Case Report

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Pathology Section

Heterotopic Ossification with Bone Marrow Elements in an Incisional Hernial Sac: A Rare Entity

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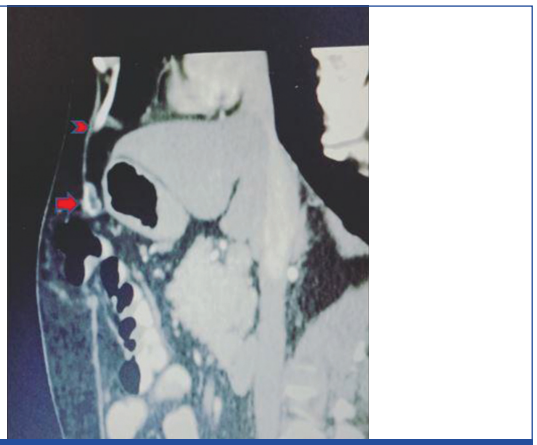
ABSTRACT

Heterotopic Ossification (HO) is usually seen in laparotomy wounds following gastrointestinal surgeries. The presence of marrow elements in an area of HO is very rare. A 57-year-old male with a history of omento-myelo-synangiosis for syringomyelia was admitted with an incisional hernia. A calcified nodule was detected in the wall of the hernia sac on Computed Tomography (CT). Histopathology of the excised nodule showed HO with marrow elements exhibiting trilineage haematopoiesis. HO is commonly an incidental finding during abdominal imaging for another condition. The presence of trilineage haematopoiesis within HO is extremely rare. As most of these patients are asymptomatic, conservative management is sufficient. Surgical excision is indicated, if any associated complications arise.

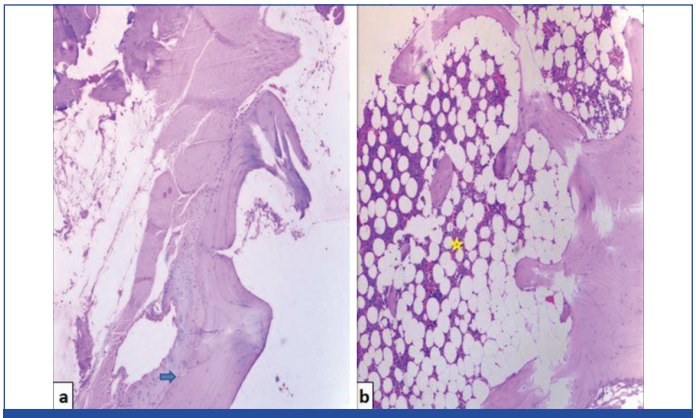
Keywords: Bone morphogenetic proteins, Fibroblast growth factor-2, Laparotomy wounds

CASE REPORT

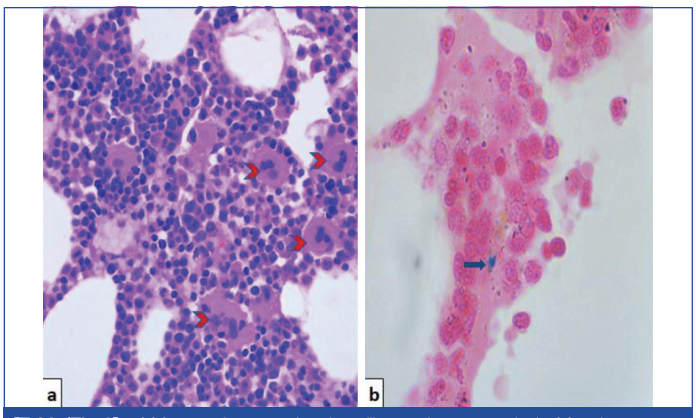
A 57-year-old male was presented with a past history of omento-myelo-synangiosis for syringomyelia was admitted with a five-year history of a bulge in the right side of the abdomen. He experienced recurrent vomiting and intermittent episodes of loose stools, but no pain. On examination, the abdomen was soft, and a scar in the epigastric region was observed. The diagnosis was an epigastric incisional hernia with bowel loop herniation. Abdominal Ultrasonography (USG) revealed a ventral hernia in the right paraincisional site at the epigastric region, with omentum and bowel loops as contents. Unfortunately, the USG images from the outside hospital could not be retrieved. A CT of the abdomen showed a hernial sac with a calcified nodule at the upper end of the sac, in the epigastric region, away from the xiphisternum [Table/Fig-1]. The diagnosis was a hernial sac with dystrophic calcification. The patient underwent open incisional hernia mesh repair. During the procedure, a calcified nodule measuring 2×2×1 cm was found in the subcutaneous plane near the upper end of the hernia defect. Gross findings showed a greyish-yellow hard nodule surrounded by fat. On the cut surface, there were densely calcified bony areas measuring 1.5 cm across, with central tiny cystic spaces. Histopathologically {Haematoxylin and Eosin (H&E)}, microscopy revealed fibroadipose tissue with a nodule consisting of lamellar bone and normocellular marrow with trilineage haematopoiesis [Table/Fig-2 (a,b), 3a]. There was no evidence of granuloma or malignancy. Perl's stain for iron showed focal iron deposits within the marrow elements [Table/Fig-3b]. The pathology report confirmed the diagnosis as HO with functioning bone marrow. The patient is currently on regular follow-up and there have been no recurrences as of the writing of the present report.



[Table/Fig-1]: A Computed Tomography (CT) abdomen (sagittal section) showing a calcified nodule in the upper end of the hernial sac (red arrow). The tip of xiphisternum is marked with an arrow head.



[Table/Fig-2]: a) Fibroadipose tissue with heterotopic bone showing enchondral ossification (blue arrow); b) Heterotopic bone showing lamellar bone with marrow elements (asterix). Haematoxylin and Eosin (H&E) stain, 100X.



[Table/Fig-3]: a) Marrow elements showing trilineage haematopoiesis. Megakaryocytes are marked with arrow heads. H&E, 400X; b) Perl's stain for iron showing iron deposits (arrow), 1000X.

DISCUSSION

The HO is defined as the formation of lamellar bone outside the skeletal system, usually observed in laparotomy wounds after

gastrointestinal surgeries [1]. It is commonly seen in males (89%) aged 18 to 81 years. The majority of cases are self-limiting, with some showing spontaneous regression [2]. The term "heterotopic" is derived from the Greek roots "hetero" and "topos," meaning "other place" [3]. Although, HO in abdominal incisions is uncommon, the presence of marrow elements showing trilineage haematopoiesis is very rare [1]. It is commonly observed in muscles and tissues adjacent to bones and less frequently in the mesentery, gastrointestinal tract, abdominal incisions, wound sites, blood vessel walls, kidneys, uterus and eyes [4,5]. HO is a typical complication of orthopaedic procedures and rarely occurs after abdominal surgeries [2]. However, a study by Kim J et al., examined postoperative CT scans of 152 patients, who underwent abdominal surgeries, found HO in 25% of cases [6]. According to Kaplan FS et al., an inciting event (e.g., trauma), a signaling pathway, a supply of mesenchymal cells and an appropriate environment are necessary factors for the development of HO [7]. Factors such as tissue hypoxia, hypercalcaemia, changes in sympathetic nerve activity, etc., are also involved in its development [8].

Several theories have been proposed regarding the pathogenesis of HO since, the exact mechanism is unknown. Initially, a theory suggested that, during vertical incisions for abdominal surgeries, there is an activation of osteoprogenitor cells from the xiphoid process or the pubic symphysis, which subsequently seed in the surgical wounds [2]. Another theory suggests that, HO results from a process of osteogenic induction, where immature pluripotent mesenchymal cells localised in muscle tissue differentiate into osteoblasts or chondroblasts, inducing subsequent bone formation [9]. The role of prostaglandins in the formation of HO has also been suggested. It is proposed that, prostaglandins regulate osteoblasts and osteoclasts involved in bone formation thereby, regulating osteogenesis [10]. The findings of studies by Shore EM et al., and Yu PB et al., suggest the critical role of Bone Morphogenetic Proteins (BMPs) in the formation of heterotopic bone [11,12]. Fibroblast growth factor-2 and other haematopoietic cytokines, such as granulocyte colony-stimulating factor, granulocyte-macrophage colony-stimulating factor, and erythropoietin, facilitate angiogenesis in the presence of mesenchymal stromal cells, which is essential for haematopoiesis [5,13,14]. BMPs can also stimulate angiogenesis by regulating osteoblast-derived vascular endothelial growth factor [15].

The presence of marrow elements within HO has been rarely reported in the literature. Wang D et al., reported a case of HO with marrow elements in the laparotomy scar of a woman, who underwent gastric reduction surgery for obesity treatment [16]. Rare complications such as fracture after trauma and traumatic perforation of abdominal viscera can occur within HO [1,17]. Additionally, there have been rare reports of malignant transformation to osteosarcoma, as well as, cases of haematological malignancies like myeloma involving the marrow elements of HO [18-20]. Borgia A et al. described a case of trilineage haematopoiesis in an excised area of HO from an enucleated blind painful eye in a patient with a history of lymphoplasmacytic lymphoma. Haematopoietic marrow within the HO in the present case showed infiltration by non-Hodgkin's B lymphoma [4].

CONCLUSION(S)

In conclusion, HO is a rare complication of abdominal surgery. The finding of trilineage haematopoiesis within HO is very rare. These cases are usually asymptomatic and diagnosed incidentally, requiring a conservative management. Surgical excision is indicated when symptomatic or when complications arise.

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