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Torqued Pedunculated Inflammatory Myofibroblastic Tumour of the Greater Omentum (GO-IMT) Presenting as Gynaecological Emergency in an Adolescent Girl: Case Report and Representative Literature Review

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Keywords:

Inflammatory myofibroblastic tumour; Torsion; Gynaecological emergency; Imaging; Greater omentum

1. Abstract

1.1. Background and Objectives: Material and methods: A rare case of a torsion of pedunculated Inflammatory Myofibroblastic Tumor (IMT) from the Greater Omentum (GO) in an adolescent girl, mimicking torsion of pedunculated leiomyoma is presented. IMT is a rare pathology, with pelvic location and torsion of pedunculated IMT from the GO being extremely rare. Surgery is the treatment of choice. However, the treatment was severely delayed in our patient due to signs of infection, unrecognized pathology and multiple transfers among departments. To our knowledge, this is the first case reported of a torsion of pedunculated GO-IMT, located in the pelvis and representing as gynaecological emergency.

2. Introduction

Inflammatory Myofibroblastic Tumour (IMT) is a distinctive neoplasm that was first identified in the lungs in 1939 [1]. Since then, it has been described in children and young adults in the head and neck region as well as in the abdomen and pelvis [2]. The World Health Organization classified IMTs as intermediate neoplastic lesions, since they may show a tendency for local recurrence, can be aggressive and sometimes even metastasize [3]. These tumours are solid, well- or ill-defined, with in-homogenous and at least some degree of delayed enhancement due to fibrotic tissue, both on contrast-enhanced Computed Tomography (CT) and Magnetic Resonance Imaging (MRI), which are the most used radiological modalities for their evaluation.4 Since these radiological features are not specific, the final diagnosis is based on histopathology where the tumour is comprised of differentiated myofibroblastic spindle cells accompanied by large amounts of plasma cells and/or lymphocyte infiltration. In approximately 50% of IMT's there is a characteristic Anaplastic Lymphoma Kinase (ALK) tyrosine kinase activation and ALK protein overexpression [2]. Complete surgical resection remains the treatment of choice with a regular follow-up after surgery due to relatively high possibility of local recurrence.4 Rarity of this pathology represents a diagnostic challenge for clinicians as well as radiologist and clinical pathologists evaluating the specimens taken in operating theatre.

We here report of a rare case of torqued pedunculated Inflammatory Myofibroblastic Tumour (IMT) of the Greater Omentum (GO)

presenting as gynaecological emergency and mimicking a torqued leiomyoma on Magnetic Resonance Imaging (MRI) in a 16-year-old girl.

3. Case Report

3.1. Consent

Written informed consent was obtained from the patient's parents for publication of this case report and any accompanying images. (A copy of the written consent is available for review at the Editor-in-Chief of this journal.)

3.2. Case History and Laboratory Values

A 16-year-old female with no comorbidities presented with acute onset of perianal pain. She was not vomiting and had no diarrhoea. She has been taking oral hormonal contraception (combination of 2 mg dienogest and 0,03 mg ethinylestradiol daily) for the last 6 months for dysmenorrhea. Her last menstrual period was 12 days before the onset of the problems, she was never pregnant (virgo) and on her last gynaecological visit 6 months earlier no leiomyoma of the uterus as well as no other pathology were seen with abdominal US. She never had any operative procedures done before or never suffered abdominal trauma. At examination, her vital signs were normal and the laboratory showed high inflammatory markers (leucocytes 13.9 × 10^9/L, C-reactive protein 39 mg/L). Since she was not constipated and urinary infection was excluded, empiric therapy with Amoxicillin/clavulanic acid and metronidazole intravenously was initiated. However, no resolution of either clinical signs or inflammatory markers was noticed.

3.3. Imaging

US per rectum was performed by a gynaecologist showing a large solid mass (78 × 53 mm in size) behind the uterus looking like a leiomyoma (Figure 1) with the uterus and both ovaries appearing normal. Since the mass had not been seen on gynaecological examination and transabdominal US preformed 6 months earlier, the decision was made to extend imaging diagnostics. Transabdominal US showed a well-defined solid lobulated mass measuring 80 x 51 mm, with minimal free fluid in the pelvis and no other pathology neither in the pelvis nor in the rest of the abdomen. Pelvic contrast-enhanced MRI was performed to further characterize the mass. A well-delineated, lobulated mass, measuring 6.2 × 4.8 × 7.2 cm, located to the right of the uterus and in front of the right ovary was seen (Figure 2). The mass was separated from both the uterus and the ovary. There was a pedicle originating from the anterior side of the mass, with the origin of the stalk not clearly seen at the time of initial evaluation. The mass had an in-homogenous internal signal on T2-weighted (T2-w) images. No fat content was shown inside the mass neither on T2-weighted fat-suppressed nor T1-w chemical shift images. The anterior part of the mass was enhanced after Gadolinium contrast application. Contrary, the posterior half showed no enhancement. No clear restriction of diffusion was seen on diffusion weighted images (DWI). The pelvic fat around the mass was structurally normal. There was a small amount of free liquid in the pelvis without any liquid collections. The uterus and both ovaries were structurally and morphologically normal, as well as the rectum, the bladder and the bowel. Although there was no clear connection of the mass to the uterus, we concluded that the mass could be a torquated pedunculated subserous leiomyoma uteri with differential diagnosis of torquated pedunculated Gastrointestinal Stromal Tumour (GIST).

3.4. Operative Treatment

The patient was transmitted to the department of gynaecology where laparoscopic surgery was performed. The mass in the pelvis was surrounded by adhesions. It was connected to the omentum by a pedicle and twisted around it four times (Figure 3). The uterus, both ovaries and uterine tubes appeared normal. During the surgery, the mass was separated from omentum, together with its pedicle inserted in the endo-bag. The mass without abdominal contamination (in endo-bag) was released from the pelvis and sent to pathohistological examination.

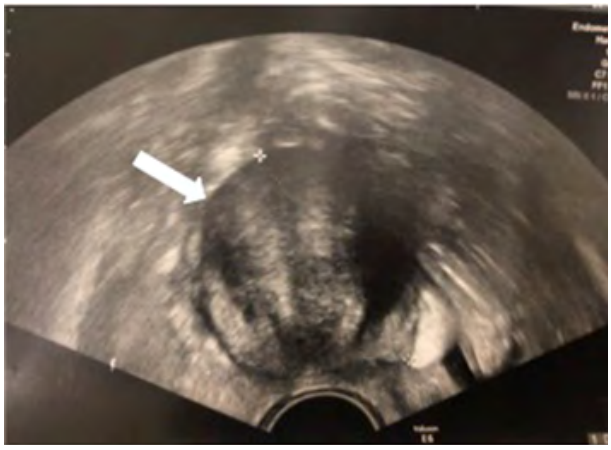

Figure 1: Ultrasound per rectum showed a large solid mass (arrow), measuring 78 × 53 mm, located behind the uterus. The mass had the apperance of a leiomyoma.

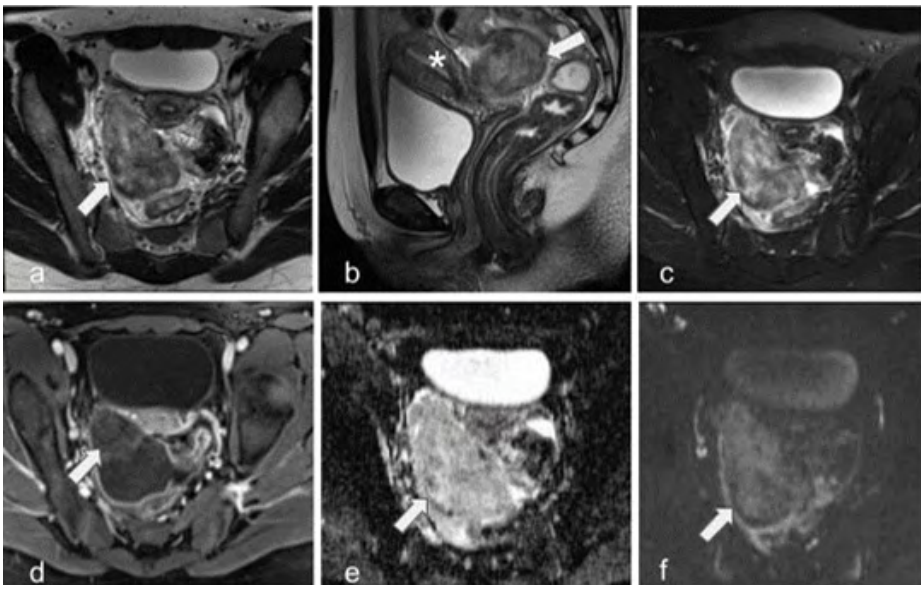

Figure 2: T2 weighted (T2w) pelvic magnetic resonance (MR) images in (a) axial and (b) sagittal plane showed well-defined pelvic mass (arrows) measuring 6.2 × 4.8 × 7.2 cm with in-homogenous internal signal. The mass was clearly separated from the uterus (*). (c) T2w fat saturated image in axial plane showed there is no fat content inside the mass (arrow). (d) Contrast-enhanced T1w image in axial plane displayed enhancement only in the anterior part of the mass (arrow). (e and f) No clear restriction of diffusion was seen neither on axial images with (e) high b values nor on (f) ADC maps (arrows).

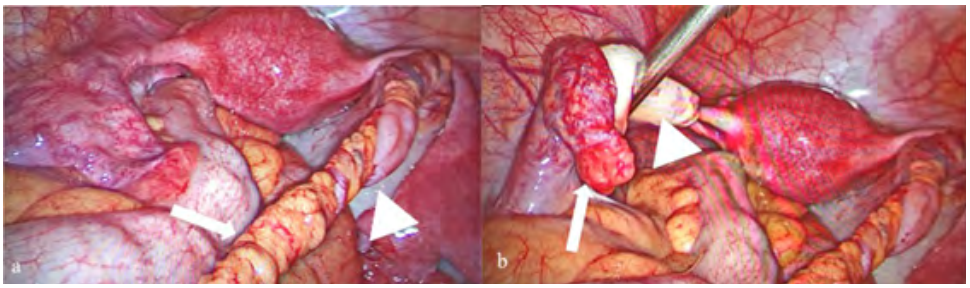

Figure 3: a and bThe tumour (arrowheads in a and b) was located in the puch of Douglas and was surrounded by adhesions. It was attached to the omentum by a slender pedicle (arrows in a and b) and torquated four times. However, vessels were not completely closed. The uterus and both adnexa were normal.

3.5. Pathohistology

a gross examination showed a tumour sized 8 x 5,5 x 5 cm that appeared white in the proximal half and haemorrhagic in the distal half, most likely due to torsion. On the cut section, the tumour was of medium hard consistency, it appeared solid and fascicular (Figure 4).

Histologically, the tumour was composed of moderately cellular spindle cells with variably collagenous and myxoid matrix. Tumour cells had ovoid or tapering nuclei and indistinct palely eosinophilic cytoplasm. There was an admixed chronic inflammatory infiltrate consisting mainly of plasma cells along with smaller numbers of lymphocytes and eosinophils (Figure 5a,b).

Immunohistochemistry stains showed positivity for SMA, CK AE1/AE3 and WT-1, whereas ER, PR, inhibin, calretinin, MUC-4, STAT-6, EMA, CD34, S100, caldesmon and desmin were negative. There was equivocal positivity for ALK (Figure 5c).

The final diagnosis was hyalinized inflammatory myofibroblastic tumour.

3.6. Hospitalisation and Follow-Up

Otherwise, the hospitalisation of our patient was uneventful and she was discharged home on the 5th day (2nd postoperative day) pain free, afebrile and with normal values of inflammatory markers. Our patient has regular follow-up appointments with her chosen gynaecologist performing abdominal US and up to this day there were no clinical, laboratory and US signs of the recurrence of IM

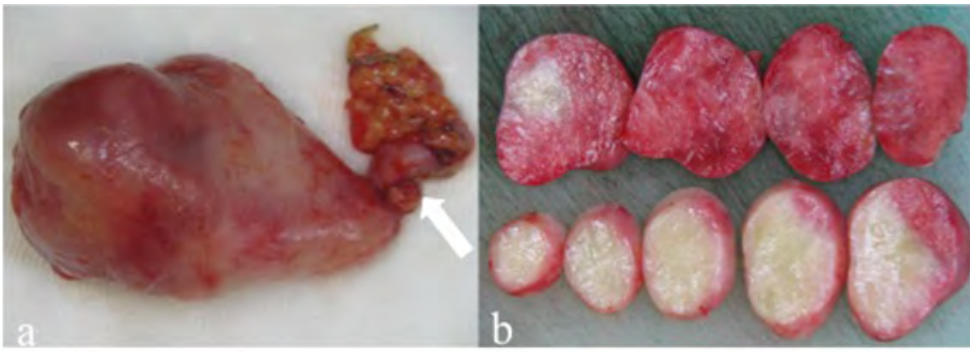

Figure 4: Macroscopically, the tumour (a) was attached to the omentum by a slender pedicle (arrow), and twisted around it four times. On the cut surface (b), the tumour was of medium hard consistency, fascicular, white in the proximal half and haemorrhagic in the distal half.

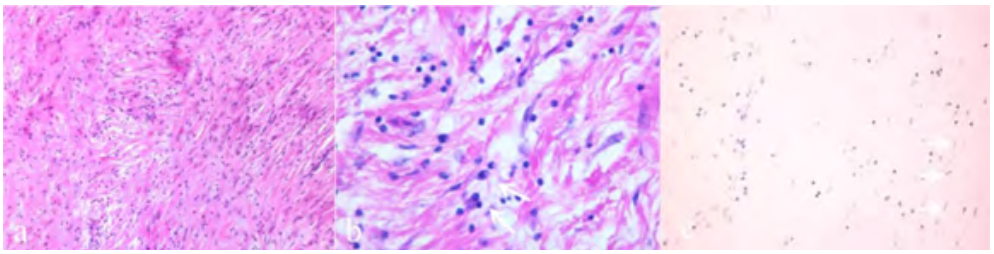

Figure 5: Histologically, the tumour (a) was composed of myofibroblastic mesenchymal spindle cells embedded in a collagenous and myxoid matrix, and (b) accompanied by an inflammatory infiltrate, consisting of plasma cells (arrow), lymphocytes, and eosinophils. Immunohistochemistry for ALK was mostly negative (c), however rare areas showed pale positivity, possibly cytoplasmic (arrows), that could not be undoubtedly distinguished from an artefact.

4. Discussion

Inflammatory Myofibroblastic Tumour (IMT) is a rare distinctive neoplastic lesion which comprises myofibroblastic spindle cells and inflammatory cells.5 It typically occurs in the abdomino-pelvic region and lungs of children and young adults. Nevertheless, IMTs can also arise in other anatomical sites and can also be found in elderly [2,6]. The World Health Organization classified IMTs as intermediate neoplastic lesions, since they can be aggressive, may show a tendency for local recurrence and can occasionally metastasize − distant metastasising occurs in <5 % of cases [2,3].

Clinical presentation varies from asymptomatic to general symptoms occurring in 15-30% of patients, such as fever, malaise and weight loss, and finally to local symptoms which depend on anatomical site of the tumour. Local symptoms in IMTs occurring in the abdominal-pelvic region include incidentally detected abdominal mass (most commonly), abdominal pain, gastrointestinal complaints but seldom intestinal obstruction, ascites and vomiting. A laboratory analysis may reveal an elevated Erythrocyte Sedimentation Rate (ESR), microcytic anaemia, polyclonal hyperglobulinemia and/or thrombocytosis [2,7].

Radiological descriptions of IMTs are rare, variable and nonspecific as well as limited to case reports or small series.8,9 IMTs are usually discovered using Ultrasound (US), Computed Tomography (CT) or Magnetic Resonance Imaging (MRI), especially in younger people to avoid CT related radiation. These lesions are usually presented on CT scans as solid, well- or ill-defined masses, which are hypo- or isoattenuating to muscles. 4,10 On contrast-enhanced CT there are variable amounts of delayed enhancement due to potential fibrotic tissue. MR imaging features are less commonly described and include hypo- or hyperintense T2-w signal and hypointense T1-w signal with heterogeneous gadolinium contrast enhancement due to the presence of some degree of fibrotic tissue in IMTs [4,8].

Since these radiological features are not specific, the final diagnosis is based on histopathology. Macroscopically, omental IMTs have a malignant appearance, are highly vascular, and may be adherent to surrounding structures. Microscopically, they are composed of interlacing fascicles of myofibroblastic slightly atypical spindle cells intermingled with chronic inflammatory stroma of lymphocytes or histiocytes or plasma cells with little mitotic activity and pleomorphism [11]. On immunohistochemistry, strong diffuse cytoplasmic reactivity to vimentin is typical for almost all IMTs, while reactivity to Muscle-Specific Actin (SMA) varies from a focal to a diffuse pattern in the spindle cell cytoplasm. IMTs show no reactivity to myogenin, myoglobin, S100 protein, or CD117. In addition, TP53 immunoreactivity is rare, and it has been associated with tumour recurrence and malignant transformation. A typical feature of IMTs is a characteristic anaplastic lymphoma kinase (ALK) tyrosine kinase activation and ALK protein overexpression that is present in about 60% of the cases. Coffin et al. discovered that ALK-positive IMTs occurred in younger patients with a tendency for recurrence, while the absence of ALK was likely to be associated with elderly and metastasizing. [5] On the other hand, Telugu et al. did not find statistically significant correlation between ALK-1 expression, recurrence and metastasis [12].

Surgical resection of IMT is the treatment of choice with a regular follow-up after surgery [4,5]. In cases of unresectable IMT cases of successful adjuvant radiotherapy or adjuvant chemotherapy are reported, although controversive conservative therapy with antibiotics, steroids, NSAIDs or observation along with intense follow-up have been also proposed in consideration [12,13]. Precise radical removal with adequate margins has great prognostic significance and avoids recurrence. In case of recurrent disease or metastasizing, a re-excision or metastasectomy ought to be performed. Possible treatment also includes radiotherapy, chemotherapy, steroids and nonsteroidal anti-inflammatory drugs [5].

We report a case of MR manifestations of torqued pedunculated GO-IMT in a young girl presenting as gynaecological emergency with a thorough review of previous literature on GO-IMTs to summarize the clinical, imaging and pathohistological features of GO-IMTs.

In the literature, a total of 23 patients with GO-IMTs were identified (Table 1) [7,10,12-27]. Ten of the patients were female (44%) and thirteen were male (56%). Age of the patients varied from a newborn to 75 years of age, with the average age of 18.4. Where data was available, the most common location of the GO-IMT was in the pelvis (six patients; 37 %), and the second most common was lower abdomen (right or both lower quadrants) (five patients; 31%) while in the remaining patients GO-IMTs were growing inside the whole abdomen (four patients; 25%) or in the upper abdomen (three patients; 19%). The majority of the patients were administered due to pain or discomfort (ten patients; 63 %), mostly with the longer course of the disease, only two patients (13 %) presented with acute pain, mimicking acute appendicitis. In six patients (38 %), a painless mass or a lump was palpated and seven patients (43%) presented with general symptoms. None of the patients was asymptomatic. Among the cases where laboratory data was given, five patients (42 %) exhibited normal laboratory values, two (17%) had elevated ESR, three (25%) had elevated leukocytes and four patients (33%) had high CRP. Diagnostic pathway included multimodal imaging approach in approximately half of the cases; in fact, transabdominal US was followed by panoramic imaging in eight cases (53%), seven using CT and one MRI. As the first imaging modality, a CT scan was used in 12 patients (80%) and an MR imaging in two patients (13%). In one case none of the diagnostic imaging tools was used due to specific clinical presentation (mimicking acute appendicitis) which led directly to diagnostic laparoscopy. 16 patients (84%) had one tumour, three (16%) had multiple tumours. The average size of the tumours was 9.7 cm (ranging from 0.4 to 26.0 cm). Surgery, either laparoscopic or open laparotomy was the method of treatment in all 19 patients. One patient (5%) received preoperative chemotherapy, three patients (16%) received adjuvant chemotherapy and one patient (5%) adjuvant radiotherapy. Where applicable, immunohistochemistry was positive for ALK in 14 patients (88 %) and negative in two (12%).

Among all cases, only three GO-IMT in the literature were reported to have a pedicle. In addition, after further careful search of the literature, only one case of a torqued pedunculated omental IMT with subsequent infarction was found. This GO-IMT in a 20-year old girl was located in the right lower abdomen and was mimicking acute appendicitis with no diagnostic imaging being performed before the surgery [13]. A case of ischemic pedunculated myxoid hamartoma of the ileum due to torsion in a 6-year old boy mimicking acute appendicitis has been described. However, it is still not clear whether omental-mesenteric myxoid hamartoma represents a variant of IMT. Secondly, the origin of this mass was ileal mesentery [14].

At MRI, the torqued GO-IMT in our patient was initially misdiagnosed as torqued pedunculated subserous leiomyoma for its close proximity to the uterus and a visible pedicle originating from the anterior part of the mass leading toward the uterus. However, careful retrograde analysis of pelvic MR images revealed that the pedicle actually led to a partly shown, T2w hyperintense structure in the lower right quadrant (probably representing edematous greater omentum) and not toward the uterus (Figure 6). According to the literature, leiomyomas tend to be extremely rare in adolescence (< 1%).15 Secondly, leiomyomas that have a pedicle are rare and torsion of subserosal leiomyoma is even a rarer condition with the diagnosis seldom established preoperatively [16]. In addition, Extragastrointestinal Stromal Tumours (EGIST), either originating from mesentery and omentum or less commonly, as free masses in the pelvic cavity can be considered among differential diagnoses. Whereas radiologic imaging findings are similar to imaging features seen in IMTs (well- defined margins, heterogeneous enhancement, exophytic growth, a size of > 5 cm and a necrotic centre), EGIST tumours are rare in young adults and are negative for desmin, keratin and ALK on immunohistochemistry staining [17,18].

Other differential diagnosis includes abscesses, tuberculous granulomas, soft tissue sarcomas, lymphomas, calcifying fibrous tumour and metastasis [7].

As already mentioned, during laparoscopic surgery, a pelvic mass that was twisted around its stalk, originating from the omentum was removed and sent to the Pathological department. Due to equivocal positivity of ALK, a second opinion from another well-qualified pathology department revealed the diagnosis of Inflammatory Myofibroblastic Tumour (IMT). As previously said, surgery is the treatment of choice for IMTs and a regular follow-up after surgery is needed due to possible aggressive course of the disease [4,5]. Our patient has regular follow-up appointments every three months with her chosen gynaecologist who performs ultrasound examination. Up to this day, 14 months after being diagnosed with IMT, there are no signs of recurrence.

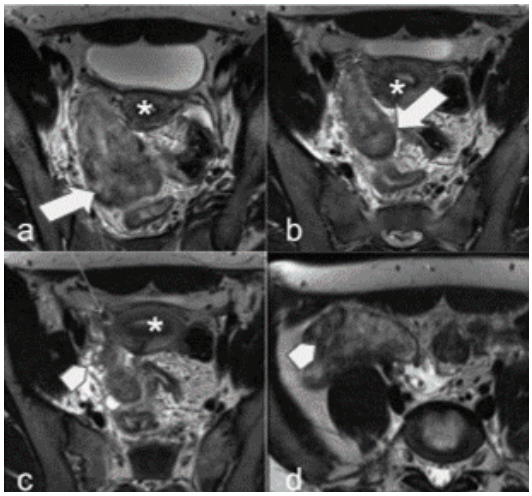

Figure 6: Careful retrograde analysis of T2 weighted pelvic magnetic resonance (MR) images in axial plane revealed (a and b) a pedicle (thin arrow in b) originating from the anterior part of the mass (wide arrow). (c) Pedicle (thin arrow) was leading toward the uterus (*), however no clear connection to it was found. The right ovary (arrowhead) appeared normal and was clearly separated from the mass.(d) Consequently, the pedicle ended in a large, well-defined hyperintense in-homogenous mass (arrowhead) that probably represented edematous greater omentum.

Table 1: Demographic, clinical, imaging and histopathological features of 23 cases of GO-IMTs reported in the literature

Author	Sex/age (years)	Location	Symptoms/signs	Inflammatory markers	Max diameter (cm)	Dg imaging	Treatment	Pedicle	IHC staining
Koltuksuz et al.[14]	F/14	RUQ	abdominal pain	normal (no values)	2.0 and 4.0 (2 masses)	US, CT	LapSE	no	N/A
Hagenstad et al.[15]	M/18	RLQ	clinical signs of acute appendicitis	N/A	N/A	CT	LapSE+ appendectomy + ACTH	N/A	ALK+
Ma et al.[16]	M/ 7 Mo	whole abdomen	abdominal distention	N/A	0.4 - 11.0 (multiple omental nodules)	N/A	LapSE	N/A	ALK+
Gupta et al.[17]	M/6	upper abdomen	abdominal pain, low fever, constipation	L 24.0× 10^9/L, others normal (no values)	5.5	US, CT	LapSE	no	
Kim et al.[18]	F/3	N/A	abdominal pain	normal (no values)	15	US, CT	LapSE	yes	N/A
Sodhi et al.[19]	M/2	whole abdomen	abdominal mass, intermittent low fever	ESR 95 mm, others not given	15.,0	CT	LapSE	no	N/A
Bertocchini et al.[12]	M/10	whole abdomen	weight loss, progressive ascites	elevated CRP (no values)	huge nodular mass, diffusely infiltrating GO	US, CT	LapSE + ACTH	no	N/A
Singhal et al.[20]	F/15	lower abdomen	abdominal pain, intermittent fever, weight loss	normal (no values)	7.2	US, CT	LapSE	no	ALK+,
Aptel et al.[10]	F/20	pelvis	impaired general condition	normal (no values)	7	CT	LapSE	no	ALK+
Backhaus et al.[21]	M/1,5	whole abdomen	painless enlarging abdominal mass	CRP 8.3 mg/L; L normal (no values)	16	US, MR	NACTH + LapSE	no	ALK+
Deepu et al.[22]	M/40	right iliac fossa	abdominal pain, low fever, abdominal mass	not given	6	US, CT	LapSE	no	N/A
Kye et al.[23]	F/22	pelvis	painless, abdominal mass	normal (no values)	6.5	CT	LapSE	yes	ALK+
Antonescu et al.[24]	F/NB	N/A	N/A	N/A	N/A	N/A	N/A	N/A	ALK+
Antonescu et al. (suppl. data)[24]	M/5	N/A	N/A	N/A	N/A	N/A	N/A	N/A	ALK+
	F/21	N/A	N/A	N/A	N/A	N/A	N/A	N/A	ALK+
Cianci et al.[25]	M/75	upper abdomen	abdominal pain, abdominal mass	ESR >20 mm	26	US, CT	LapSE	N/A	ALK-
El Hage Chehade et al.[7]	M/38	lower abdomen	abdominal pain, anorexia, nausea	L 11.8 × 10^9/L, CRP 121 mg/L	10	MR	LapSE	no	ALK+
Pennington et al.[13]	F/20	RLQ, pelvis	clinical signs of acute appendicitis	L 11.3× 10^9/L, CRP 81.2 mg/L	N/A (multiple omental nodules)	none	LapSE (dg LPS)	yes	N/A
Yamamoto et al.[26]	F/22	N/A	N/A	N/A	N/A	N/A	N/A	N/A	ALK+
Liang et al. [27]	F/25	pelvis	abdominal pain	N/A	7	CT	LPS	no	ALK+
Telugu et al.[12]	M/10	N/A	N/A	N/A	7	N/A	LapSE	N/A	ALK+

	M/41	N/A	N/A	N/A	8.5	N/A	LapSE + RTH	N/A	ALK-
	M/13	N/A	N/A	N/A	13	N/A	LapSE + ACTH	N/A	ALK+

Legend: abd (=abdominal), ACTH (=adjuvant chemotherapy), CRP (=C-reactive protein), CT (=computed tomography), CTH (=chemotherapy), NACTH (=neoadjuvant chemotherapy), ACTH(=adjuvant chemotherapy), Dg (=diagnostic), ESR (=erythrocyte sedimentation rate), F (=female), GO (=great omentum), IHC (=immunohistochemical), L (=leukocytes), LapSE (=laparatomic surgical excision), LPS (=laparoscopy), M (=male), Max (=maximum), Mo (=months), MRI (=magnetic resonance imaging), N/A (=not available), NB (=newborn), RLQ (=right lower quadrant), RTH (=radiotherapy), RUQ (=right upper quadrant), LapSE (=laparotomic surgical excision), US (=ultrasound), + (=positive), - (=negative).

5. Conclusion

We have described a case of acute pelvic pain because of a torqued Inflammatory Myofibroblastic Tumour (IMT) arising from the Greater Omentum (GO), firstly thought to be torqued pedunculated subserous leiomyoma uteri, in a previously healthy young female, which was successfully laparoscopically removed.

Rarity of this pathology, with torsion of pedunculated pelvic GO-IMT being even rarer, represents a diagnostic challenge for clinicians as well as radiologists and pathologists. Awareness of this entity may lead to faster decision making and avoidance of multiple transfers between departments. In addition, the possibility of more aggressive forms of this disease, the potential recurrence and/or metastasizing prompts the patients for regular follow ups.

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