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Heterotopic mesenteric and abdominal wall ossification – Two case reports in one institution

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ABSTRACT

INTRODUCTION: Heterotopic ossification occurs when bone develops in tissues which usually don't undergo ossification. Heterotopic mesenteric ossification, also known as intra-abdominal myositis ossificans, is a rare and benign form of ossification, usually related with previous abdominal surgery or trauma.
PRESENTATION OF CASES: We report two cases of heterotopic ossification both after multiple abdominal surgeries, with intraoperative findings of mesenteric and abdominal wall ossification. Histopathology revealed metaplastic bone deposition, without evidence of atypia or malignancy.
DISCUSSION: This rare entity shares clinical and pathological characteristics of myositis ossificans. It is important to consider the differential diagnosis with sarcomas. In the cases described, the patients were proposed for elective surgery and this pathology was as an incidental finding.
CONCLUSION: The simultaneous presence of mesenteric and abdominal wall ossification in both patients makes these cases even rarer.

1. Introduction

Heterotopic ossification (HO) is related with abnormal bone formation in tissues which usually don't undergo ossification. It was first described in 1883 by Riedel, as a complication after injury of the spinal cord [1].

The heterotopic mesenteric ossification (HMO), also known as ossificant myositis, is a rare and benign form of HO. It is characterized by the formation of an ossificant pseudotumor in the mesenteric base, generally after abdominal trauma (surgical or other). Lemershev et al. and Hansen et al. made the first report of heterotopic ossification of the intestinal mesentery in 1983, but it was Wilson et al. in 1999 who named this condition [2–4].

Regarding the ossification of the abdominal scar, this was first reported in 1901 by Askanazy. It is more common than HMO, considering the large volume of abdominal surgeries performed every year worldwide [5].

The incidence of HO is not clear, given the rarity and possible underreporting of these findings. Although many causes have been proposed, the aetiology of this pathology is not consensual [1,6].

We report two cases of combined heterotopic ossification of mesentery and abdominal wall.

2. Case report 1

We report a case of a 45 years-old male, previously healthy, that presented to the emergency room with an abdominal pain, fixed and severe, that didn't alleviate with classic analgesics. He was submitted to multiple exams, with the diagnosis of a superior mesenteric ischemia. The patient underwent urgent surgery with segmental enterectomy and temporary ileostomy. He was admitted to the Intensive Care Unit (ICU) in the postoperative period, needing multiple surgeries with small bowel resection. The patient was discharged home in the 86th postoperative day (POD).

Ileostomy reversal was scheduled 8 weeks later. Preoperatively the patient was assessed with an analytic panel and computed tomography (CT) that showed calcifications with linear morphology of the adipose and muscular planes near the ostomy, which prolonged to the mesenteric fat and peritoneal peritoneum in the middle line (Fig. 1).

Intraoperatively we found bone formations in the abdominal wall above the scar, as well as within the mesentery, compatible with the CT scan findings (Figs. 1 and 2). These bone formations were carefully resected, because they involve the ileostomy and also to prevent injury to the small bowel, since there were spiculated bones.

Histopathology revealed metaplastic bone deposition with mature bone trabeculae, fibrous and adipose tissue, with no evidence of atypia or malignancy.

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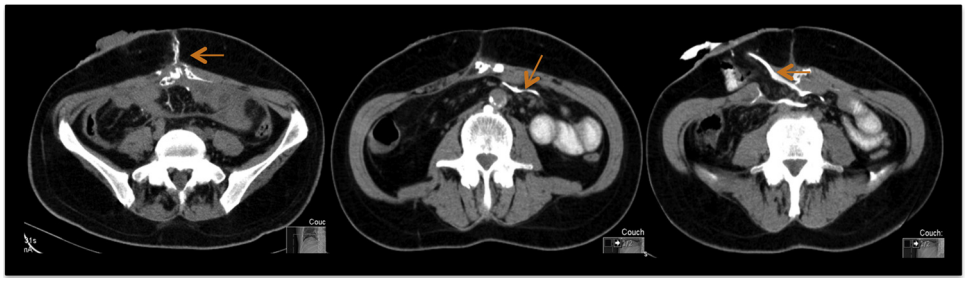

Fig. 1. Abdominal Computed Tomography scan showing calcification of the abdominal midline scar, mesentery and fat surrounding the ileostomy. Linear branching opacities (arrows).

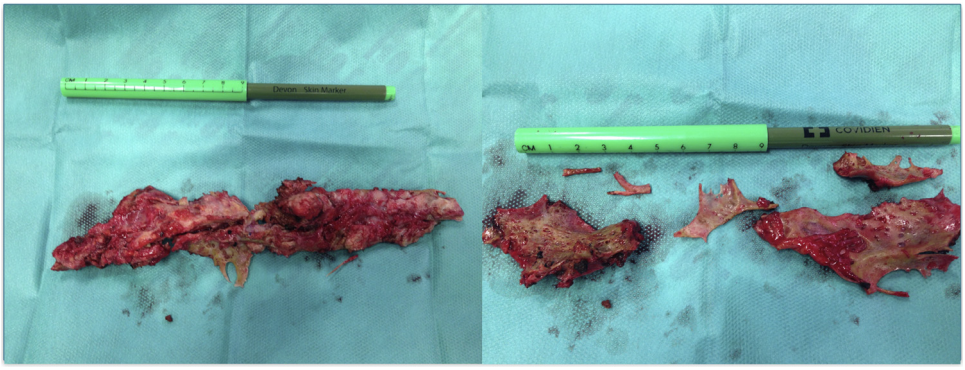

Fig. 2. Excised well-formed bone fragments from the midline scar (left image) and from the mesentery (right image).

He was discharged in the 15th POD. We maintain follow-up as an outpatient, with no complications after 20 months.

3. Case report 2

We report the case of a 60 years-old male, submitted to radical right colectomy for a malignant condition, complicated with anastomotic dehiscence and septic shock, which led to anastomosis take down and vacuum pack laparostomy. Abdominal wall closure was achieved primarily after 2 revisions with a terminal ileostomy and a mucous fistula of colon.

Eighteen months after, the patient was proposed for ileostomy reversal.

Intra-operatively we found bone formations in the abdominal wall and also in the mesentery (Fig. 3), like the previous case. The bone formations were also resected, to prevent future complications, like bowel obstruction or perforation.

The patient's postoperative course was uneventful, and he was discharged free of symptoms.

Histopathology revealed also metaplastic bone deposition, without evidence of atypia or malignancy.

He was discharged in the 13th POD, with no complications after 18 months.

4. Discussion

HO is distinct from dystrophic calcification (DC), which refers to the deposition of amorphous calcium salts in the absence of osteoblastic activity, unlike the organized calcium deposition comprised of osteoblasts and structured bone layers observed in HO [5,20].

There are three distinct types of HO described: myositis ossificans circumscripta, myositis ossificans progressiva and traumatic myositis ossificans, the last one being present in our patients [5].

Traumatic myositis ossificans is related with the heterotopic ossification of extra-skeletal tissue following a traumatic stimulus [7]. It usually occurs adjacent to skeletal tissue, but there are reports of traumatic myositis ossificans happening in the abdomen, generally in patients subjected to abdominal trauma and/or abdominal surgery. In these cases, the formation of an ossifying pseudotumor occurs at the base of the mesentery, known as heterotopic mesenteric ossification (HMO) [8].

According to the literature, there are four main factors that probably have an influence on the pathogenesis of ectopic bone formation [6].

First, there must be an inciting event, like a trigger, that in our two cases was abdominal surgery. Second, after this trigger, a signaling vehicle must emanate from the site of the injury, this being the most important step. However we do not know for sure which are the implicated agents [7,19]. Third, it should exist a supply of mesenchymal cells that have the potential to differentiate into many different cell types, including chondroblasts or osteoblasts, if the appropriate signal emanates from the site of the injury. Lastly, a permissive environment is necessary for this process to occur [7].

It's important to be aware of the existence of HMO, because it is crucial to distinguish between this entity and extraskeletal osteosarcoma. This awareness has increased since 1999, when Wilson et al. reported five similar cases [9].

Classic histopathologic features of HMO include nodules of dense fibrous tissue within the mesentery mostly composed of trabeculae of well-formed lamellar bone trabeculae with dystrophic calcification [13]. The osteoblastic cells show usually no evidence of atypia, necrosis or increase in mitotic activity. HMO can sometimes exhibit a number of features that can raise concerns for a malignant process including high cellularity, mitotic figures, and a "lace-like" osteoid production [13,14]. The most important histological feature of HMO is the zone formation. This refers to the progressive maturation from the central, inner immature zone consisting of hypercellular reactive fibrous tissue that can contain atypia and mitoses, to the middle zone of organized osteoid, to finally the

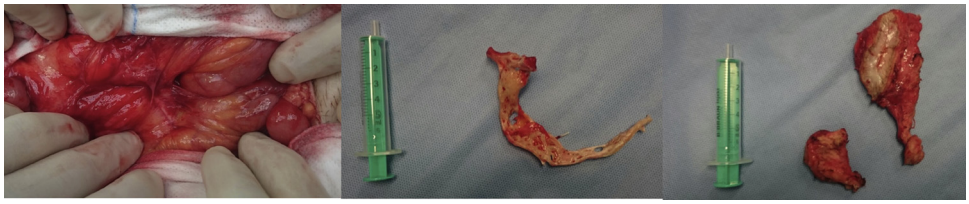

Fig. 3. Bone formations of the mesentery (arrow and central picture) and of the abdominal wall (right picture).

peripheral zone of the lesion consisting of well-formed mature lamellar bone [8,15–17]. Thus, it is important to make differential diagnosis with sarcomas, which exhibit their most aggressive histopathologic changes at the periphery of the lesion [13].

The pathophysiology of HO is not well-established, although it has been postulated to be due to osteoblastic metaplasia of multipotent mesenchymal cells in response to severe inflammatory stimulus due to trauma, or occasionally it can be due to traumatic or surgical implantation of bone or periosteum into the soft tissues [1,9,18,19]. In our two cases, HMO and abdominal wall ossification probably occurred due to repetitive surgical injuries, with maintained inflammatory stimulus until bone formation. We do not know the exact time of bone formation since the stimulus was applied, but generally those lesions grow and develop within weeks or even days after surgery.

To date there are approximately fifty cases of HMO described in the literature.

This entity has a strong male preponderance, with ages ranging from 22 to 80 years old, mostly middle-aged individuals, and one case described in a child [10,11]. In nearly all of the cases described, abdominal surgery or trauma preceded HMO, and the patients were symptomatic, usually with small bowel obstruction (nausea, vomiting, abdominal pain and distension). Diagnosis is usually made intraoperatively, but CT scans can also provide some evidence [12]. In our cases, the patients were both males, but asymptomatic, with the HMO incidentally found at CT (first case) or surgery (second case). Both were submitted to multiple surgeries, and the second case had also a generalized peritonitis, providing the permissive environment for ossification.

The prognosis of HO is generally good, with no evidence of malignant potential so far and treatment should be conservative whenever possible, avoiding surgery to prevent further ossification [5,20]. Cases with bowel obstruction in symptomatic patients usually require surgical intervention and a complete excision of bone formations should be made. There is some evidence in the literature suggesting prophylactic treatment with non-steroidal anti-inflammatory drugs, bisphosphonates or even radiotherapy after surgery to prevent recurrence in these patients, but these measures need scientific corroboration [20].

5. Conclusion

This is a very rare pathology, so it is difficult to reach consensus on the best approach for each case and clinicians are guided by the existing literature.

Awareness on this reactive process will prevent pathologist to misdiagnosis this lesions as sarcomatous.

These two cases were the first in our hospital and there are no other cases reported to date in Portugal.

Our patients were proposed for elective surgery for other reasons, and this pathology was found intraoperatively, otherwise they would not be proposed for surgery, since they were asymptomatic. The patients may have recurrences in the future, so they should be followed on an outpatient basis.

In both cases we observed the occurrence of mesenteric and abdominal wall ossification, which is an even rarer event than the occurrence of either one individually.

With this work, the authors intend to emphasize the importance of reporting these cases, to make clinicians more aware of this pathology and to allow better definition of etiology and therapeutic approach.

SCARE guidelines

This work has been reported in line with the SCARE criteria [21].

Ethical approval

Not submitted to ethical approval – Case report.

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Author contributions

Cátia Ferreira: study design, data collection and writing.
Artur Ribeiro, Herculano Moreira, Bruno Pinto: writing and revision.
Carina Gomes, Ana Melo, Nádia Tenreiro: data collection.
Paulo Avelar: revision of the manuscript.

Conflicts of interest

No conflict of interests to disclose.

Consent

Written informed consent was obtained from the patients for publication of this case report and accompanying images.

Guarantor

None.

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