprising 48 patients (42.86%) with pelvic pain lasting <3 months. Table 1 presents the demographic characteristics of all patients in the two groups. Compared with the non-CPP group, the CPP group comprised a significantly larger percentage of patients experiencing residual urine sensation (CPP vs non-CPP = 47.3% vs 17.0%, $p < 0.05$) and bearing-down sensation (CPP vs non-CPP = 33.0% vs 8.9%, $p < 0.05$). The CPP group also comprised a significantly larger percentage of patients experiencing dysmenorrhea (CPP vs non-CPP = 26.6% vs 5.4%, $p < 0.05$) and general myofascial pain (CPP vs non-CPP = 50.0% vs 18.8%, $p < 0.05$).

3.2. Physical examination of painful sites in patients with pain in the entheses of the LAM

Table 2 presents the physical examination results of painful sites in patients with pain in the entheses of the LAM. Among all patients, 89 (79.5%) experienced pain originating from location A, whereas 100 (89.3%) and 94 (83.9%) experienced pain originating from locations B and C, respectively. Regardless of the origin of pain, no statistically significant difference was observed between the CPP and non-CPP groups (Table 2).

In the study sample, 75 (66.9%) of patients experienced not only LAM entheses pain but also myofascial pelvic pain. However, no statistically significant difference was observed between the APP and CPP groups ($p = 0.539$).

3.3. Referred pain resulting from LAM entheses pain

In this study, we observed that LAM entheses pain resulted in referred pain in 10 specific areas. Table 3 presents the frequency of referred pain corresponding to each entheses location. Palpation at locations A, B, and C revealed referred pain in 66.96%, 77.68%, and 70.54% of patients, respectively, and <35% of patients had no referred pain.

Regardless of the origin of pain, the area that was most commonly associated with referred pain was the bilateral lower abdominal quadrant. Specifically, 47.3%, 47.3%, and 40.0% of patients with pain originating from locations A, B, and C, respectively, reported referred pain in the bilateral lower abdominal quadrant. Additionally, regardless of the origin of pain, the second most common site for referred pain was the inguinal area. Specifically, 8.93%, 14.3%, and 15.2% of patients with pain originating from locations A, B, and C, respectively, reported referred pain in the inguinal area.

Among patients with pain originating from location A, no referred pain was reported in the urethral area or labia minora. Similarly, among patients with pain originating from location C, no referred pain was reported in the pelvic crest or lumbosacral area. These findings can be explained by considering both the distribution of nerves and the three-dimensional structure of the body surface in relation to the location of pain.

4. DISCUSSION

According to the anatomy of the LAM from our prior study, tender sites are observed along the attachment of the aponeurosis of the LAM to the pubic bone, as demonstrated by bimanual examination. As described earlier in the text, the aponeurosis of the LAM extends beyond the ATLA and connects to the inferior border of the superior ramus of the pubic bone.¹¹ This attachment site is referred to as an entheses. In entheses, the pathophysiology of pain differs from that of myofascial pain, which involves the identification of painful taut bands in related muscles. Similar to myofascial pain, CPP is associated with frequently unrecognized

Table 1**Characteristics of patients with LAM entheses pain in the chronic pelvic pain and non-CPP groups**

Variable	Non-CPP (n = 48)	CPP (n = 74)	Total, %	p
Age, y	50.53 ± 14.32	49.81 ± 14.47	112 (100)	0.804
Height, cm	159.06 ± 5.88	158.39 ± 5.90	112 (100)	0.597
Weight, kg	58.33 ± 10.61	56.26 ± 8.46	112 (100)	0.299
BMI	23.12 ± 4.47	22.52 ± 3.18	114 (100)	0.447
Non-nulliparous, %	24 (21.4)	50 (44.6)	74 (66.1)	0.641
Urinary frequency, %	25 (22.3)	56 (50.0)	81 (72.3)	0.268
Urgency, %	13 (11.6)	35 (31.3)	48 (42.9)	0.185
Nocturia, %	20 (17.9)	40 (35.7)	60 (53.6)	0.886
Urinary incontinence, %	9 (8.0)	26 (23.2)	35 (31.3)	0.216
Weak urine stream, %	16 (14.3)	39 (34.8)	55 (49.1)	0.288
Residual urine sensation, %	19 (17.0)	53 (47.3)	72 (64.3)	0.024
Painful bladder, %	12 (10.7)	27 (24.1)	39 (34.8)	0.606
Bearing-down sensation, %	10 (8.9)	37 (33.0)	47 (42.0)	0.016
Dysmenorrhea, %	6 (5.4)	32 (28.6)	38 (33.9)	0.004
IBS, %	15 (13.4)	41 (36.6)	56 (50.0)	0.110
Myofascial pain from any body part, %	21 (18.8)	56 (50.0)	77 (68.8)	0.027
Morning stiffness from any body part, %	16 (14.3)	44 (39.3)	60 (53.6)	0.081

BMI = body mass index; CPP = chronic pelvic pain; IBS = irritable bowel syndrome; LAM = levator ani muscle.

Table 2**Enthesis in LAM-related painful sites**

Location	Non-CPP (n = 48)	CPP (n = 74)	Total, %	p
Location A, %	29 (25.4)	60 (52.6)	89 (79.5%)	0.554
Location B, %	34 (29.8)	66 (57.9)	100 (89.3%)	0.963
Location C, %	34 (29.8)	60 (52.6)	94 (83.9%)	0.252
Obturator muscle, %	20 (17.5)	50 (43.9)	70 (62.5%)	0.122
Levator ani posterior, %	21 (18.4)	42 (36.8)	63 (56.3%)	0.880
Levator ani anterior, %	16 (14)	36 (31.6)	52 (46.4%)	0.511

CPP = chronic pelvic pain; LAM = levator ani muscle.

Table 3**Referred pain originating from each painful site in LAM entheses**

Enthesis location	Referred pain location, n (%)										
	1	2	3	4	5	6	7	8	9	10	11
A	4 (3.57)	10 (8.93)	3 (2.68)	0	1 (0.89)	0	6 (5.36)	2 (1.69)	1 (0.89)	53 (47.32)	37 (33.04)
B	4 (3.57)	16 (14.29)	5 (4.46)	1 (0.89)	1 (0.89)	2 (1.79)	4 (3.57)	1 (0.85)	2 (1.79)	53 (47.32)	25 (22.32)
C	0	17 (15.18)	8 (7.14)	3 (2.68)	3 (2.67)	5 (4.46)	2 (1.79)	0	3 (2.68)	45 (40.18)	33 (29.46)

1 = pelvic crest; 2 = inguinal area; 3 = labia majora; 4 = urethral area; 5 = vaginal outlet; 6 = labia minora; 7 = inner thigh; 8 = lumbosacral area; 9 = anus; 10 = lower quadrant of the abdomen; 11 = non-referred pain; A = great sciatic notch; B = head of the inferior border of the superior ramus of pubic bone; C = caudal part of the inferior border of the superior ramus of pubic bone; LAM = levator ani muscle.

and untreated symptoms, which are typically diagnosed after multiple visits to different health-care providers.¹⁸ To the best of our knowledge, LAM entheses pain was never discussed in the literature before.

In this study, we identified both LAM entheses pain and myofascial pelvic pain in 66.9% of our patients. According to Bismil et al,¹⁹ entheses serve as specialized attachment sites for myofascial units to the bone. These sites involve stress concentration and are thus susceptible to microtrauma and chronic inflammation. Because muscles are more elastic than tendons and bones, the mechanical stress that transfers from muscles to bones may dissipate in the muscle-tendon-bone unit (entheses), minimizing the effects of stress concentration. However, following exercise or spasms, muscle stiffness may prevent the dissipation of this stress, resulting in microinjury in the entheses.⁸ Abate et al²⁰ and

Killian et al²¹ have indicated that the overuse of the entheses and loading-induced microdamage may result in enthesopathy. Horita et al²² also reported that in certain conditions, such as overexertion, the rigidity of muscles and tendons is altered, resulting in muscle dysfunction during the early stages and in enthesopathy during the late stages. In our previous study, we discovered that the thickness of the fused obturator fascia with the aponeurosis of the LAM was four times thicker in women than in men.¹¹ Other studies have indicated that the volume of the LAM is approximately twice larger in women than in men.^{23,24} Thus, we assume that the overuse of the LAM and the microtrauma resulting from weight bearing may occur more easily in women than in men. Although myofascial pain may be associated with entheses pain during the course of the disease, it manifest at different stages.

Notably, in this study, 33.1% of patients experienced pain, specifically in the enthesis of the LAM, without any myofascial pain in the muscular region of the LAM. This finding suggests that the enthesis of the LAM serves as the primary source of pain. Schilders et al^{25,26} reported that enthesopathy plays a role in musculofascial unit dysfunction and pain. They also described that enthesopathy in the enthesis of a specific muscle causes pain upon passive stretching of the affected muscle and eventually causing resisted muscle activation. Pain caused by enthesopathy or inflammation in the enthesis of the LAM, referred to as enthesitis, may be the underlying cause for the later development of myofascial pain. Thus, LAM enthesis pain must be identified as early as possible. Thus, our previous research highlighting the LAM (especially the iliococcygeus part) and its connection to the pelvic bone provides crucial anatomical findings.

In this study, patients with LAM enthesis pain exhibited certain LUTSs. Specifically, 72.3%, 42.9%, 53.6%, 64.3%, 30.3%, and 47.9% of patients experienced urinary frequency, urgency, nocturia, residual urine sensation, urinary incontinence, and a weak urine stream, respectively. Generally, the LAM plays a crucial role in maintaining urethral tension, which directly affects proper urination.²⁷ Meister et al²⁸ demonstrated that myofascial pelvic pain is strongly correlated with difficulties in bladder emptying and increased urinary frequency. The aforementioned hypothesis, which suggests an interaction between myofascial pelvic pain and LAM enthesis pain, may explain why patients experiencing LAM enthesis pain exhibit similar LUTSs to those with MPPS. These findings may contribute to a better understanding of the origins of LUTSs.

The noteworthy discovery of an increase in dysmenorrhea rates within the CPP group as shown in Table 1 suggests a potential link between CPP, enthesis pain in LAM, and dysmenorrhea. This is further supported by the higher percentages of pain in the enthesis of LAM and myofascial pain in the CPP group as indicated in Table 2, although these differences did not reach statistical significance. Li et al²⁹ also underscored 2.5 times higher odds (95% CI, 2.02-3.10) of a positive association between dysmenorrhea and chronic pain, extending beyond the pelvic region.

We found that LAM enthesis pain was accompanied by referred pain in at least 10 areas, namely the pelvic crest, inguinal area, labia majora, urethral area, vaginal outlet, labia minora, inner thigh, back gird, anus, and lower quadrant of the abdomen. According to a previous study,¹¹ locations A and B belong to the iliococcygeus part of the LAM. Hence, they exhibit a pattern of referred pain similar to that observed in the lower abdominal quadrant (47.32% and 47.32%) and the inguinal area (8.93% and 14.29%). Because of its proximity to the cranial area, location A is associated with an increased likelihood of referred pain in the pelvic crest and lumbosacral area. As described by Kim et al,^{14,15} location C involves the enthesis of the pubococcygeal and puborectal parts of the LAM. In this study, we discovered that compared with locations A and B, location C tended to be associated with a higher likelihood of referred pain in specific regions, such as the labia majora (7.14%), labia minora (4.46%), urethra (2.68%), and anus (2.68%). This phenomenon may be caused by the distribution of the variable muscular part of the LAM and the three-dimensional structure of the body surface in relation to the location of painful sites. Pastore and Katzman¹⁸ discovered that similar to LAM enthesis pain, myofascial pelvic pain resulted in referred pain in the suprapubic region, urethra, perineum, sacrococcygeal region, deep vaginal region, posterior thigh, and anus. This finding provides insights into the origins of CPP and its management.

In this study, among patients with LAM enthesis pain, 53.6% experienced morning stiffness. Morning stiffness and enthesis

inflammation, referred to as enthesitis, are hallmarks of autoimmune rheumatic diseases.³⁰ Entheses are regarded as a primary target organ in psoriatic arthritis and ankylosing spondylitis.^{30,31} Further research is required to investigate the potential correlation between LAM enthesis pain and autoimmune diseases.

Overall, we discovered that 50.4% and 33.9% of patients with LAM enthesis pain had a history of IBS and dysmenorrhea, respectively. Because some women may experience suprapubic pain or discomfort in the lower abdominal quadrant, the location of referred pain resulting from LAM enthesis pain should be considered during diagnosis. These symptoms may sometimes be misleading and may be due to gastrointestinal diseases such as IBS.¹⁸ Lillemon et al³² defined viscerosomatic reflexes as a mechanism through which visceral pain originating from the bladder, uterus, and bowel results in myofascial trigger points and muscle pain. Hence, clinical physicians must be aware of the difficulty of identifying the primary source of pain, especially LAM enthesis pain, during the management of CPP.

In this study, we examined pain originating from a novel anatomical structure, namely the enthesis of the LAM. Specifically, we investigated the connection between the iliococcygeus part of the LAM and pelvic bone, which we have discussed in a previous study. We also conducted an extensive review of relevant articles. To the best of our knowledge, this is the first article to report on LAM enthesis pain and its symptoms. Some of our proposed theories are unique and are built upon this understanding.

This study has some limitations. First, the number of patients included in each group was small. Second, because the enthesis of the LAM is regarded as a recent discovery and has not yet been comprehensively investigated, limited references were available. Given the numerous clinical symptoms associated with LAM enthesis pain, further research is required to facilitate detailed comparisons.

Taken together, recent anatomical findings have revealed novel sources of pain in the pelvic area, specifically referred to as LAM enthesis pain. This finding can serve as a valuable reference for evaluating CPP. In this study, we developed a screening strategy for the evaluation of CPP by palpating the enthesis of the LAM to the pubic bone. Clinicians should be aware of this potential source of pain and should identify the precise anatomical locations of tenderness whenever possible.

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