ISSN 2307-8960 (online)

World Journal of *Clinical Cases*

World J Clin Cases 2021 December 26; 9(36): 11122-11508

Published by Baishideng Publishing Group Inc

World Journal of Clinical Cases

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The *WJCC* is now indexed in Science Citation Index Expanded (also known as SciSearch®), Journal Citation Reports/Science Edition, Scopus, PubMed, and PubMed Central. The 2021 Edition of Journal Citation Reports® cites the 2020 impact factor (IF) for *WJCC* as 1.337; IF without journal self cites: 1.301; 5-year IF: 1.742; Journal Citation Indicator: 0.33; Ranking: 119 among 169 journals in medicine, general and internal; and Quartile category: Q3. The *WJCC*'s CiteScore for 2020 is 0.8 and Scopus CiteScore rank 2020: General Medicine is 493/793.

RESPONSIBLE EDITORS FOR THIS ISSUE

NAME OF JOURNAL
World Journal of Clinical Cases

ISSN
ISSN 2307-8960 (online)

LAUNCH DATE
April 16, 2013

FREQUENCY
Thrice Monthly

EDITORS-IN-CHIEF
Bao-Gan Peng

EDITORIAL BOARD MEMBERS
https://www.wjgnet.com/2307-8960/editorialboard.htm

PUBLICATION DATE
December 26, 2021

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https://www.wjgnet.com/bpg/gerinfo/242

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World J Clin Cases 2021 December 26; 9(36): 11406-11418

DOI: 10.12998/wjcc.v9.i36.11406

ISSN 2307-8960 (online)

CASE REPORT

Inguinal endometriosis: Ten case reports and review of literature

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Author contributions: Li SH performed the conception and design; Li SH and Wang SZ performed the administrative support; Li SH and Sun HZ performed the provision of study materials or patients; Sun HZ and Li WH performed the collection and assembly of data; Sun HZ performed the data analysis and interpretation; all authors contribute to manuscript writing and final approval of manuscript.

Informed consent statement: Written informed consent was obtained from the patients for publication of this case report and accompanying images and clinical data.

Conflict-of-interest statement: All authors declare no conflicts of interest related to this article.

CARE Checklist (2016) statement: The authors have read the CARE Checklist (2016), and the manuscript was prepared and revised according to the CARE Checklist (2016).

Supported by 2017 "Sailing" Plan of Beijing, No. ZYLX201713.

Country/Territory of origin: China

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Abstract

BACKGROUND

To describe the characteristics, diagnosis and surgical treatment of inguinal endometriosis (IEM).

CASE SUMMARY

We retrospectively analyzed 10 patients diagnosed with IEM at Beijing Chao-Yang Hospital from 2011 to 2019. Relevant features, symptoms, images, surgical treatment, hormonal therapy and follow-up were collected and discussed. A total of 10 cases of IEM diagnosed by surgery and pathology were characterized by a lesion on the right side (9/11); five patients had symptoms related to the menstrual cycle, and only 3 patients were clearly diagnosed before surgery. Ultrasonography was of little assistance in confirming the diagnosis, but magnetic resonance imaging showed specific, high-intensity patterns. Anatomically, most of the IEM lesions were located in the extraperitoneal ligament (10/11); nine patients had inguinal hernias (IH), five had concurrent or prior pelvic endometriosis, and four had infertility. The clinical results from extensive resection were satisfactory.

CONCLUSION

IEM is an extremely rare condition that can easily be misdiagnosed prior to surgery. A right IH may contribute to the formation of right-sided IEM, and extensive resection involving the round ligament and hernia sac is essential to prevent recurrence.

Key Words: Inguinal endometriosis; Clinical characteristics; Imaging examination; Pelvic endometriosis; Treatment; Case report

Core Tip: An inguinal hernia on the right side may be one of the causes of the formation

Specialty type: Medicine, research and experimental

Provenance and peer review: Unsolicited article; Externally peer reviewed.

Peer-review model: Single blind

Peer-review report's scientific quality classification

Grade A (Excellent): 0
Grade B (Very good): 0
Grade C (Good): C, C
Grade D (Fair): 0
Grade E (Poor): 0

Received: June 22, 2021
Peer-review started: June 22, 2021
First decision: September 1, 2021
Revised: September 29, 2021
Accepted: November 15, 2021
Article in press: November 15, 2021
Published online: December 26, 2021

P-Reviewer: Palmeri M, Parra RS
S-Editor: Fan JR
L-Editor: A
P-Editor: Fan JR

of right inguinal endometriosis. This condition may present clinically as a painful mass that can vary in size, possibly according to menstrual cyclicity. Preoperative imaging using ultrasound and/or magnetic resonance imaging may be useful for preoperative diagnosis. Extensive resection involving the round ligament and hernia sac is necessary to prevent recurrence.

Citation: Li SH, Sun HZ, Li WH, Wang SZ. Inguinal endometriosis: Ten case reports and review of literature. *World J Clin Cases* 2021; 9(36): 11406-11418
URL: https://www.wjgnet.com/2307-8960/full/v9/i36/11406.htm
DOI: https://dx.doi.org/10.12998/wjcc.v9.i36.11406

INTRODUCTION

Endometriosis (EM) is a disease characterized by the presence of tissue resembling endometrium outside the uterus, causing a chronic inflammatory reaction[1]. The disease is mainly found in women of childbearing age, mostly in the pelvis, with a prevalence in healthy women of 6%–10%[2]. Inguinal EM (IEM) occurs in 0.3% to 0.6% of patients affected by EM, a rare form of extraperitoneal partial round ligament involvement. It is often confused with other groin pathologies and is mostly diagnosed during histological examination[3,4]. IEM was first described by in 1986[5], and to date, fewer than 150 cases have been reported in the literature[6]. In addition, most of the cases related to IEM have been reported as individual cases.

IEM mainly occurs in the right groin area and is characterized by a groin mass with pain and periodic exacerbations or no pain but with menstrual symptoms. The size of the mass may change with the menstrual cycle or have a bleeding tendency[7]. Catamenial symptoms are present in 50% of patients[8,9]. Niitsu *et al*[10] classified IEM into three types according to the location of the endometrial tissue: Type I, hernial sac or hydrocele in the Nuck canal; type II, round ligament; and type III, under the skin. The etiology of IEM is still unclear, although several theories have been proposed, including but not limited to metaplasia of the mesothelium, retrograde menstruation and seeding into a hernia, oxidative stress, inflammation, autoimmune dysfunction, apoptosis suppression and regeneration of stem cells[11].

IEM is extremely rare, and its occurrence is usually associated with pelvic EM. It can also be easily misdiagnosed before surgery due to its coexistence with inguinal hernia (IH) and other inguinal lesions, which increases the difficulty of diagnosis. Therefore, it is essential to explore the pathogenesis, clinical features, diagnosis and differential diagnosis of IEM. This article retrospectively analyzed the clinical presentation, diagnosis, and treatment of 10 patients with IEM at our hospital and reviewed the relevant literature findings.

CASE PRESENTATION

Chief complaints

The medical records of all patients who underwent IEM surgery in Beijing Chao-Yang Hospital between 2011 and 2019 were retrospectively reviewed. The initial inclusion criteria were as follows: (1) Complete surgical resection; and (2) The presence of both endometrial glands and stromal cells in all lesions excised from the inguinal region. When IEM was diagnosed before or during the operation due to thickening of the hernia sac wall or round ligament, the following inclusion criteria were applied: (1) Complete removal of the hernia sac; (2) Relatively wide surgical margins (approximately 1 cm) for broad ligament resection; and (3) Adequate hernia repair, if necessary. Furthermore, we identified the number of female patients treated for pelvic EM or IH during the same period to estimate the incidence of IEM.

This research project was approved by the constituted Ethics Committee of our hospital (2016-Science-166) and conformed to the Declaration of Helsinki. Informed consent was given by all subjects, and patient anonymity was preserved.

Ten IEM patients were included in this case report; all diagnosed through surgery and pathology, and 4 were nulliparous. Table 1 summarizes the characteristics of all 10

Table 1 Clinical data of patients with inguinal endometriosis in this study

Index	Cases, *n* (%)	Percentage, *n* (%)
Age at diagnosis (yr)	38 (32-53)	
Age distribution	Total: 10	
30–39 yr	5	50
40–49 yr	3	30
50 yr and older	2	20
Parity		
Nulliparous	4	40
Dysmenorrhea		
Yes	7	70
No	3	30
Surgical history	6	60
Cesarean section	1	10
Pelvic endometriosis	4	40
Inguinal hernia	1	10
No surgical history	4	40
Department of admission		
Gynecology	2	20
General surgery	8	80
Laterality of inguinal endometriosis		
Right	8	80
Left	1	10
Right and left	1	10
Lesion size (cm)	3.2 ± 1.2	
Symptoms		
Presenting symptoms	10	100
Symptoms related to menstruation	5	50
Symptoms not related to menstruation	5	50
No symptoms (incidental findings)	0	0
Swelling		
Yes	10	100
No	0	0
Block size varies with body position		
Yes	9	90
No	1	10
Pain		
Yes	10	100
No	0	0

patients, including age, surgical history, and lesion characteristics. The median age at diagnosis was 38 years (range 32–53 years). Of the 10 IEM patients, four had a history of pelvic EM surgery, 8 in the right area of the groin, 1 on the left, and 1 on both sides.

All patients had pain and swelling in the groin. Among them, 9 had an increase in groin mass when standing and a slight decrease in groin mass when lying supine. Five patients had catamenial symptoms, including bloating and pain in the groin during menstruation, but the symptoms disappeared between menstruation periods. The duration of complaints prior to surgery ranged from 3 to 108 mo.

The mean size of the lesions was 3.2 ± 1.2 cm. Eight patients were admitted to the hernia department, and only two patients were treated by a gynecologist. A total of 2478 female patients underwent surgery for a hernia or swelling in the groin, and 1958 female patients underwent surgery for EM during the same period in our hospital. The prevalence of IEM in the two groups was 0.4% (10/2478) and 0.5% (10/1958), respectively.

History of present illness

The 10 cases were diagnosed of inguinal EM by surgery and pathology.

History of past illness

Five had prior pelvic EM, and four had infertility.

Personal and family history

All patients had no family history.

Physical examination

No special findings except for masses in the groin area were found under physical examinations.

Laboratory examinations

The blood ca125 of case 9 was slightly elevated to 78.5 U/mL, the blood ca125 of case 10 was normal, and the remaining 8 patients did not receive laboratory examinations.

Imaging examinations

Ultrasonography (US) was performed in all 10 patients (Table 2); 8 patients showed inconclusive heterogeneous lesions, and one patient with bilateral IEM had a heterogeneous, hypoechoic lesion on the left side and a cystic lesion on the right side. Only one patient was diagnosed with IEM by US, in which the mass appeared as a hypoechoic lesion with a cluster of small cysts in the right groin.

Magnetic resonance imaging (MRI) was performed on 2 patients who were treated by a gynecologist. As shown in Figure 1, case 10 (cystic lesion) showed a hyperintense nodule in the wall of a cystic lesion on T1-weighted axial imaging. Furthermore, T1-weighted contrast-enhanced imaging showed an enhanced hyperintense lesion in the wall of the cystic lesion. Case 9 (solid mass) showed mixed hypointense and hyperintense patterns on T1-weighted and T2-weighted imaging, but T1-weighted contrast-enhanced images showed an enhanced hyperintense pattern at the same lesion. Therefore, the IEM lesion showed specific performance on MRI, in which a hyperintense pattern was found on T1-weighted images or T1-weighted contrast-enhanced images, particularly the latter.

FINAL DIAGNOSIS

A total of 10 cases of IEM diagnosed by surgery and pathology were characterized by a lesion on the right side.

TREATMENT

As shown in Table 1, all patients received surgical intervention through a skin incision in the groin, and the involved extraperitoneal round ligaments were resected extensively. In addition, 9 patients underwent complete excision of the IH sac and hernia repair. Simultaneous laparoscopic exploration of the pelvis was performed only for case 9, and the EM lesion in her right sacral ligament was completely removed.

Case 10 (Figure 2) had EM lesions in both inguinal areas. The inner wall of the right hernia sac was locally thickened with dark red fluid and adhesions, and the left extraperitoneal round ligament was significantly thickened with a purplish-blue lesion on the cut surface. Extensive excision of the left extraperitoneal round ligament and

Table 2 Characteristics of patients who underwent surgery for inguinal endometriosis

Case	Age (yr)	EM history	Parity	Surgery year	Laterality	ICS	Preoperative diagnosis	US/MRI	Site of EM	Surgery	Postoperative diagnosis	Follow-up (mo)
1	47	-	P	2018	R	-	IH/cyst of Nuck's canal?	US: Heterogeneous echo; MRI:/	Extraperitoneal round ligament	Wide excision of the lesion; Hernia repair	IEM (R); IH (R)	24
2	52	+	P	2016	R	-	IH/IEM?	US: Low echo with cluster; Cysts (IEM); MRI:/	Extraperitoneal round ligament	Wide excision of the lesion; Hernia repair	IEM (R); IH (R)	41
3	33	-	P	2017	R	+	IH/cyst of Nuck's canal?	US: Heterogeneous echo; MRI:/	Extraperitoneal round ligament	Wide excision of the lesion; Hernia repair	IEM (R); IH (R)	24
4	53	-	P	2018	R	+	IH	US: Heterogeneous echo; MRI:/	Extraperitoneal round ligament	Wide excision of the lesion; Hernia repair	IEM (R); IH (R)	12
5	35	+	N	2018	R	-	IH	US: Heterogeneous echo; MRI:/	Extraperitoneal round ligament	Wide excision of the lesion; Hernia repair	IEM (R); IH (R)	19
6	36	+	N	2019	R	-	IH	US: Heterogeneous echo; MRI:/	Extraperitoneal round ligament	Wide excision of the lesion; Hernia repair	IEM (R); IH (R)	13
7	42	-	P	2015	R	+	IH	US: Heterogeneous echo; MRI:/	Extraperitoneal round ligament	Wide excision of the lesion; Hernia repair	IEM (R); IH (R)	60
8	40	+	N	2012	L	-	IH	US: Heterogeneous echo; MRI:/	Extraperitoneal round ligament	Wide excision of the lesion; Hernia repair	IEM (L); IH (L)	70
9	32	-	N	2014	R	+	IEM; Pelvic EM	US: Heterogeneous echo; MRI: IEM (R)	Extraperitoneal round ligament	Wide excision of the lesion	IEM (R); Pelvic EM	88
10	36	-	P	2011	B	+	Bilateral IEM; IH (R)	US: Heterogeneous echo (L) + cyst (R); MRI: IEM (bi)	Extraperitoneal round ligament (L); Hernial sac (R)	Wide excision of the lesion (L + R); Hernia repair (R)	IEM + IH (R); IEM (L)	97

ICS: Inguinal catamenial symptom; IH: Inguinal hernia; IEM: Inguinal endometriosis; N: Nulliparous; P: Parous; US: Ultrasonography; MRI: Magnetic resonance imaging; EM: Endometriosis.

the right hernia sac was performed. In addition, the hernia on the right side was repaired. All specimens underwent routine pathological examination, and characteristic endometrial glands with stromal cells were found in all cases.

Only one patient (Case 9), who was surgically confirmed to have pelvic EM, was treated with gonadotropin-releasing hormone agonist (GnRH-a) for six months postoperatively. The remaining nine patients did not receive any drugs.

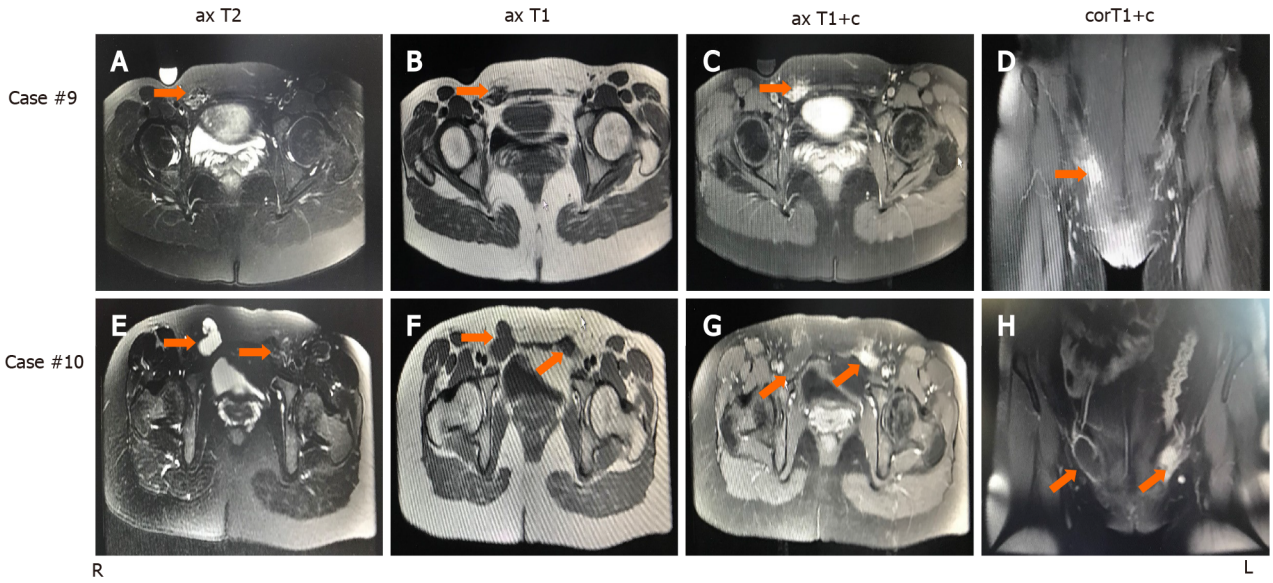

Figure 1 Magnetic resonance imaging findings of inguinal endometriosis. Magnetic resonance imaging findings of Case 9 (A–D) and Case 10 (E–H). A: Fat-suppressed T2-weighted axial image shows a mixed hyperintense and hypointense lesion with irregular edges in the shape of a cluster of dots and foci in the right groin; B: T1-weighed axial image shows a mixed hyperintense and hypointense lesion in the right groin; C: T1-weighted contrast-enhanced axial image shows an enhanced hyperintense lesion in the right groin; D: T1-weighted contrast-enhanced coronal image shows an enhanced hyperintense lesion in the right groin; E: Fat-suppressed T2-weighted axial image shows a hyperintense cyst in the right groin and a mixed hyperintense and hypointense lesion with irregular edges in the shape of a cluster of dots and foci in the left groin; F: T1-weighed axial image shows hyperintense nodules in part of the wall of the right hernia sac and a mixed hypointense and hyperintense lesion in the left groin; G: T1-weighted contrast-enhanced axial image shows an enhanced hyperintense lesion in part of the thickened wall of the right hernia sac and an enhanced hyperintense lesion in the left groin; H: T1-weighted contrast-enhanced coronal image shows an enhanced hyperintense nodule in the wall of the right hernia sac and an enhanced hyperintense lesion in the left groin.

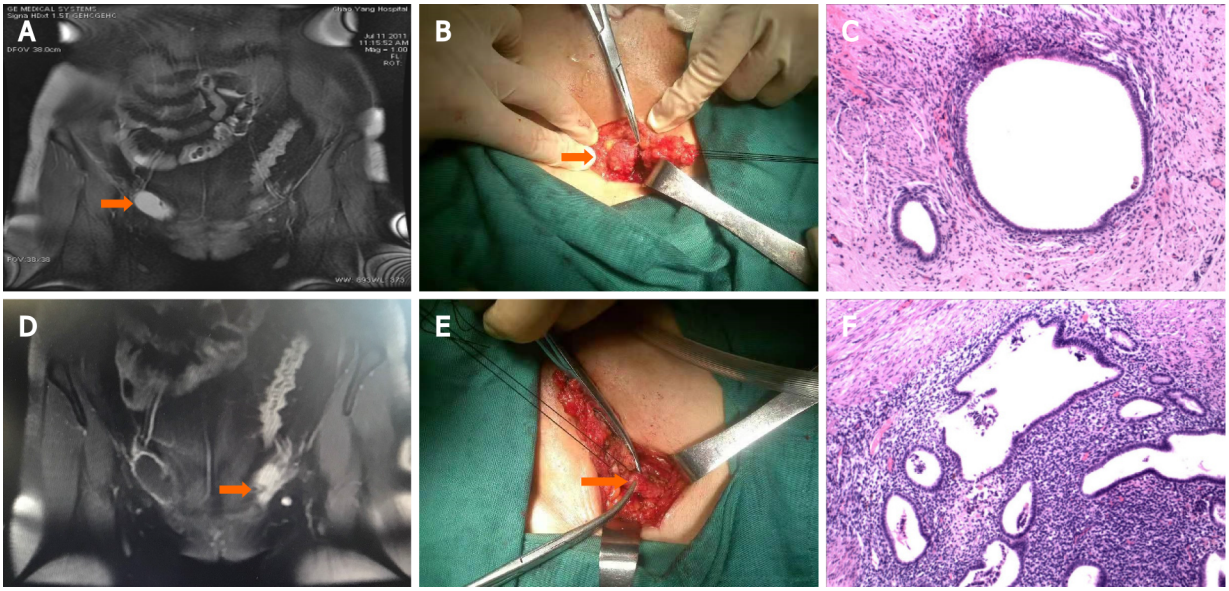

Figure 2 Magnetic resonance imaging, intraoperative findings and histological appearance of case 10. A: The arrow points to the endometriosis lesion in the sac of the right inguinal hernia; B: The arrow points to the sac of the right inguinal hernia; C: Typical endometrial glands and stroma in the right hernia sac. Hematoxylin and eosin (H&E) staining, original magnification × 100; D and E: The arrow points to the extraperitoneal round ligament in the left inguinal region; F: Typical endometrial glands and stroma surrounded by smooth muscles. H&E, original magnification × 100.

OUTCOME AND FOLLOW-UP

The median follow-up time was 36 mo (range 12–108 mo), and no complications or recurrence of IEM were observed. In the follow-up of 4 patients, 2 conceived spontaneously and delivered at full term within 2 years. One patient underwent assisted reproduction and full-term delivery 18 mo after surgery, and one patient remained infertile with adenomyosis 2 years after surgery.

Table 3 summarizes the characteristics, symptoms, imaging examinations, surgical treatments, medications and follow-up results of our cases and 14 other retrospective case reports related to IEM.

DISCUSSION

It is challenging to recognize IEM, which is therefore often confused with more common diseases, such as hernias, soft tissue tumors, cysts, lymphadenopathy, granulomas, and hydrocele. Catamenial symptoms, such as changes in size and tenderness, appear in 50% of patients, indicating that the suspicion index for IEM should be increased[12]. IEM is prone to misdiagnosis before surgery[11,13], possibly due to its uncommon location, the noncyclic pain pattern in a number of patients[14-16] or the inconclusive results from US[17,18]. Symptomatic patients often visit a surgeon rather than a gynecologist[3,6,10,19], so IEM is often misdiagnosed as a common groin disease such as IH[17,20]. More explicitly, IEM can be discovered in the extraperitoneal part of the round ligament, subcutaneous fat tissue, inguinal lymph nodes, and even in the sac wall of the inguinal or femoral hernia[6,21].

In our study, 0.5% of patients affected by EM developed IEM, which is consistent with the literature[3]. According to reports, 72%–90%[3,10,17,19] of patients experience right IEM. Similarly, our series of results showed that 9 out of 11 lesions were on the right. Hypotheses for the predominance of the right side in IEM are as follows[3,8,9,19]. First, due to its clockwise circulation, peritoneal fluid persists on the right side for an extended period of time in patients with EM. Second, the sigmoid colon prevents peritoneal fluid from flowing into the left inguinal ring. Third, pre-existing EM of the pelvis makes the round ligament a route for the groin. Pathological diagnosis is still the key for identifying IEM. The characteristic finding is the presence of endometrial glands with stromal cells in the connective tissue.

In our study, 8 patients had a right IH, which thus may be one of the causes for the formation of right IEM. The exact pathogenesis of IEM has not been well explained[15,22]. In the present study, most of the patients with IEM had IHs on the same side, and approximately half of the patients also had pelvic EM. Therefore, regional anatomical defects in the groin and pre-existing pelvic EM may be the main causes of IEM. Although there is an association between abdominal wall EM (*e.g.*, scar EM) and cesarean delivery, a possible association of IEM with cesarean delivery has not been reported. Only one of the 10 patients in this study had a history of cesarean delivery, and this patient also had ipsilateral IH, which did not suggest an association with IEM. The preoperative diagnosis of IEM remains a clinical challenge. Although catamenial symptoms should increase the index of suspicion for IEM[3,8,9,19], typical catamenial symptoms are present in only 50% of IEM cases. Due to the uncommon location of IEM lesions, symptomatic patients usually see a general surgeon rather than a gynecologist[3,6,15,17,19,20], and the diagnosis is frequently incidental or is made upon histologic examination.

A groin lump or subcutaneous mass of the inguinal area can be difficult to distinguish from a wide range of entities, such as hernia, lymph node enlargement, cancer, EM, Nuck hydrocele, lipomas, and abscess[23]. Cutaneous EM with a hernia sac can occur in the absence of normal EM symptoms such as dysmenorrhea. The differential diagnosis for inguinal painless nodules might include IEM[24]. If EM in the inguinal area is detected, it might be within the IH sac or EM of the Nuck canal (NC), round ligament, and subcutaneous tissue[25]. The most frequent manifestation of IEM is a painful lump with cyclical discomfort and growth[26]. Preoperative imaging using US and/or MRI may be useful in this regard, although surgical exploration and histology may clearly establish the genesis and ultimate diagnosis of the disease[13]. Additionally, features of the discomfort, such as frequency, duration, relationship with menstruation, and location, should be extensively questioned. Likewise, appropriate preoperative imaging is important. Ultrasound has been widely used in the examination of the pathology of IEM[27,28].

Computed tomography (CT), MRI, or even a positron emission tomography-CT (PET-CT) has been used to augment the US findings in some situations. High-resolution ultrasound is a low-cost and dependable technique for establishing the suspicion of an IEM diagnosis[29]. Nonetheless, the final diagnosis is based on histology and immunohistochemistry.

In our study, eight patients were treated by a non-gynecologist, and only three patients were diagnosed preoperatively, which is consistent with less than 50% of most reports reported in the literature[3,13,17,19]. In addition, IEM is often associated

Table 3 Literature reviewed of this study

Ref.	Case number	Age (yr)	Side	Size (cm)	EM history, *n* (%)	Reproductive history	Presenting symptoms	Imaging	Preoperative diagnosis	Site of lesion	Operation	Recurrence
Kapan *et al* [14], 2005	3	39-51	R: 2; L: 1	2-4	/	/	Groin lump (100%)	/	0	Type I (1); Type II (2)	Wide excision of lesion (3); Hernia repair (3)	/
Gaeta *et al* [22], 2010	8	30 (22-46)	R: 8	1.5-4.5	/	/	Groin lump (100%); Pain (50%); Catamenial symptom (25%)	MRI: Type I: Prevalently cystic (2/8); type II: Prevalently solid with small scattered cysts (6/8)	100%	Type I (8)	Wide excision of lesion (8)	/
Sun *et al* [9], 2010	9		R: 8; L: 1	/	/	/	Catamenial symptom (66%)	/	33%		Excision of inguinal lesion (8/9); Extra round ligament (1/9); Laparo-scopy (4 pelvic EMs)	0%
Wong *et al* [39], 2011	1	48	R	4 × 5	No	Gravida 3, para 3	Period pain at the groin during menses	US: A slightly bulky uterus	Fineneedle aspiration biopsy of the mass revealed EM	Proliferative endometrium	Solid, fibroid-like tumor was removed from the right groin	Remained asymptomatic and underwent a second exploratio: Revealed a multinodular subinguinal endometriotic lesion
Rajendran *et al*[15], 2012	1	36	L	2 × 2		Crampy lower abdominal pain and a lump in her left groin. The lump present for 3 yr		CT: Mass adjacent to the rectus femoris muscle. US: A 2 × 2 cm solid mass with evidence of blood flow at the posterior aspect		Biopsy of the lesion revealed endometrial tissue		
Albutt *et al* [44], 2014	1	23	L	2.1	No	No	A new-onset tender bulge with subjective fevers and chills	US: Avascular complex cystic lesion measuring 2.1 cm in the left groin. CT: A tubular cystic structure along left inguinal canal, round ligament	Inguinal hernia	Fpathology: A hydrocele with concordant EM	The cystic structure was dissected away from the round ligament	no
Mourra *et al*[17], 2015	42	35 (20-53)	R: 29; L: 11; Unk: 2	3.36 (1-5)	5	/	Groin lump (100%)	/	31%	/	Wide excision of lesion (42); Hernia repair (8); Laparo-scopy (4)	1 (2.38%)
Çayır *et al* [37], 2018	1	35	R	2.5 × 1.5				US: Hypoechoic solid mass of 2.5 cm × 1.5 cm				

Wolfhagen *et al*[13], 2018	9	32.5 (27-43)	R: 7; L: 2	/	0	P: 4; N: 5; S: 2	Groin lump (100%); Catamenial symptom (44%)	US: 1/7 suggestive for IEM; MRI: 0/4 suggestive for IEM; Image: 3/7 suggestive for hernia; Fin: 1/2 suggestive for IEM; 1/2 inconclusive	33%	Type I (7); No mention (2)	Wide excision of lesion (9); Laparo-scopy (1 pelvic EM)	0%
Niitsu *et al* [10], 2019	28	20-50	R: 25; L: 3	1-3.3	4	/	Groin lump (100%); Catamenial symptom (57.1%)	US: Low or with cyst (28/28); CT: Soft tissue density (18/18); MRI: T1 low/T2 low (3/3)	71.4%	Type I (15); Type II (10); Type III (3)	Wide excision of lesion (28)	2 (7.1%)
Arakawa *et al*[18], 2019	20	37.2 ± 6.7	R: 13; L: 5; R and L: 2	2.4 ± 1.1	11	P: 3; N: 17	Groin lump (100%); Pain (100%); Swelling (70%); Catamenial symptom (80%)	US: Solid mass (15/20), cystic mass (2/20), mixed (1/20), no record (2/20); CT: Inguinal mass (13/13); MRI: Solid (9/18), cystic + solid (8/18), cystic (1/18)	5/6	No mention	Operation: Radical excision of lesion (5/6), Wide excision of lesion (1/6); Hormone: OC (4/8), DNG (4/8); Chinese medicine (1); No treatment (5)	1 (5%)
Jena *et al* [4], 2020	1	25	r	3 × 2	No		2-yr history of painful persistent mass in the right groin and her symptoms fluctuated with the menstrual cycle	MRI: 2.7 cm × 1.7 cm × 1.6 cm heterogeneous nodular lesion in the right inguinal subcutaneous plane superficial to the adductor muscles and at the lower edge of the rectus abdominis muscle	Inguinal hernia	Mass showed the possibility of intramuscular endometriosis	Excision of the lesion and the	Patient was symptom free on subsequent follow-up
Zihni İ *et al*[25], 2020	1	31	r	2.1 × 1.2		The patient had given birth by caesarean section 2 yr previously	Pain and swelling in the right inguinal area. The complaints had been ongoing for approximately 1 yr, and the pain and swelling increased undertaking strenuous labour	US: A cystic structure, 21 mm × 12 mm in size, was seen within the hernia pouch in the right inguinal canal				
Basnayake *et al*[23], 2020	1	27	r	4 cm × 4 cm	No	No	Enlarging, painless, right inguinal swelling of 4 mo duration	US: Multiloculated, thin septated, anechoic cystic swelling without increased internal vascularity at	There was no demonstrable hernia	The histology: Type I endometriosis	A complete excision of the cyst was performed	Follow-up after 1 yr showed no evidence of recurrence

							the right inguinal region					
This study	10	38 (32-53)	R: 8; L: 1; B: 1	3.2 ± 1.2	4	P: 6; N: 4	Groin lump (100%); Pain (100%); Swelling (100%); Catamenial symptom (50%)	US: Low echo with cluster cysts (1/10); heterogeneous mass (8/10); heterogeneous low echo lesion (L) + cyst echo (R) (1/10); MRI: T1 +c high (2/2)	30%	Type II (9); Type I + II (1)	Wide excision of lesion (10); Hernia repair + mesh (9)	0%

EM: Endometriosis; MRI: Magnetic resonance imaging; N: Nulliparous; P: Parous; S: Subfertile; US: Ultrasonography.

with IH. In 2 large case studies reported by Niitsu *et al*[10] and Mourra *et al*[17], the prevalence of IEM with IH was 54% and 19%, respectively. In our study, the incidence was 90% (9/10). General surgeons tend to focus on the IHs and ignore the presence of IEM, leading to preoperative misdiagnosis. Compared with gynecologists, general surgeons encounter IEM more often. Since this diseases happens rarely, diagnosis is very challenging. It has been stated in many series that general surgeons have performed operations on patients diagnosed with IEM[30]. In these rare cases, it may be helpful if the patient has a history of swelling in the groin area, especially during menstruation or a history of uterine surgery. However, a definite diagnosis can only be made after pathological examination[31]. Although most patients experience pelvic EM, there are also a considerable number of patients with extrapelvic manifestations [32]. Association of the anterior abdominal wall or inguinal groin area is usually correlated with previous surgery, secondary to implantation, and it is rare for IEM lesions to appear in these areas in patients who had not undergone prior surgery[33].

Ultrasound is usually used as the first noninvasive imaging approach for inguinal lesions, but it has limited diagnostic value for IEM. US and CT usually cannot diagnose IEM[3]. Since IEM simulates common surgical conditions in the area, including hydrocele of the canal of Nuckand IH, imaging methods such as US are helpful before surgery. These diseases can yield imaging signs such as cystic components or hypoechoic solid masses[34]. US is usually unable to provide a definitive diagnosis because IEM masses frequently show indeterminate hypoechoic, heterogeneous, or cystic lesions[17,35,36]. We also confirmed that only one patient with a history of pelvic EM was suggested to have IEM on ultrasound. Therefore, the preoperative diagnosis rate based on US alone is very low. If general surgeons focus on the periodic pain pattern of the inguinal mass and the patient's previous history of EM, the preoperative diagnosis rate might be greatly improved (71.4%)[10].

It has been stated that MRI is a valuable diagnostic method for pelvic EM, with a specificity of 98% and a sensitivity of 90%[22], as MRI can recognize iron in hemosiderin deposits in endometrioma, so pure fibrous lesions are hypointense on T1- and T2-weighted scans, whereas hemorrhages are hyperintense on fat-suppressed T1-weighted or T1-weighted scans. Gaeta *et al*[22] indicated that there are two main MRI patterns of IEM on T1-weighted images: A solid hypointense mass with a hyperintense hemorrhagic cyst and clusters of hemorrhagic cysts. The role of MRI in the diagnosis of thoracic/pelvic EM has been investigated, and it was discovered that MRI has significantly greater accuracy than other tools, but the role of MRI in abdominal wall EM is undefined[36]. It has been reported that IEM was accidentally found in systemic iodine 131 (I-131) scans following total thyroidectomy[37].

In our study, the IEM lesion located in the round ligament presented with mixed hyper- and hypointensity with an irregular edge in the shape of a cluster of dots and foci on T1- or T2-weighted images, but the lesion in the hernia sac presented as a cystic lesion with hyperintense nodules in part of the cyst wall on T1-weighted images. All lesions presented an enhanced, hyperintense pattern on T1-weighted contrast-enhanced images. T1-weighed contrast-enhanced imaging clearly showed a slight lesion in the hernia sac of case 10. Therefore, extensive use of MRI can improve the accuracy of the preoperative diagnosis of IEM.

Drug treatment for IEM has been poorly reported and is controversial. For isolated pelvic ectopic EM, postoperative suppressive hormone therapy is not necessary[3]. Hormone suppression therapy should only be used for patients with pelvic EM treated laparoscopically[38].

Surgical treatment has been reported in most cases[10,14,39]; primary treatment of IEM relies on extensive resection of the lesion to avoid subsequent recurrence[40]. Niitsu *et al*[10] reported three cases of recurrence separately from the subcutaneous area, the remnant hernia sac and the distal end of the round ligament. In our study, removal of the affected round ligament with a 1 cm surgical margin was carried out in all patients. Additionally, all-inclusive resection of the hernia sac and hernia repair were performed on nine patients. It is critical to diagnose IEM prior to surgery. Based on the preoperative diagnosis, surgeons should select suitable surgical techniques depending on the anatomical location of the IEM lesion[41]. When the general surgeon performs IH surgery and finds significant thickening of the hernia sac wall, adhesions, bloody fluid inside the sac, or significant thickening of the extraperitoneal round ligament and purple–blue lesions on the cut surface, the possibility of IEM should be considered, and a gynecologist should be asked to consult on the staging, remove the hernia sac and perform complete excision of the IEM lesion as well as evaluate the presence of pelvic EM and, if necessary, perform laparoscopic exploration, especially in patients with infertility.

Finally, to prevent intraoperative cell implantation, proper normal saline irrigation should be performed prior to wound closure. To minimize the risk of artificially inducing EM, it is recommended that gloves, suture materials and sponges be changed in the final stage of uterine surgery. In cesarean section surgery, it is beneficial to clean the operative field with a high-flow saline solution before closing the incision line[42]. Due to the possibility of pelvic EM, laparoscopic diagnosis or postoperative referral to a gynecologist should be performed at the same time, especially for infertile women[3, 18-20]. Individualized clinical treatment should be provided in collaboration with general practitioners and obstetrics and gynecology specialists[43].

In this study, there was only one patient for whom the coexistence of pelvic EM was surgically confirmed and who received GnRH-a treatment six months after surgery. The remaining nine patients did not receive any medication. In the short-term and long-term follow-ups, none of the patients experienced postoperative groin discomfort. Therefore, for IEMs without pelvic EM, extensive surgical resection is sufficient. For larger lesions that are highly suspected of IEM, preoperative hormone therapy may sometimes be considered to help achieve radical resection. IEM has been shown to be associated with pelvic EM, and some authors recommended simultaneous laparoscopy to evaluate concomitant pelvic EM[3,10,18,20,44].

Follow-up showed that none of the patients had recurrence of IEM or IH. Therefore, extensive resection of the involved round ligament and hernia sac is the key to preventing recurrence, but this needs to be confirmed by more clinical IEM cases. Because IH occasionally coexists in patients with IEM, peritoneal pseudomyxoma, perivascular epithelioid cell tumors, and especially malignancy have been infrequently found in the hernia sac, some of which may not show serious abnormalities; hence, even if the surgeon is not aware of the coexistence of IEM, the hernia sac should be removed as widely as possible, and pathological examination should be performed. If malignant tumors are suspected, cryosectioning should be performed, as this may help in the decision to perform more aggressive surgical procedures. No developmental abnormalities or malignant changes were encountered in our series. It is very rare for EM lesions in the lateral wall of the pelvis to undergo malignant transformation. However, oncologists should pay greater attention to patients with a history of EM or pelvic surgery[45-48]. In our study, 5 patients had pelvic EM, including 4 patients with a surgical history of pelvic EM and 1 patient who underwent simultaneous pelvic EM surgery. Four out of these 5 patients were infertile. Therefore, it is necessary for a gynecologist to evaluate concomitant pelvic EM, especially for infertile patients. Treatments include complete surgical excision with minimal spillage to avoid recurrence, hormonal therapy and anticipatory management[3]. Gynecology referral is recommended because pelvic EM often occurs at the same time as IEM. IEM rarely recurs after appropriate extensive surgical resection[10,17,19], and postoperative hormonal therapy is not recommended for patients with isolated pelvic EM who have undergone appropriate surgical treatment[38].

CONCLUSION

IEM is extremely rare and can easily be misdiagnosed before surgery. MRI helps in making a preoperative diagnosis. Right IH may be one of the reasons for the formation of right IEM, and extensive resection is the key to preventing IEM recurrence. Since IEM is usually associated with pelvic EM, consultation with a gynecologist is

recommended. In this article, the clinical characteristics, pathogenesis, diagnosis and differential diagnosis of IEM were further enriched by the clinical management experience of the 10 patients we treated and by a systematic literature review. In women with inguinal masses associated with menstrual cycle changes (especially on the right side), gynecological disease-IEM cannot be excluded.

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