Heterotopic Mesenteric Ossification after Gastrectomy for Watermelon Stomach

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Abstract

Heterotopic mesenteric ossification is a rare disorder. Only a few cases have been reported in the literature, associated with previous abdominal surgery or trauma. We report a case of heterotopic mesenteric ossification leading to abdominal sepsis, after abdominal operation for recurrent gastric bleeding, due to gastric antral vascular ectasia (GAVE), otherwise called "watermelon stomach", another rare disorder.

Key words

Heterotopic ossification – watermelon stomach – gastric antral vascular ectasia – GAVE disease.

Introduction

Heterotopic mesenteric ossification, lately described as intra-abdominal myositis ossificans, is an ossifying pseudosarcoma, associated with intestinal obstruction [1]. Only 20 cases have been recently reviewed in the literature, while Mayo Clinic have also announced 9 more new cases [2]. Patients are predominately males who have undergone recent abdominal surgery or trauma. The precise pathophysiologic mechanism of bone formation in soft tissues, in this case the omentum, is not known, although heterotopic ossification mechanism has been thoroughly described in human or animal model [3, 4]. On the other hand, gastric antral vascular ectasia (GAVE), otherwise called watermelon stomach, is a rare and often misdiagnosed cause of recurrent upper gastrointestinal (GI) bleeding.

A previously unreported case of greater omentum ossification following total gastrectomy for a GAVE disease variant is the subject of this report.

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Case report

A 42-year-old male was initially admitted to our hospital for hematemesis. He was offered an endoscopy, where a bleeding vessel was found and was injected with epinephrine. Hemostasis was initially achieved. Two days later recurrent and massive hematemesis caused hemodynamic instability and laparotomy was urgently performed. Gastrotomy and ligation of a vessel that was considered a Dieulafoy lesion was considered the proper therapeutic management. The patient had an uneventful postoperative period of 5 days when a massive hematemesis recurred again. A decision to do a second laparotomy was made and a total gastrectomy with a Roux-en-Y oesophagojejunal anastomosis was offered to the patient. Histologically, there was a superficial fibromuscular hyperplasia of the mucosa of the pylorus accompanied by microvascular thrombosis and capillary ectasia (dilated and focally dysplastic vesells) in the lamina propria (Fig. 1). This diffused angiodysplasia was classified by the pathologists as GAVE or watermelon stomach disorder.

The patient was transferred to the ICU, where he suffered an acute respiratory distress syndrome (ARDS) and long lasting fever. Because of the critical condition of his health and the unknown origin of uncontrollable septic fever, a diagnostic laparotomy was performed. An inflammatory greater omentum was found and excised. The peritoneal cavity was thoroughly washed. No other pathology was found. Previous anastomoses were checked intact. The excised omentum was very rough and scratchy. Pathologic examination revealed inflammatory heterotopic ossification of the greater omentum. In detail, the corresponding tissue sections were characterized by well-formed bone trabeculae and prominent osteoblastic rimming, as a result of progressive metaplasia. Furthermore, medium to high cellularity and sporadic mitotic figures were correlated to cartilage formation, and bone production (Fig. 2).

In the postoperative period a low flow enterocutaneous fistula occurred, which was managed conservatively. The

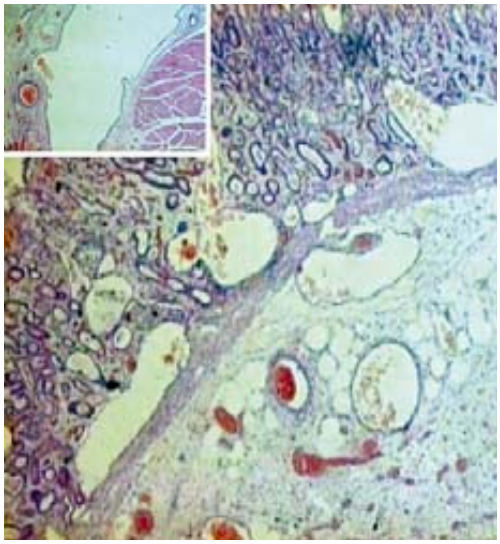

Fig 1. Watermelon stomach. Note the superficial fibromuscular hyperplasia of gastric mucosa of the pylorus accompanied with microvascular thrombosis and capillary ectasia (dilated and focally dysplastic vesells) in the lamina propria. H&E stain, original magnification 10x (main images) and 40x (inside images).

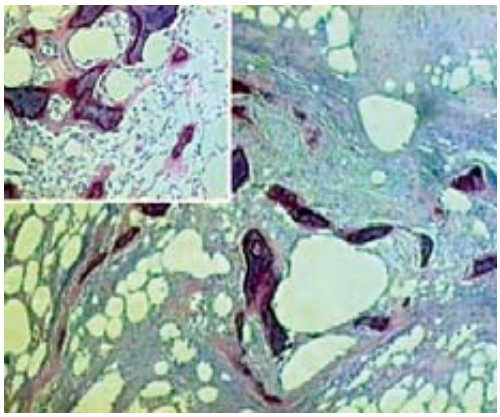

Fig 2. Heterotopic mesenteric ossification. Note the cartilage formation, and bone production as a result of excessive osteoblastic activity. H&E stain, original magnification 10x (main images) and 40x (inside images).

patient had a urinary tract infection and bacteraemia treated with antibiotics. He was discharged from the ICU after 18 days and was dismissed from the ward after 70 days of hospitalization. The patient has remained in good health for 3 years now.

Discussion

Watermelon stomach is a rare disease, as well as intra-abdominal ossification of the omentum. Watermelon stomach is associated with chronic blood loss and anaemia. It is characterized by distinct pathologic findings such as superficial fibromuscular hyperplasia of the gastric mucosa of the antrum accompanied with microvascular thrombosis and capillary ectasia in the lamina propria [5]. Pathogenesis of the disease is not really known. Mechanical or humoral factors such as malfunction of capillary sphincters and vasoactive substances, hormones or neuropeptides may induce transfusion-dependent anemia with characteristic endoscopic appearance [5]. Therapeutic solutions include endoscopic treatement, pharmacologic agents, thermal ablation and surgery, in order to reduce or eliminate blood transfusions [6, 7]. Watermelon stomach is most frequently associated with autoimmune disorders, such as Raynaud's disease and sclerodactyly or primary biliary cirrhosis, systemic lupus erythematosus and scleroderma [1 -10]. Watermelon stomach is not a frequent disorder but can easily unstabilize a blood transfusion dependent patient due to recurrent bleedings.

Heterotopic bone formation in abdominal wounds is a subgroup of myositis ossificans traumatica [2], very often described in orthopedic surgery, especially hip replacement operations [3, 4]. All cases described are correlated with vertical or midline incisions [5]. Two theories are suggested to explain the mechanism of bone formation. Either small particles from periostum or perichondrium of sternum or pubic symphysis are inoculated during laparotomy, or immature pluripotent mesenchymal cells differentiate to osteoblasts or chondroblasts due to local injury. The pathogenesis of the ossification, omentum and mesentery are the affected abdominal structures causing in most cases small bowel obstruction, as well as peritonitis and sepsis, as reported in one case [1]. Intraabdominal ossification of the greater omentum after treatment for watermelon stomach is an extremely bizarre sequence of disorders that has never been described before. Histologically, the entity of heterotopic mesenteric ossification shares common characteristics with other benign mesenteric tumours such as intra-abdominal myositis ossificans or of different origin tumours, such as giant cell tumor of the soft tissues and hemangioendothelioma. Differential diagnosis between benign and malignant entities must be based on the grade of nuclear polymorphism, level of cellularity and also on the number of mitoses [11-14].

In **conclusion,** abdominal surgery with midline incision may conclude to heterotopic intra-abdominal ossification that can cause gastrointestinal obstruction and sepsis. Clinical, endoscopic, surgical, radiologic and pathologic alertness can be vital for patients with such an unusual pathology.

References

1. Patel RM, Weiss SW, Folpe AL. Heterotopic mesenteric ossification: a distinctive pseudosarcoma commonly associated with intestinal obstruction. Am J Surg Pathol 2006; 30: 119-122.
2. Zamolyi RQ, Souza P, Nascimento AG, Unni KK. Intraabdominal myositis ossificans: a report of 9 new cases. Int J Surg Pathol 2006; 14: 37-41.
3. Kaplan FS, Glaser DL, Hebela N, Shore EM. Heterotopic ossification. J Am Acad Orthop Surg 2004; 12: 116-125.
4. Cohly HH, Buckley RC, Pecunia R, Das SK. Heterotopic bone

formation: presentation of an experimental rat model and a clinical case. Biomed Sci Instrum 2003; 39:446-453.
5. Novitsky YW, Kercher KW, Czerniach DR, Litwin DE. Watermelon stomach: pathophysiology, diagnosis, and management. J Gastrointest Surg 2003; 7: 652-661.
6. Gretz JE, Achem SR. The watermelon stomach: clinical presentation, diagnosis, and treatment. Am J Gastroenterol 1998; 93: 890-895.
7. Dulai GS, Jensen DM. Treatment of watermelon stomach. Curr Treat Options Gastroenterol 2006; 9: 175-180.
8. Gostout CJ, Viggiano TR, Ahlquist DA, Wang KK, Larson MV, Balm R. The clinical and endoscopic spectrum of the watermelon stomach. J Clin Gastroenterol 1992; 15: 256–263.
9. Archimandritis A, Tsirantonaki M, Tzivras M, Hatzis G, Davaris P. Watermelon stomach in a patient with vitiligo and systemic lupus erythematosus. Clin Exp Rheumatol 1996; 14: 227–228.
10. Watson M, Hally RJ, McCue PA, Varga J, Jimenez SA. Gastric antral vascular ectasia (watermelon stomach) in patients with systemic sclerosis. Arthritis Rhem 1996; 39: 341–346.
11. Wilson JD, Montague CJ, Salcuni P, Bordi C, Rosai J. Heterotopic mesenteric ossification (intraabdominal myositis ossificans): report of five cases. Am J Surg Pathol 1999; 23: 1464–1470.
12. Cope R. Heterotopic ossification. South Med J 1990; 83: 1058-1064.
13. Hinck SM. Heterotopic ossification: a review of symptoms and treatment. Rehabil Nurs 1994; 19: 169-173.
14. Reardon MJ, Tillou A, Mody DR, Reardon PR. Heterotopic calcification in abdominal wounds. Am J Surg 1997; 173: 145-147.