

Case Report

Extensive Circumferential Heterotopic Ossification Discovered at the Base of a Loop Ileostomy

Ahmad Bosaily,¹ John Edminister ,¹ Samarchitha Magal ,¹ Mohammad Jamil,¹ Amy Lynn,² and Glenn Hall ³

¹College of Medicine and Life Sciences, University of Toledo, Toledo, OH, USA

²Department of Pathology, ProMedica Health System, Toledo, OH, USA

³Department of Colorectal Surgery, ProMedica Health System, Toledo, OH, USA

Correspondence should be addressed to Glenn Hall; glenn.hallmd@promedica.org

Received 25 April 2019; Revised 17 July 2019; Accepted 21 November 2019; Published 2 December 2019

Academic Editor: Sibiu P. Saha

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Heterotopic ossification is a rare phenomenon defined by the formation of bone within nonossifying soft tissues. A rare variant of heterotopic ossification is heterotopic mesenteric ossification (HMO), in which there is involvement of the mesentery and surrounding intra-abdominal structures. There are only four previously reported cases of HMO involving an ileostomy. We present a case of HMO affecting an ileostomy which was discovered during elective stoma reversal in a 52-year-old male who required fecal diversion following perineal necrotizing fasciitis.

1. Introduction

Heterotopic ossification (HO) is a peculiar, benign disease characterized by organized bone formation in nonossifying muscle or soft tissue [1]. Ossification of soft tissue following orthopedic surgery, prolonged immobilization, or trauma has been well described. However, intra-abdominal cases of HO appear to be rare, with approximately 50 cases appearing in the medical literature [1]. Furthermore, there are even fewer reports of HO involving a stoma site [2–5]. In this report, we describe a case of HO involving a loop ileostomy.

2. Case

The patient is a 52-year-old African American male with an unwanted diverting loop ileostomy who was scheduled for an elective stoma reversal. Originally, the loop ileostomy was created for fecal diversion after a large perineal defect was made for the treatment of necrotizing fasciitis of the perineum. A high splenic flexure, foreshortened colonic mesentery, and morbid obesity precluded performing a diverting colostomy at that time. Of note, the patient had

no family history of HO or abdominal surgical history prior to the ileostomy creation.

During the reversal of the loop ileostomy, a circumferential incision was made at the mucocutaneous junction. Dissection was carried towards the serosal surface of the small bowel with electrocautery. There were significant adhesions between the small bowel and surrounding soft tissue which was much denser than expected. At that time, the base of the ostomy was reached and a dense structure was identified by palpation. It was circumferential in both limbs of the loop ileostomy. It was hard and fixed with a bone-like consistency. We encountered difficulty in freeing the small bowel from this circumferential dense structure, and this, combined with the patient's body habitus, rendered it difficult to take the stoma down safely through the peristomal incision. As a result, we converted the operation to an open procedure using a lower midline incision to reverse the ileostomy. After significant lysis of adhesions, the ileostomy was reached from the midline incision, and the hard structure was excised circumferentially using a combination of sharp dissection with Metzenbaum scissors and Bovie electrocautery. Upon gross inspection, it was observed that the structure was seated at the level of the rectus abdominis muscle and formed a

complete circle around the ileostomy. A portion appeared to have the consistency of thin connective tissue, while the majority of the specimen appeared osseous. The specimen grossly consisted of three irregular portions of ragged skeletal muscle and bone measuring 5.5, 7, and 12.5 cm in the greatest dimension (Figure 1). The skeletal muscle and bone were intertwined with flattened strands of bone interdigitating with the skeletal muscle fibers. Delicate, branching wisps of bone protruded from the specimen. No foreign body was identified. The specimen was radiographed using the Faxitron and showed both trabecular and cortical bone within the skeletal muscle (Figure 2). Microscopically, both thick cortical bone and trabecular bone with normocellular bone marrow were found within the skeletal muscle. Bone encircled and was immediately adjacent to skeletal muscle fibers (Figures 3 and 4).

The patient had follow-up visits after ileostomy reversal at weeks 2, 3, 7, 10, and 16. He developed transient fecal incontinence given the extensive nature of his perineal wound, which was successfully treated with pelvic floor biofeedback therapy by week 10. His ileostomy and midline wounds healed without abdominal pain, hernias, or signs of recurrence.

3. Discussion

Heterotopic ossification is a rare, benign disorder in which osteoblasts organize into discrete bone layers and deposit in nonskeletal tissues [1]. The phenomenon of HO was initially described in 1883 by Riedel in the context of a complication following spinal cord injury [6]. Previous literature reviews have described a 1901 case by Askanazy involving an abdominal scar [7]. The first reports of heterotopic ossification involving the intestinal mesentery were made by Lemershev et al. and Hansen et al. in 1983. In 1999, Wilson et al. coined the term heterotopic ossification to describe this pathology. Heterotopic ossification of soft tissue structures has been attributed to surgical, traumatic, and idiopathic etiologies involving different parts of the body [1, 3]. However, it appears that the amount of cases involving the intra-abdominal structures is low and that involvement of the stoma, as in our case, is even rarer [1, 4, 5, 8].

We describe a case of HO involving an ileostomy site, which we theorize could represent a rare subtype of intra-abdominal heterotopic mesenteric ossification (HMO). It appears that stoma involvement of HMO is a rare presentation of an already rare phenomenon, as only 4 other reports have specifically noted involvement of an ileostomy [2–5]. The morbidity associated with this case of HMO included a longer operation as necessitated by the need to convert to an open lower midline incision. Additionally, the patient suffered the less desirable cosmetic consequences of having this midline incision in addition to any potential risk of incisional hernia at the midline.

To date, there have been approximately 50 cases reported of HO [1, 9]. In 2012, Hiramata et al. performed a literature review of 47 cases of HO in which they further classified the occurrence by location in the abdomen. Twenty-seven cases involved the mesentery, 8 cases involved the intestinal



FIGURE 1: Gross specimen consists of three irregular portions of ragged skeletal muscle and bone measuring 5.5, 7, and 12.5 cm in the greatest dimension.

wall, and 6 cases involved the abdominal wall among other rarer sites [4]. Since that review in 2012, there have been a few more reports involving the mesentery. We also found recent cases involving relatively rare sites such as the abdominal wall and parietal peritoneum [1, 7, 9–13]. Considering all types of HMO, the mean age was 53.7 years old with at least 37 cases affecting males [14–16]. The lesions typically occurred after surgical interventions, but also sometimes without any previous surgeries, and when symptomatic, they most commonly presented as a small bowel obstruction [17].

Heterotopic ossification must be differentiated from other phenomena, such as dystrophic calcification and malignant bone tumors [1, 7]. Dystrophic calcification is a common pathology in which insoluble calcium salts are deposited in previously damaged tissue, despite a lack of osteoblastic activity [1]. Malignant tumors of the bone can be differentiated from HO by their growth pattern, both radiologically and histologically. The malignant bone tumors generally have indistinct borders on imaging, with histology showing central ossification and a lack of peripheral ossification [7]. This can be compared to the distinct HO lesions found on radiographs and primarily peripheral ossification seen on histology [7, 18].

There are three forms of HO: myositis ossificans progressiva (MOP), myositis ossificans circumscripta (MOC), and myositis ossificans traumatica (MOT). Myositis ossificans progressiva, also called fibrodysplasia ossificans progressiva or stone man disease, is an autosomal dominant connective tissue disorder caused by mutations in the *ACVR1/ALK2* gene, which codes for a bone morphogenetic protein type I receptor [19]. At birth, MOP is characterized by bilateral congenital deformities of the big toes and an otherwise normal appearance, with the heterotopic bone formation beginning only in childhood [20]. This disorder eventually progresses to immobilization due to extensive HO throughout the body and extra-articular joint fusion [19]. Myositis ossificans circumscripta is a disease in which there is isolated HO of a single muscle or muscle group; it is nonprogressive and can be idiopathic or occur following a trauma [21]. Myositis ossificans circumscripta and MOT are often used to define the same phenomenon, as they both can be products of trauma.

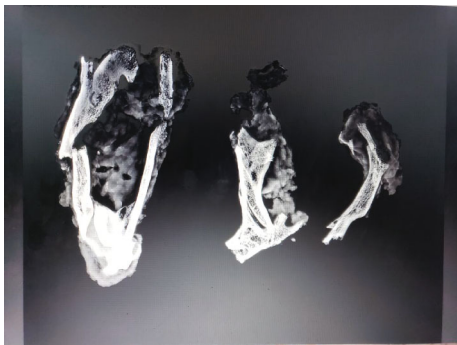


FIGURE 2: Specimen radiograph demonstrates both thick cortical and lacy trabecular bone irregularly distributed through the skeletal muscle fragments (12 cm, 7 cm, and 5.5 cm).

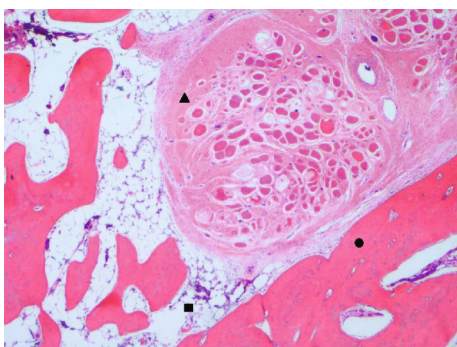


FIGURE 3: Trabecular bone (circle) with bone marrow (square) wrap around bundles of skeletal muscle (triangle) as is typical of myositis ossificans (2x magnification, H&E stain).

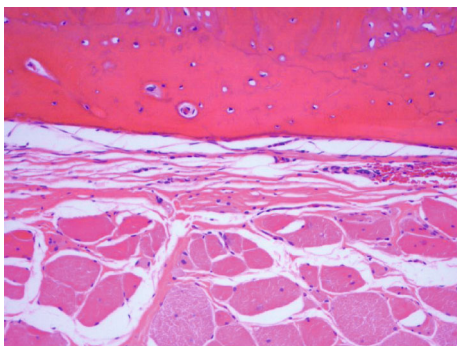


FIGURE 4: Cortical bone (top) demonstrating cement lines adjacent to skeletal muscle shown in cross section with peripherally located nuclei and myofibrils (10x magnification, H&E stain).

While MOT typically describes the phenomenon of HO in tissues adjacent to skeletal muscle, there have been reports of intra-abdominal MOT following abdominal surgery and/or trauma [22]. In these cases, an ossifying pseudotumor forms at the base of the mesentery and is often referred to as heterotopic mesenteric ossification, intra-abdominal myositis ossificans, mesenteritis ossificans, or heterotopic ossification of the intestinal mesentery [12]. Although the pathophysiology behind MOT remains unclear, there have been a few hypothesized theories. One theory speculates that MOT arises from

an exaggerated inflammatory response that induces activation of mesenchymal osteogenic progenitor cells, thus producing HO in nonskeletal tissue [23]. Another theory proposes that disruption of the xiphoid process during abdominal surgery results in the seeding of these disturbed cells along the abdominal incision [24]. Risk factors associated with MOT are the same as those for severe inflammatory response: severe trauma to the extremities, higher disease burden, and traumatic brain injury [24].

Due to the rare nature of HMO and its nonspecific radiological appearance, it may be challenging to diagnose preoperatively without a biopsy. Imaging modalities may not be able to accurately distinguish between differential diagnoses such as contrast leakage or osseous neoplasia. This difficulty with radiologic identification is compounded by the only mild increase in soft tissue density that can be observed in early stages of heterotopic ossification [25]. Laboratory markers are nondiagnostic and often unrevealing, but some HO reports have described a nonspecific but sensitive elevation in alkaline phosphatase theorized to be due to increases in osteoblastic activity [26].

The exact natural history of heterotopic ossification regarding onset may depend on the location. The time from initial injury to development of ossification has been reported as early as 4 weeks following burn injury and as early as 3 months following elbow injury [27, 28]. One case report suggests that the time from initial injury could be as early as days for development of HO specifically affecting the mesentery [1]. The literature suggests that HO lesions carry no malignant potential; therefore, we presume that without intervention a lesion may remain benign indefinitely [1]. In our case, the time between ileostomy creation and reversal was 7 months.

The benign pathology of this condition lends credence to a conservative management if the lesions are asymptomatic, but a symptomatic case (i.e., bowel obstruction) may be an indication for surgical excision [1]. Excision of symptomatic lesions comes with the risks expected of operating on a patient [29]. It would seem intuitive to avoid unnecessary surgeries or incisions in a patient with a history of HO because surgery itself is a causative factor, and caution should be taken when operating on these patients due to reports of recurrence or further progression [1, 9]. Prophylactic use of nonsteroidal anti-inflammatory drugs, bisphosphonates, or radiation therapy in high-risk patients have been shown to reduce the incidence of HO [30–32].

Heterotopic ossification must be distinguished from other pathologies that may present similarly. As in our case, the surgical team may initially confuse HO with a foreign body due to the shape and location of the ossification. This is an important consideration because a surgeon may choose to simply leave ossification behind or at most perform an incisional biopsy, as opposed to removal if it is a foreign body. Furthermore, a tissue capsule usually forms around a foreign body making its removal more straightforward. Knowledge of such cases may deter the surgeon from performing further dissection due to a benign pathology such as HMO [1]. However, in our case, dense adhesions between the ossification and small bowel required extensive dissection

and removal of the entire osseous structure to allow for ileostomy reversal. An on-table X-ray may also be used to help differentiate between a foreign body and ossification.

4. Conclusion

We report a rare case of HMO that was identified at loop ileostomy takedown. A thorough literature review revealed only four previous cases involving an ileostomy. Heterotopic mesenteric ossification can result in serious complications, including but not limited to small bowel obstruction, fistula, sepsis, and death [33]. However, surgical excision appears to be an acceptable and suitable treatment in symptomatic patients. This is a particularly rare condition described in the literature, though it may be underreported. As such, further investigation of the etiology, diagnosis, prevention, and treatment of HMO is essential to delineate appropriate prophylactic and therapeutic options for these patients.

Conflicts of Interest

The authors have no conflicts of interests to disclose.

Authors' Contributions

All authors have made substantial and direct contribution to the manuscript and approved it for publication.

References

- [1] C. Ferreira, C. Gomes, A. Melo et al., "Heterotopic mesenteric and abdominal wall ossification - two case reports in one institution," *International Journal of Surgery Case Reports*, vol. 37, pp. 22–25, 2017.
- [2] K. Sapalidis, T.-M. Strati, L. Liavas et al., "Heterotopic mesenteric ossification of ileostomy - "intraabdominal myositis ossificans"," *Romanian Journal of Morphology and Embryology*, vol. 57, no. 1, pp. 277–281, 2016.
- [3] C. W. Hicks, C. G. Velopulos, and J. M. Sacks, "Mesenteric calcification following abdominal stab wound," *International Journal of Surgery Case Reports*, vol. 5, no. 8, pp. 476–479, 2014.
- [4] K. Hiram, M. Yamaya, H. Jin, and S. Narumi, "A case of diverting stoma stenosis caused by heterotopic ossification," *Nihon Rinsho Geka Gakkai Zasshi (Journal of Japan Surgical Association)*, vol. 73, no. 12, pp. 3167–3171, 2012.
- [5] H.-W. Kao, C.-H. Liou, W.-C. Chang, C.-Y. Yu, and C.-Y. Chen, "Imaging features of heterotopic mesenteric ossification: a case report and literature review," *Chinese Journal of Radiology*, vol. 30, no. 1, pp. 55–58, 2005.
- [6] T. A. Balboni, R. Gobezie, and H. J. Mamon, "Heterotopic ossification: pathophysiology, clinical features, and the role of radiotherapy for prophylaxis," *International Journal of Radiation Oncology, Biology, Physics*, vol. 65, no. 5, pp. 1289–1299, 2006.
- [7] R. J. H. van Leeuwen, T. Kraal, S. Scholtens, and G. Visser, "A large heterotopic ossification in a 25 years old laparotomy scar," *Quantitative Imaging in Medicine and Surgery*, vol. 6, no. 4, pp. 470–473, 2016.
- [8] A. Rosenberg and A. Oliveira, "Myositis ossificans and fibro-osseous pseudotumor of digits," in *WHO Classification* *Tumours of Soft Tissue and Bone*, C. B. Fletcher, H. P. JA, and F. Mertens, Eds., pp. 50–51, IARC, Lyon, 4th ed edition, 2013.
- [9] M. Amalfitano, B. Fyfe, S. V. Thomas et al., "A case report of mesenteric heterotopic ossification: histopathologic and genetic findings," *Bone*, vol. 109, pp. 56–60, 2018.
- [10] T. Akinbiyi and S. Kaul, "Heterotopic ossification encountered during a complex ventral hernia repair: case report and literature review," *Eplasty*, vol. 17, article e29, 2017.
- [11] W.-T. Li, S.-Y. Horng, and H.-F. Chien, "Abdominis rectus intramuscular myositis ossificans," *Formosan Journal of Surgery*, vol. 49, no. 1, pp. 20–26, 2016.
- [12] O. Ioannidis, A. Sekouli, G. Paraskevas et al., "Intra-abdominal heterotopic ossification of the peritoneum following traumatic splenic rupture," *Journal of Research in Medical Sciences : The Official Journal of Isfahan University of Medical Sciences*, vol. 17, no. 1, pp. 92–95, 2012.
- [13] G. Sahsamani, P. Triantafylakis, K. Gkouzis, K. Katis, and G. Dimitrakopoulos, "Intra-abdominal myositis ossificans in an asymptomatic patient during closure of a Hartmann's colostomy," *Journal of Surgical Case Reports*, vol. 2016, no. 11, article rjw203, 2016.
- [14] M. A. Myers and J. P. Minton, "Heterotopic ossification within the small-bowel mesentery," *Archives of Surgery*, vol. 124, no. 8, pp. 982–983, 1989.
- [15] R. M. Patel, S. W. Weiss, and A. L. Folpe, "Heterotopic mesenteric ossification: a distinctive pseudosarcoma commonly associated with intestinal obstruction," *The American Journal of Surgical Pathology*, vol. 30, no. 1, pp. 119–122, 2006.
- [16] J. Charles and J. A. Hunt, "Heterotopic bone formation in abdominal incisions," *Hawaii Medical Journal*, vol. 51, no. 3, pp. 65–69, 1992.
- [17] Ø. Hansen, F. Sim, P. F. Marton, and O.-P. N. Grüner, "Heterotopic Ossification of the Intestinal Mesentery: Report of a Case Following Intraabdominal Surgery," *Pathology, Research and Practice*, vol. 176, no. 2–4, pp. 125–130, 1983.
- [18] L. M. White and R. Kandel, "Osteoid-producing tumors of bone," *Seminars in Musculoskeletal Radiology*, vol. 4, no. 1, pp. 25–43, 2000.
- [19] F. S. Kaplan, M. Xu, P. Seemann et al., "Classic and atypical fibrodysplasia ossificans progressiva (FOP) phenotypes are caused by mutations in the bone morphogenetic protein (BMP) type I receptor ACVR1," *Human Mutation*, vol. 30, no. 3, pp. 379–390, 2009.
- [20] R. J. Pignolo, E. M. Shore, and F. S. Kaplan, "Fibrodysplasia ossificans progressiva: clinical and genetic aspects," *Orphanet Journal of Rare Diseases*, vol. 6, no. 1, p. 80, 2011.
- [21] S. O'Brien, "Non-traumatic myositis ossificans circumscripta," *Sonography*, vol. 4, no. 2, pp. 88–92, 2017.
- [22] J. Kim, Y. Kim, W. K. Jeong, S. Y. Song, and O. K. Cho, "Heterotopic ossification developing in surgical incisions of the abdomen: analysis of its incidence and possible factors associated with its development," *Journal of Computer Assisted Tomography*, vol. 32, no. 6, pp. 872–876, 2008.
- [23] M. B. K. Potter LJAF, T. A. Davis, C. K. N. Evans et al., "Heterotopic ossification following combat-related trauma," *The Journal of Bone & Joint Surgery*, vol. 92, Supplement_2, pp. 74–89, 2018.
- [24] J. D. Wilson, C. J. Montague, P. Salcuni, C. Bordi, and J. Rosai, "Heterotopic mesenteric ossification (intraabdominal

- myositis ossificans’): report of five cases,” *The American Journal of Surgical Pathology*, vol. 23, no. 12, pp. 1464–1470, 1999.
- [25] B. A. Tonino, H. G. van der Meulen, K. C. Kuijpers, W. M. Mallens, and A. P. van Gils, “Heterotopic mesenteric ossification: a case report (2004: 10b),” *European Radiology*, vol. 15, no. 1, pp. 195–197, 2005.
- [26] G. Bovo, F. Romano, E. Perego, C. Franciosi, R. Buffa, and F. Uggeri, “Heterotopic mesenteric ossification (“intraabdominal myositis ossificans”): a case report,” *International Journal of Surgical Pathology*, vol. 12, no. 4, pp. 407–409, 2004.
- [27] S. K. He, M. Yi, G. Zhong, S. Q. Cen, J. L. Chen, and F. G. Huang, “Appropriate excision time of heterotopic ossification in elbow caused by trauma,” *Acta Orthopaedica et Traumatologica Turcica*, vol. 52, no. 1, pp. 27–31, 2018.
- [28] R. Kornhaber, N. Foster, D. Edgar et al., “The development and impact of heterotopic ossification in burns: a review of four decades of research,” *Scars, Burns & Healing*, vol. 3, article 205951311769565, 2017.
- [29] Z. Torgersen, A. Osmolak, J. Bikhchandani, and A. R. Forse, “Ectopic bone in the abdominal cavity: a surgical nightmare,” *Journal of Gastrointestinal Surgery*, vol. 17, no. 9, pp. 1708–1711, 2013.
- [30] Y. Lemeshev, C. J. Lahr, J. Denton, S. P. Kent, and A. G. Diethelm, “Heterotopic bone formation associated with intestinal obstruction and small bowel resection,” *The Alabama Journal of Medical Sciences*, vol. 20, no. 3, pp. 314–317, 1983.
- [31] T. J. Blokhuis and J. P. Frolke, “Is radiation superior to indomethacin to prevent heterotopic ossification in acetabular fractures?: a systematic review,” *Clinical Orthopaedics and Related Research*, vol. 467, no. 2, pp. 526–530, 2009.
- [32] M. Milakovic, M. Popovic, S. Raman, M. Tsao, H. Lam, and E. Chow, “Radiotherapy for the prophylaxis of heterotopic ossification: a systematic review and meta-analysis of randomized controlled trials,” *Radiotherapy and Oncology*, vol. 116, no. 1, pp. 4–9, 2015.
- [33] H. Honjo, Y. Kumagai, T. Ishiguro et al., “Heterotopic mesenteric ossification after a ruptured abdominal aortic aneurism: case report with a review of literatures,” *International Surgery*, vol. 99, no. 4, pp. 479–484, 2014.