http:// ijp.mums.ac.ir
Case Report (Pages: 17443-17447)

Gastric Inflammatory Myofibroblastic Tumor as a Cause of Gastric Outlet Obstruction in Children

Seyed Ali Jafari [1], Mahdi Rouhbakhsh [2], Hamidreza Kianifar [3], Mohammadali Kiani [4], Maryam Khalesi [5], Nadia Danaei [6], * Fatemeh Basirinezhad [7]

[1] MD, Assistant Professor, Department of Pediatric Gastroenterology, Akbar children hospital, Mashhad University of medical sciences, Mashhad, Iran.

[2] Clinical Research Development Unit of Akbar Hospital, Faculty of Medicine, Mashhad University of Medical Sciences, Mashhad, Iran.

[3] Professor, Department of Gastroenterology, Faculty of medicine, Mashhad University of medical sciences, Mashhad, Iran.

[4] Professor, Department of Pediatric Gastroenterology, Akbar children hospital, Mashhad University of medical sciences, Mashhad, Iran.

[5] Assistant Professor, Department of Pediatric Gastroenterology, Akbar children hospital, Mashhad University of medical sciences, Mashhad, Iran.

[6] Department of Pediatric Gastroenterology, Akbar children hospital, Mashhad University of medical sciences, Mashhad, Iran.

[7] Clinical Research Development Unit of Akbar Hospital, Faculty of Medicine, Mashhad University of Medical Sciences, Mashhad, Iran.

Abstract

Gastric inflammatory myofibroblastic tumor (IMT) is a rare tumor that is almost seen in children with a wide range of clinical features. However, radiologic imaging and gastro endoscopy can be helpful, but the confirmation of the IMT diagnosis is via a histopathologic gastric specimen evaluation. Here we present the 2nd case of IMT with the clinical manifestations of a gastric outlet obstruction (GOO) in an eleven month old infant. Surgical treatment should be considered for the treatment of IMT.

Key Words: Case study, Gastric outlet obstruction, Histopathologic gastric specimen evaluation, Inflammatory Myofibroblastic Tumor

* Please cite this article as: Jafari SA, Rouhbakhsh M, Kianifar H, Kiani M, Khalesi M, Danaei N, Basirinezhad F. Gastric Inflammatory Myofibroblastic Tumor as a Cause of Gastric Outlet Obstruction in Children. Int J Pediatr 2023; 11 (02):17443-17447. DOI: **10.22038/ijp.2023.68757.5126**

**** Corresponding Author:***

Fatemeh Basirinezhad, Clinical Research Development Unit of Akbar Hospital, Faculty of Medicine, Mashhad University of Medical Sciences, Mashhad, Iran. Email: basirinezhadf@mums.ac.ir.

Received date: Dec.02,2022; Accepted date:Feb.05,2023

1- INTRODUCTION

Gastric outlet obstruction (GOO) is a condition in which mechanical obstruction in the gastric pylorus region leads to epigastric pain and postprandial vomiting (1). Idiopathic (or infantile) hypertrophic pyloric stenosis (IHPS) is the most common cause of GOO in children with a prevalence of 2 to 5 per 1000 live births (2, 3). Other causes of GOO include:

a) Gastric volvulus,

b) Gastric antral web,

c) Gastric duplication cysts,

d) Gastric polyps,

e) Neoplasm, and other rare causes (4). When IHPS is excluded, the prevalence of other causes encounters only 1 in 100 000 live births (5).

Gastric inflammatory myofibroblastic tumor (IMT) is a rare and unique mesenchymal tumor characterized as a low-grade malignant or borderline tumor. In recent years, the World Health Organization (WHO) proposed the term IMT, which has gradually been recognized by experts and scholars (6).

We presented here a child with Gastric IMT, causing GOO.

2- CASE PRESENTATION

An 11-month-old boy, weighing 10 kg, presented to our hospital with a history of a mildly distended abdomen, fever, and recurrent nonbilious postprandial projectile vomiting for 2 months. In clinical examination, his sclera was pale and anthropometric measures were normal. Primary laboratory tests indicated microcytic anemia, leukocytosis, and elevated inflammatory markers such as Erythrocyte Sedimentation Rate (ESR), C-reactive protein (CRP) level, and platelet count (hemoglobin 7.1g/dl; WBC count 20.82×103 cells/ml; PLT count 962×103 cells/MCL; ESR 69 mm/hour; CRP 102 mg/dl). Abdominal ultrasound indicated a mass between the anatomical position of the gastric outlet and the proximal part of the duodenum. In the contrast study, distended stomach, filling defect in the antral region and little transit of contrast media after 15 minutes were shown (**Fig. 1**).

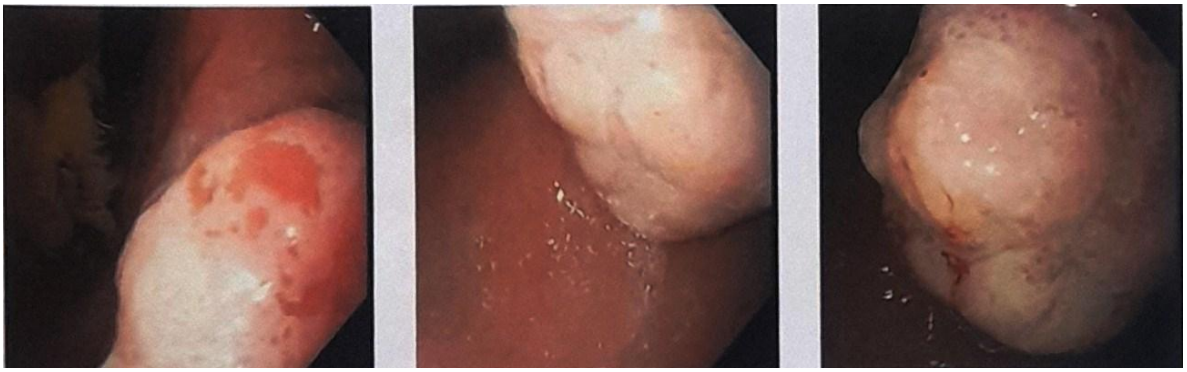

Fig. 1: Esophagogastroduodenoscopy indicating a mass between the anatomical position of the gastric outlet and the proximal part of the duodenum, The lesion in gastric fundus, incisura, and antrum from right to left

Esophagogastroduodenoscopy showed a large polypoid verrucous mass that lied from cardia to antrum and obstructed the pylorus region. **Fig. 2** showed the lesion in gastric fundus, incisura, and antrum from right to left. The elective laparotomy was

done via the transverse supraumbilical incision. There was an 18×12 cm polypoid mass at the antrum and body of the stomach near the distal margin. Several lymphadenopathies were seen around the stomach and subhepatic space. Billroth II gastrectomy was done and 13 lymph nodes with a maximum size of 1 cm were resected.

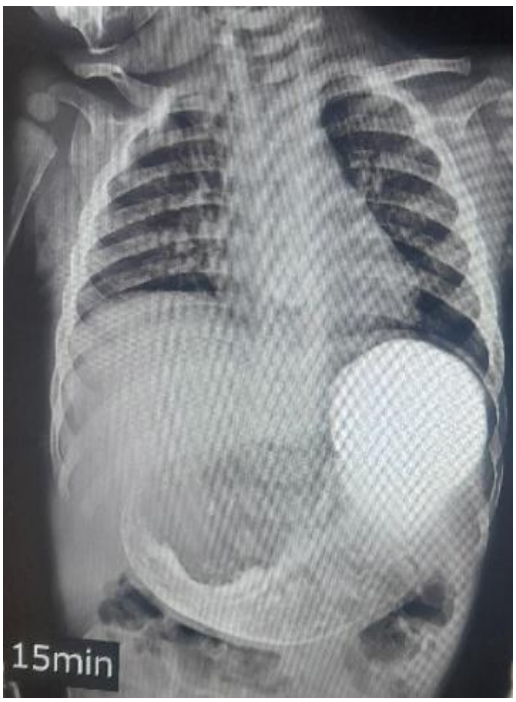

Fig. 2: The lesion in gastric fundus, incisura, and antrum from right to left

The histopathological evaluation showed a gastric stromal neoplasm with a fascicular growth pattern, moderate cellularity composed of the spindle to stellate myofibroblastic cells with vesicular and nucleolated nuclei (**Fig. 3** and **4**). Immunohistochemistry staining (IHC) were positive for smooth muscle actin (SMA), Vimentin, and Anaplastic Lymphoma Kinase (ALK). The patient did well with no recurrence or complication over 2 years of follow-up.

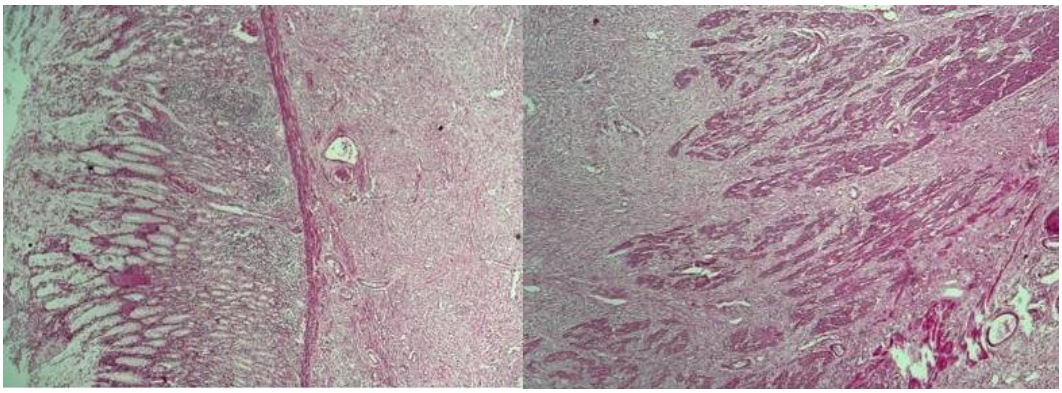

Fig. 3: Inflammatory myofibroblastic tumor cells infiltrating from submucosa to muscularis properia and serosa (H & E staining, low power field).

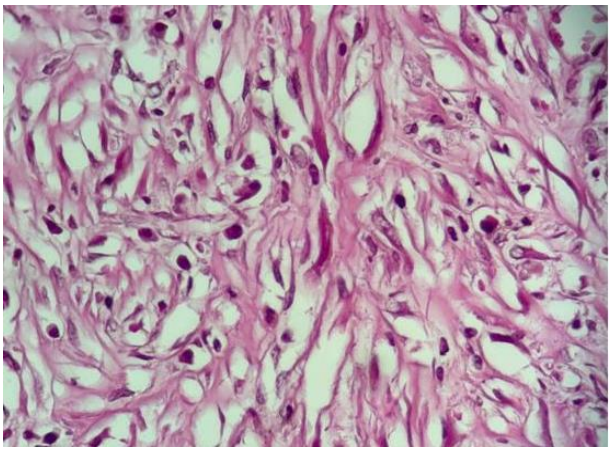

Fig. 4: Infiltration of spindle cell myofibroblasts intermixed with inflammatory cells (H & E staining, high power field)

3- DISCUSSION

IMT is defined as a histologically distinctive lesion of uncertain behavior. It was formerly known as pseudoinflammatory tumor, plasma cell granuloma, inflammatory fibroblastoma, and inflammatory fibromyalgia. IMT frequently recurs and rarely metastasizes (7). It has shown that the most common locations of IMT in children are abdomen and pelvis, respectively (8). The clinical features of gastric IMT are variable. Some of them include abdominal pain, upper gastrointestinal bleeding, vomiting, growth retardation, and an abdominal mass. Furthermore, fever and/or weakness are seen in almost all patients. Severe microcytic hypochromic anemia, thrombocytosis, elevated ESR, and hypergammaglobulinemia are common laboratory data in a patient with IMT (9).

As yet, it is unclear what causes gastric IMT, but it may develop from stem cells in the gastric mesenchyme that differentiate into myofibroblasts. Risk factors such as inflammation, surgery, trauma, and unique infection are associated with the occurrence of IMT (10).

To our knowledge, this is the 2nd case of gastric IMT with the clinical features and presentation of GOO in children. The previous case was an 8-month girl who presented with persistent vomiting for several days, an epigastric palpable mass, and normal laboratory data (11). However, in our patient, the clinical manifestations were low-grade fever, abdominal distention, a gastric mass, and recurrent vomiting for 2 months. Microcytic hypochromic anemia, elevated ESR, and thrombocytosis were also seen in the laboratory data.

IMT in most cases mimics the features of malignancy in upper endoscopy and radiologic imaging. Hence, the diagnosis of IMT is based on the histopathological findings in postoperative evaluation. In histopathology, IMT is characterized by spindle, the epithelioid proliferation of myofibroblasts, a lymphoplasmacytic infiltrate, and a myxoid background stroma (7).

A complete or partial surgical resection is the most common treatment for IMT (12). However, the case was treated with combined endoscopic and laparoscopic gastric wedge resection (13).

In conclusion, gastric IMT is a rare tumor that can cause GOO. Complete surgical resection is the first line of treatment for IMT. Because of the risk of recurrence, a long-term follow-up should be considered.

4- ACKNOWLEDGEMENT

This work was supported by the Clinical Research Development Center of Akbar Hospital, Mashhad University of Medical Sciences, Mashhad, Iran. We also gratefully acknowledge Dr Paria Dehghanian, pathologist, for her contribution in this case report.

5- REFERENCES

1. Iqbal U, Khara HS, Hu Y, Kumar V, Tufail K, Confer B, Diehl DL, EUS-guided gastroenterostomy for the management of gastric outlet obstruction: a systematic review and meta-analysis. Endoscopic ultrasound, 2020. 9(1): p. 16.

2. Hernanz-Schulman, M., Infantile hypertrophic pyloric stenosis. Radiology, 2003. 227(2): p. 319-331.

3. Peters |B, Oomen MWN, Bakx R, Benninga MA, Advances in infantile hypertrophic pyloric stenosis. Expert review of gastroenterology & hepatology, 2014. 8(5): p. 533-541.

4. Otjen JP, Iyer RS, Phillips GS, Parisi MT, Usual and unusual causes of pediatric gastric outlet obstruction. Pediatr Radiol, 2012. 42(6): p. 728-37.

5. Cremin, B., Neonatal pre-pyloric membrane. South African Medical Journal, 1967. 41(41): p. 1076-1079.

6. Wang Y, Shen L, Yun T, Zhu C, Wang P, Wang S, Clinicopathological features of gastric inflammatory myofibroblastic tumor: Report of five cases. Experimental and Therapeutic Medicine, 2021. 22(3): p. 1-7.

7. Coffin, C.M., J.L. Hornick, and C.D. Fletcher, Inflammatory myofibroblastic tumor: comparison of clinicopathologic, histologic, and immunohistochemical features including ALK expression in atypical and aggressive cases. The American journal of surgical pathology, 2007. 31(4): p. 509-520.

8. Dalton BGA, Thomas PG, Sharp NE, Manalang MA, Fisher JE, Moir CR, Peter SDS, Iqbal CW, Inflammatory myofibroblastic tumors in children. Journal of pediatric surgery, 2016. 51(4): p. 541-544.

9. Mirshemirani A, Khaleghnejad Tabari A, Sadeghian N, Shariat-Torbaghan S, Pourafkari M, Mohajerzadeh L, Abdominal inflammatory myofibroblastic tumor: report on four cases and review of literature. Iranian Journal of Pediatrics, 2011. 21(4): p. 543.

10. Lemale J, Boudjemaa S, Parmentier B, Pointe HDL, Coulomb A, Dainese L, A pseudotumoral lesion revealing Meckel's diverticulum. Archives de Pediatrie: Organe Officiel de la Societe Francaise de Pediatrie, 2016. 23(11): p. 1157-1160.

11. Hirschburger M, Enders J, Alzen G, Padberg W, Wagner HJ, An inflammatory myofibroblastic tumor of the stomach as a rare cause of gastric outlet obstruction in an 8-month-old infant. Klin Padiatr, 2010. 222(3): p. 192-3.

12. Albayrak F, Dursun H, Albayrak Y, Altas S, Uyanik A, Yildirim R, Inflammatory myofibroblastic tumor of the stomach in an adult woman: a rare intermittent cause of gastric outlet obstruction. Tumori, 2010. 96(3): p. 492-5.

13. Hayashi M, Kawakubo H, Mayanagi S, Nakamura R, Suda K, Wada N, Kitagawa Y, Gastric inflammatory myofibroblastic tumor treated with combined laparoscopic and endoscopic gastric wedge resection: a case report. World Journal of Surgical Oncology, 2018. 16 (1): p. 1-5.