Heterotopic intraabdominal ossification: report of a case and review of the literature

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Ossificação mesentérica heterotópica: relato de caso e revisão da literatura

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key words

Myositis ossificans

Soft tissue neoplasms

Ossification heterotopic

abstract

There are 28 unequivocal reports of heterotopic mesenteric ossification (HMO) in the medical literature. Most cases are poorly defined lesions in intra-abdominal structures that cause intestinal obstruction. A small well-delineated solid mass was reported in only one patient with no previous history of trauma. We report herein the case of a 67 year-old female patient with calcified mass in the left adnexal region. The awareness of HMO may avoid an erroneous diagnosis of extraskeletal osteosarcoma. This case differs from most cases of HMO as it is the third one reported in females and does not present a diffuse involvement, which leads to obstructive symptoms.

unitermos

Miosite ossificante

Neoplasias de tecidos moles

Ossificação heterotópica

resumo

A literatura médica apresenta 28 relatos inequívocos de ossificação mesentérica heterotópica (OMH). A maioria apresenta-se como lesões mal definidas em estruturas intra-abdominais causando obstrução intestinal. Em apenas um caso a lesão apresentava-se como uma pequena massa bem delimitada, num paciente sem histórico de trauma. Apresentamos o caso de uma paciente de 67 anos com massa calcificada em topografia anexial esquerda. O conhecimento da entidade OMH previne eventual diagnóstico errôneo de osteossarcoma extraesquelético. Esse caso difere da maioria dos casos de OMH visto que é apenas o terceiro relatado no sexo feminino, e não apresentava envolvimento difuso levando aos sintomas obstrutivos.

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Introduction

Heterotopic ossification (HO) is a metaplasic process that usually follows trauma and infections. The most familiar synonym of this process in somatic soft tissues is myositis ossificans. In 1999, Wilson *et al.* suggested the designation heterotopic mesenteric ossification (HMO) for the five cases of intraabdominal lesions reported by them[11], in addition to two other previous reports[7, 12]. These lesions share with classical soft tissue myositis ossificans the characteristics of frequent association with trauma, rapid but predictable course, an mesenchymal proliferation with zoning phenomenon (ordely pattern of bone formartion)[11]. Importantly, myositis ossificans is a misnomer since this process is not confined to muscles and usually exhibit little inflammation[13].

In English literature, there are 28 reports of HMO ("intraabdominal myositis ossificans" [IMO]) meeting the criteria proposed by Wilson *et al.*[11]. Most of these cases present a diffuse reaction or poorly defined lesions in intraabdominal structures (18 of 19 cases), usually involving the mesentery (19 of 28 cases), and induce a high frequency of intestinal obstruction (14 of 20 cases). Small well-delineated solid masses are rare and indeed reported in only one case with no previous history of trauma[11]. We reported herein a rare case of HMO/IMO presenting an isolated pelvic mass in a female patient with long term history of pelvic surgeries (cesarean operations).

Case report

A 67 year old female patient sought the surgery service because of an incidental finding of calcified mass in the left adnexal region. She had a past history of urolithiasis diagnosed two years before when an abdominal radiograph suggested the presence of calcified leiomyomas. By that time, a double-J ureteral catheter was placed in the left side. After a year and a half, she started complaining of dysuria, urinary incontinence and nocturia. A pelvis radiograph performed six months before the current admission was also indicative of calcified leiomyomas (**Figure 1**). A magnetic resonance (MR)was performed two months before admission, revealing a heterogeneous mass in left adnexal region suggestive of an ovarian tumor. The tumor had a bilobated appearance with a densely calcified posterior lobe and an anterior lobe with lower density (**Figure 2**). Serum tests of tumor markers one month before admission indicated non elevated levels of CA-125, human chorionic gonadotropin, α-fetoprotein, carcinoembryonic antigen and lactate dehydrogenase.

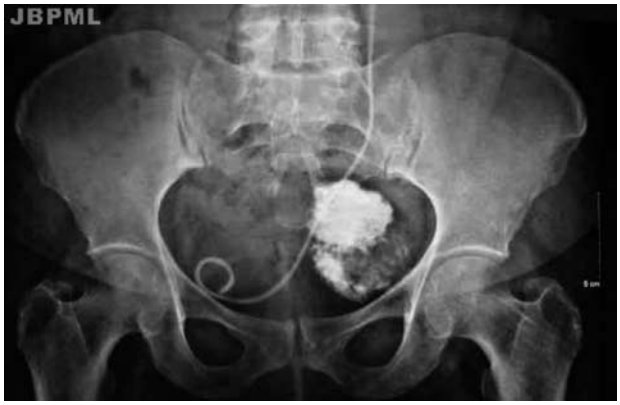

Figure 1 – *Radiograph of the pelvis. A calcified mass with ovoid shape in left pelvis*

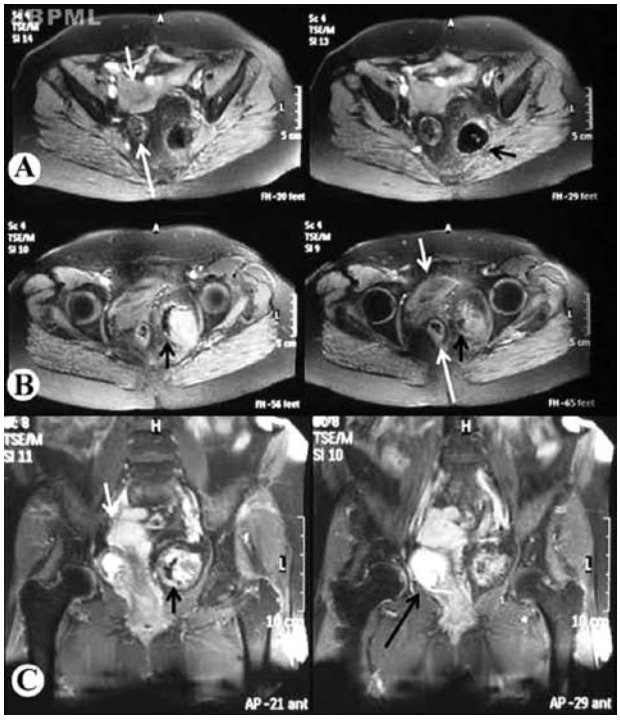

Figure 2 – *Magnetic resonance imaging. (A) Fat-suppressed T1-weighted axial images: superior sections show the posterior tumoral lobe with a densely calcified appearance (black arrow). (B) Fat-suppressed T1-weighted axial images: inferior sections show the anterior tumoral lobe with lower density. The black arrow indicates the dense calcification observed only in the periphery. In both A and B figures the short white arrows indicate the uterus and the long white arrows indicate the rectum. Note that the posterior tumoral lobe in "A" has a density even higher than the femur heads in "B". (C) Gadolinium-enhanced fat-suppressed T1-weighted coronal images: the short black arrow indicates again the densely calcified periphery of the anterior tumoral lobe. The short white arrow indicates the uterus and the long black arrow indicates the bladder, highlighted by the contrast*

The patient had a long term history of systemic arterial hypertension and dyslipemia. Her obstetric history includes 10 pregnancies and ten deliveries (with two cesarean operations). The last surgical obstetric delivery occurred 35

years ago. She also reported a previous surgical procedure for uterine leiomyoma removal 10 years ago.

At surgery, the tumor was unrelated to left ovary and uterine tube which exhibited normal appearance. The tumor emerged from the retro-rectal adipose tissue and broad ligament. At macroscopy, it was a 9 x 4.5 x 4.5 cm well delineated tumor with bilobated appearance. The external surface had a yellowish appearance. At cut surface, one lobe had a 2 cm cavity with yellowish serous content. The cut surface had a soft grayish glistening center while the periphery appeared grayish-white, firm and densely calcified. Microscopically, the tumor was mainly composed by hypercellular population of immature stromal cells. The classic zonation phenomenon described in myositis ossificans was easily observed with orderly progression of osteoid formation to mature bone from the center to tumor periphery (**Figure 3**). Stromal cells including osteoblasts lacked cytologic features of malignancy. Chronic inflammation was minimal. Foreing body or granulomas were not observed.

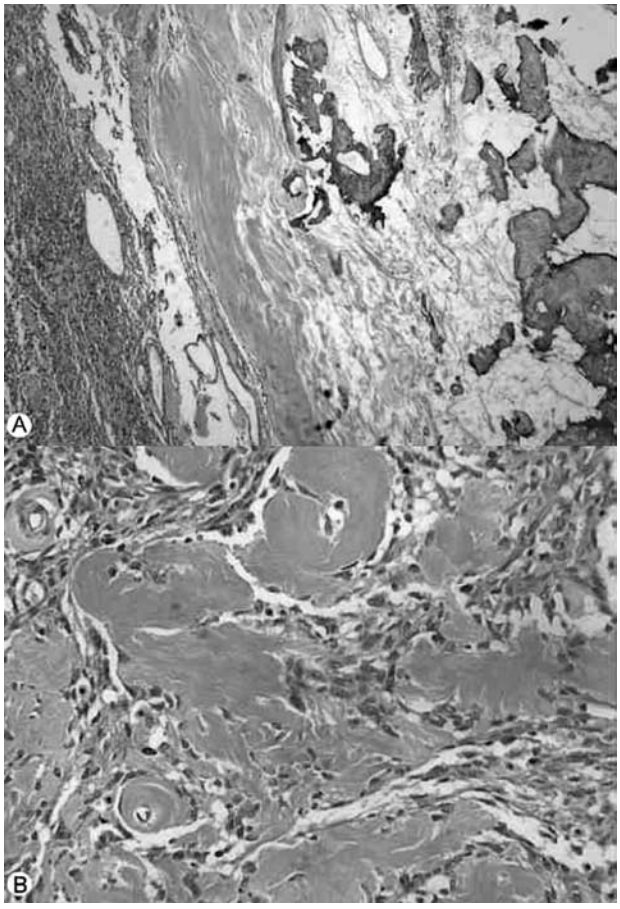

Figure 3 – *Microscopy appearance. (A) Classic zonation phenomenon described in myositis ossificans was easily observed with orderly progression of osteoid formation to mature bone from the center to tumor periphery (hematoxylin & eosin, 200x magnification); (B) Hypercellular population of immature stromal cells lacking cytologic atypia (hematoxylin & eosin, 400x magnification)*

The patient reported relief of the urinary symptoms three months after surgery. No recurrence was detected during a 12-month period of follow-up.

Discussion

Awareness on the existence of HMO/IMO is important to avoid an erroneous diagnosis of extraskeletal osteosarcoma, which is its most important morphologic mimic[11, 13]. Typical zonation of stromal cell proliferation, immature osteoid and mature bone formation, and lack of cytologic atypia are important clues for the identification of HMO/IMO[9]. In addition, the rare well differentiated extraskeletal osteosarcoma will most probably exhibit a reverse zonation effect: immature woven bone or osteoid in the tumor periphery while mature osseous trabeculae are centrally located[4].

Alertness to HMO/IMO increased since 1999 when Wilson *et al.* reported five similar cases and reviewed previous reports. The authors found only two similar cases in previous reports[7, 12] and suggested that this entity should not be confounded with previous reported of well-documented ossificafion of laparotomy scars or heterotopic bone formation in metastatic colonic carcinoma[11]. Since them, 21 new cases have been reported in English literature.

HO is a nonspecific term used to describe bone formation in tissues that normally do not ossify. It occurs in soft tissue locations associated with trauma, burns, prolonged immobilization, prior surgery, and neoplasia. HO is distinct from dystrophic calcification (DC), which refers to the deposition of amorphous calcium salts in the absence of osteoblastic activity. The origin of HO is not known, although it has been postulated to be due to osteoblastic metaplasia of multipotent mesenchymal cells in response to trauma, or occasionally by traumatic or surgical implantation of bone or periosteum into the soft tissues[8].

Table 1 summarizes the available information of all cases of HMO/IMO in English literature. The mean ± SD age at the diagnosis is 53.8 ± 18.7 years. The process is strongly associated with male gender with 26 reports to date from men and only two, similar to our case, occurring in women. These features suggest an even higher male predominance when compared to the soft tissue counterpart (which is 3:2) and a concentration of cases at an older age group (mean age of soft tissue myositis ossificans is 32 years)[9].

Most cases of IMO present a small bowel obstruction and only three among 28 cases were incidental findings, mirroring the present case. A history of trauma or previous surgery was positive in 18 of 27 cases (67% of all). Only one case is described as a well defined lesion such as in the present case, and this case had no known history of trauma[11]. In the present case, the long period between pelvic surgeries and the incidental detection of the lesion due to imaging tests may have been responsible for the organization of this reactive process that seems to be diffuse soon after triggering traumatic events. Unfortunately, we could not locate the previous material from the reported myomectomy 10 years ago to verify if the same process could be already present at that moment.

It is common to find hemorrhage or gelatinous material in the center of soft tissue myositis ossificans, and some cases may indeed present cystic changes with yellowish clear fluid paralleling the observation of the present case[4]. In contrast to a single report that described elevated serum levels of CA-125 associated with a HMO/IMO tumor with similar dimensions (9 x 3.5 cm)[2], we did not observe abnormal levels of that and other tumor markers.

In conclusion, we report a rare case of HMO/IMO involving pelvis and leading to differential diagnosis with leiomyomas and ovarian tumors. Awareness on this reactive process will prevent pathologist to misdiagnosis such lesions as sarcomatous process, such as extraskeletal osteosarcoma. This case is clearly different from most cases of HMO/IMO since is the third reported in a female patient and lacks the diffuse mesenteric involvement leading to obstructive symptoms. In addition, past trauma/surgical events are too distant to be obviously implicated in tumor emergence. Alternatively, such an insidious course could lead to the organization of this reactive process into a silent isolated pelvic mass.

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This work was approved by ethics committee of Hospital Espanhol and the patient gave written consent for publication of this case report (September 22, 2008 – Register Number 026/2008).

Table 1 Previous reports of heterotopic mesenteric ossification in English literature

Author, year	Gender	Age at diagnosis	Presentation	Location	Intraoperative findings	Trauma history
Lemershev, 1983[7]	Male	44	Small bowel obstruction	Mesentery	Diffuse enterocutaneous fistula	Laparotomies
Yannopoulos, 1992[12]	Male	63	Intestinal obstruction	Mesentery	Diffuse	Aortic bifemoral bypass
Wilson, 1999[11]	Male	75	Small bowel obstruction	Mesentery	Diffuse cementing loops	Repair of aortic aneurysm
Wilson, 1999[11]	Male	76	Small bowel obstruction	Mesentery	Diffuse cementing loops	Left colectomy repair of aortic aneurysm
Wilson, 1999[11]	Male	43	Small bowel obstruction	Mesentery	Diffuse satellite nodules	None
Wilson, 1999[11]	Male	80	Incidental	Mesentery	2 cm mass	None
Wilson, 1999[11]	Male	43	Small bowel obstruction	Mesentery	Poorly defined mass	Repaired incarcerated umbelical hernia
Hakim, 2001[5]	Male	50	Enterocutaneous fistulae	Fistulae Mesentery	Diffuse	Stab wound nephrectomy, left colectomy
Bovo, 2004[2]	Male	76	Small bowel obstruction	Mesentery	Poorly defined mass satellite nodules	None

Table 1 (continued)

Androulaki, 2005[1]	Male	74	Intestinal obstruction	Mesentery	Diffuse cementing loops	Surgical reconstruction of umbelical hernia
Kao, 2005[6]	Male	60	Incidental	Mesentery	Diffuse	Colonic diverticulitis hartmanns operation
Tonino, 2005[10]	Male	39	Incidental	Mesentery	Diffuse	Laparotomy for gunshot injury, relaparotomies to close fistulae
Zamolyi, 2006 [13]	Male	43		Omentum		
Zamolyi, 2006[13]	Female	32		Omentum		
Zamolyi, 2006[13]	Male	37		Mesentery		
Zamolyi, 2006[13]	Male	24		Mesentery		
Zamolyi, 2006[13]	Male	74	(not reported)	Left psoas muscle	(not reported)	5/9 in this series had history of trauma or surgery
Zamolyi, 2006[13]	Male	47		Colon		
Zamolyi, 2006[13]	Male	36		Right obturator muscle		
Zamolyi, 2006[4]	Male	68		Mesoappendix		
Zamolyi, 2006[4]	Female	76		Peritoneal surface		
Patel, 2006[12]	Male	51	Small bowel obstruction	Omentum, Mesentery		
Patel, 2006[12]	Male	21	Peritonitis	Omentum		
Patel, 2006[12]	Male	65	Small bowel obstruction	Mesentery	Described as poorly defined masses from 3.5 cm to 15 cm.	4/6 in this series had history of trauma or surgery
Patel, 2006[12]	Male	62	Small bowel obstruction	Mesentery		
Patel, 2006[12]	Male	22	Small bowel obstruction	Mesentery		
Patel, 2006[12]	Male	72	Small bowel obstruction	Mesentery		
Como, 2008[13]	Male	51	Enteric fistula	Mesentery	Diffuse	Multiple gunshot wounds

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