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HETEROTOPIC MESENTERIC OSSIFICATION. A CASE REPORT

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Palabras clave: Cirugía General; Osificación Heterotópica; Huesos; Mesenterio; Osteoblastos.

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ABSTRACT

Introduction: Heterotopic ossification (HO) is a rare condition characterized by bone formation in tissues that do not normally ossify. Its occurrence in the soft tissues of the mesentery, known as heterotopic mesenteric ossification (HMO), is even rarer and is usually found incidentally as an intraoperative finding. Although the etiological and pathophysiological mechanisms leading to overstimulation of osteoblastic activity have not been clearly elucidated, some hypotheses suggest the involvement of mesenchymal osteoprogenitor stem cells differentiation into osteoblasts.
Clinical case: A 41-year-old man attended a tertiary care center in Bogotá (Colombia) for ileostomy closure, which had been performed a year earlier following acute perforated appendicitis. On admission, he underwent a contrasted computed tomography of the abdomen that showed a hyperdense lesion in the root of the mesentery suggestive of bone formation. During surgery, a bony multisegmental mass attached to the mesentery was found, which was resected and sent for histopathological study. The report of this study indicated that the mass was a benign soft tissue with mature bone trabeculae. The patient was discharged 5 days after the procedure with satisfactory outcomes and resolution of his condition.
Conclusion: HMO is often a diagnostic challenge for surgeons, as it is usually found incidentally in the operating room. Therefore, it is recommended to maintain a high index of suspicion in patients who have a surgical history and atypical abdominal symptoms.

RESUMEN

Introducción. La osificación heterotópica (OH) es una condición benigna que consiste en la formación de hueso en tejidos que normalmente no se osifican. La osificación heterotópica del mesenterio (OHM) es una forma rara de OH que normalmente se descubre de forma incidental como un hallazgo intraoperatorio. Su patogénesis no se conoce del todo, pero hay hipótesis que apuntan a la estimulación de células madre osteoprogenitoras mesenquimales para diferenciarse entre los osteoblastos. Se presenta el caso de un paciente con OHM secundaria a un procedimiento quirúrgico.
Presentación del caso. Hombre de 41 años que asistió a una institución de salud de tercer nivel de atención de Bogotá (Colombia) para cierre de ileostomía, la cual se le había realizado un año antes cuando presentó una apendicitis aguda perforada. Al ingreso se realizó una tomografía computarizada de abdomen contrastada que mostró imagen hiperdensa en la raíz del mesenterio sugestiva de formación de hueso. Durante la cirugía se halló una masa multisegmentaria ósea adherida al mesenterio, la cual fue resecada y enviada a estudio histopatológico. El reporte

de este estudio indicó que la masa correspondía a tejidos blandos benignos con trabéculas óseas maduras. El paciente fue dado de alta 5 días después del procedimiento con evolución satisfactoria y resolución de su patología. **Conclusión.** La OHM suele ser un reto diagnóstico para el cirujano debido a que su hallazgo se da principalmente de forma incidental en el quirófano. En este sentido, se recomienda mantener un alto índice de sospecha en pacientes con antecedentes quirúrgicos y síntomas abdominales atípicos.

INTRODUCTION

Heterotopic ossification (HO) is a common condition in rehabilitation that occurs as a result of mature lamellar bone formation in extraskeletal soft tissue that normally does not contain bone, mostly around major joints (1). Patients who have suffered burns, stroke, spinal cord injury, traumatic amputations, joint replacements, and traumatic brain injury are at increased risk of developing HO (1).

HO may be classified into traumatic and neurogenic, but the exact pathophysiology of both is unknown. However, it has been established that bone morphogenetic proteins (BMP) stimulate mesenchymal spindle stem cells, also known as satellite cells, to migrate to the injured area and transform into fibroblasts and eventually osteoblasts (1).

Heterotopic ossification of the mesentery (HMO) is a rare and benign form of HO characterized by the formation of an ossifying pseudotumor at the mesenteric root, usually following abdominal trauma (2). It was first described in 1983 (2,3), and it has received different names over the years, including intra-abdominal myositis ossificans and mesenteritis ossificans (4), until Wilson *et al.* renamed it HMO in 1999 (2).

The pathogenesis of HMO is not yet fully understood due to its rare occurrence. However, it is believed that it results from stimulation of mesenchymal osteoprogenitor stem cells to differentiate into osteoblasts due to mechanical trauma, ischemia, inflammation, or intra-abdominal infections (2,3).

HMO is difficult to diagnose in patients with abdominal pain and discomfort due to its low prevalence, but abdominal computed tomography (CT) scans can help identify it preoperatively. However, differentiation between this condition and dystrophic calcification, bone neoplasms, leakage of contrast, foreign bodies, or extraskeletal osteosarcoma can be difficult (3).

The following is a case report of a patient with HMO discovered incidentally during an ileostomy closure procedure, which will allow us to broaden the understanding of this rare disease.

CASE PRESENTATION

A 41-year-old man attended a tertiary care center in Bogotá (Colombia) to undergo ileostomy closure, which had been performed a year earlier when he presented with acute perforated appendicitis resulting in appendiceal plastonectomy and requiring right hemicolectomy and end ileostomy. The patient was referred by the general surgery service of a secondary care institution in Bogotá, where he had undergone the procedure described above.

On admission, the patient was found to be in good general condition, without abdominal pain or fever, and with a functional ileostomy. While planning the surgical procedure, a contrasted CT scan of the abdomen was performed, showing a hyperdense image in the mesentery (Figure 1).

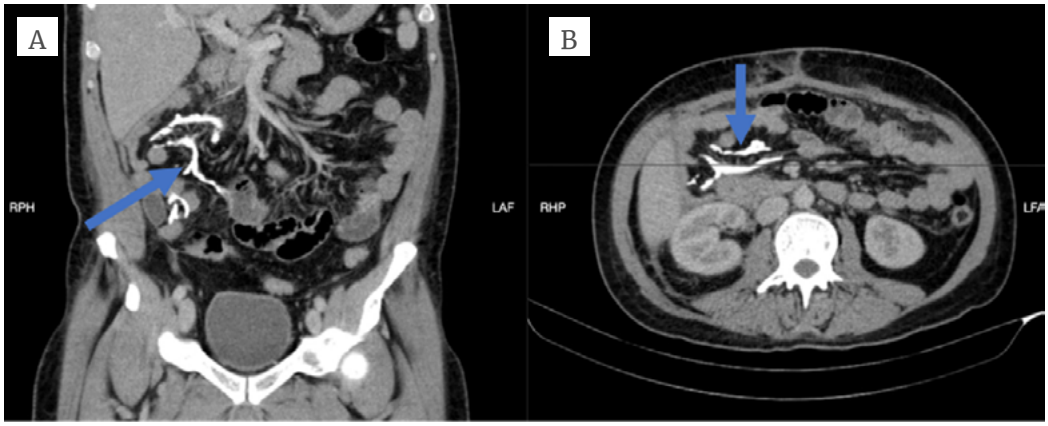

Figure 1. Contrast-enhanced computed tomography of the abdomen showing a hyperdense image suggestive of bone tissue in the mesentery. A) coronal view: the arrow points to a hyperdense image at the root of the mesentery, suggestive of bone formation; B) axial view: the arrow points to evidence of hyperdense foreign body suggestive of bone.

Source: Images obtained while conducting the study.

The patient was immediately taken to surgery, and during the procedure a bony multisegmental mass was found in the retroperitoneal region attached to the mesentery by abdominal adhesions (Figure 2), which was resected and sent for histopathological study. The biopsy report indicated that the mass was a benign soft tissue with mature bony trabeculae.

The patient stayed in the hospital for postoperative recovery and was discharged 5 days later with indications for analgesic treatment with acetaminophen 1g orally every 8 hours for 5 days and diclofenac 50mg orally every 12 hours for 3 days. Two weeks after surgery, he attended a general surgery follow-up and, due to his satisfactory progress, he was discharged by this service.

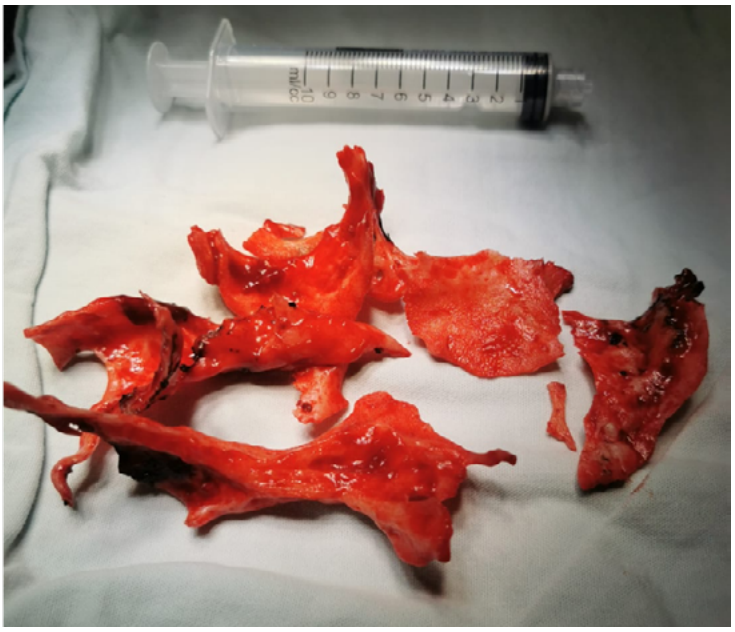

Figure 2. Multisegmental bony mass removed during ileostomy closure.
Source: Image obtained while conducting the study.

DISCUSSION

HMO is a rare condition with an unknown incidence; however, Althaqafi *et al.* (3), in a case report and literature review of this condition, found that 73 cases (51 publications) had been described worldwide in the early 2020s. The age of onset varies widely, ranging from 22 to 80 years, but most cases are described in middle-aged individuals (between 40 and 80 years of age) (5). HMO is often associated with previous traumatic injuries, a history of abdominal surgery (2,3,5,6), and even intra-abdominal infections (2,3,5).

There is difficulty in diagnosing HMO, especially in patients with abdominal pain and discomfort. In this sense, and due to the increase of cases reported in the last decade, it should be considered in the differential diagnosis of patients presenting with intestinal obstruction or enterocutaneous fistula (3). Usually, the time elapsed between the abdominal trauma and surgery is 2 to 4 weeks, although it may take up to 7 years (3), and for this reason it is usually an incidental finding by imaging or during an abdominal surgical procedure (2,3), as in the presented case.

HO is histologically characterized by calcium deposits consisting of both osteoblasts and structured bone layers (2,3,6,7). In macroscopic terms, it has been described as irregular bony tissues of hard consistency and sharp edges with some fusiform structures that look like needles (3).

Although the pathogenesis of HMO has not yet been fully elucidated, it is thought to be formed by stimulation of mesenchymal osteoprogenitor stem cells to differentiate into osteoblasts due to mechanical and surgical trauma, ischemia, edema, and intra-abdominal infection (3). In this regard, it has been described that the so-called osteogenic factors are involved in bone formation and remodeling processes (i.e., chemotaxis, proliferation and differentiation of cells involved in both bone formation and resorption), and in the development of blood vessels, nerves and medullary tissue (8). BMPs are multifunctional cytokines that are part of the transforming growth factor beta (TGF-β) family released from inflammatory cells at the site of inflammation, injury, sepsis, or wound (3). Studies in rats and rabbits have demonstrated that the systemic administration of TGF-β causes endosteum formation, generalized osteoblast hypertrophy, and increased cartilage formation (8).

According to the literature, there are four main factors that have a significant effect on the pathogenesis of heterotopic bone formation in the mesentery: 1) a trigger causing inflammation, 2) cell signaling at the site of injury via proinflammatory cytokines, 3) the presence of mesenchymal cells with potential for cellular differentiation (e.g. chondroblasts or osteoblasts), and 4) an environment that allows bone formation (2,9). Furthermore, as described by McCarthy & Sundaram (9), heterotopic bone can occur in five clinical contexts: genetic, post-traumatic, neurogenic, post-surgical, and as a consequence of reactive joint injuries. It is worth pointing out that HO has not been associated with metabolic alterations such as calcium and phosphorus disorders (9).

Based on clinical suspicion, a three-phase radionuclide bone scan can be requested to establish an early diagnosis, since this test is useful to evaluate the maturity of HO foci (10,11). However, abdominal CT is considered the imaging test of choice for the evaluation of mesenteric masses (2), as it allows determining the origin and relationship between the lesion and adjacent structures. In the present case, CT was used for surgical treatment planning.

In CT scans of patients with HMO and intestinal obstruction, dense and irregular calcified shadows, calcified densities, opacities of fat chains, among others, can be observed (3). However, this condition often resembles contrast extravasation or dystrophic calcification, or may be misinterpreted as a bone neoplasm, so radiological differentiation during CT interpretation may be difficult (6). The only way to establish the definitive diagnosis is by excision and histopathological analysis (3).

In asymptomatic patients, watchful waiting and follow-up with contrast-enhanced imaging is recommended, as surgical intervention is restricted to symptomatic patients because HMO can recur and even worsen with repeated surgeries (6). The indication for surgery is usually secondary to conditions such as generalized peritonitis, acute appendicitis, acute abdomen, intestinal obstruction, among others (2,6,12-13).

According to the literature, the prognosis of HO is favorable, with no evidence of malignant potential, and treatment should be conservative when possible, avoiding surgery to prevent further ossification (2). It has also been established that there is evidence suggesting the prophylactic treatment of HO with non-steroidal anti-inflammatory drugs, bisphosphonates, or even radiotherapy after surgery to prevent recurrence of ossification; however, there are still no statistically significant studies that prove their usefulness (2).

CONCLUSION

HMO is a benign neoformation of bone tissue in tissues that do not normally ossify. It is a rare condition (so its incidence has not been established) that is associated with a history of intra-abdominal surgery, as it is thought to develop as a result of inflammation or trauma. It is usually diagnosed via imaging or incidentally during surgery for other conditions, making it a diagnostic and therapeutic challenge for general surgeons. In this sense, it is recommended to always be highly suspicious in patients with a surgical history and atypical abdominal symptoms, as well as to implement a multidisciplinary approach to confirm the diagnosis and define the best treatment strategy for patients with abdominal pain and discomfort.

ETHICAL CONSIDERATIONS

For the preparation of this case report, informed consent was obtained from the patient for reviewing his medical records and using his diagnostic images. Moreover, his identity was always kept confidential, and the scientific, technical and administrative standards for health research established in Resolution 8430 of 1993 issued by the Colombian Ministry of Health were observed (14).

CONFLICTS OF INTEREST

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