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Case Report
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Inflammatory Myofibroblastic Tumor of the Stomach: A Rare Case Report in a 10-Year-Old Girl

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Abstract

Inflammatory Myofibroblastic Tumor (IMT), also called inflammatory pseudo tumor, is a rare childhood disease that can mimic malignancy. The etiology is not fully understood. It is usually diagnosed with histopathological examination performed after mass excision. In this case report, we present a rare case of inflammatory myofibroblastic tumor of the stomach.

Introduction

Inflammatory myofibroblastic tumor is a rare disease that can occur in many different parts of the body but most frequently in the lungs [1]. It is more common in children and young adults [2]. Although it can be diagnosed preoperatively via computed tomography and magnetic resonance imaging, the final diagnosis is usually established by postoperative histopathological evaluation. In most cases, lesions are well-circumscribed [3,4]. Although it may result from inflammation, whether it is a true neoplasm is controversial [5]. There are different opinions in the literature regarding conservative or medical treatment options and resection in the diagnosis and treatment [1,6].

In this paper, we present a rare case of inflammatory myofibroblastic tumor of the stomach which was histopathologically diagnosed after excision of the mass.

Case Presentation

A-10-year old female patient referred to the hospital with the complaints of weakness and bloody vomiting two days after the use of acetylsalicylic acid due to respiratory tract infections. In laboratory studies, hemoglobin was 3.4 g/dL. The patient underwent erythrocyte replacement. The patient had a history of anorexia and intermittent epigastric pain in the last three months. After patient's vital signs and laboratory values become stable, gastroduoendoscopy was performed for the etiological evaluation purposes. Gastroduoendoscopy revealed a polyploidy mass in the gastric corpus (Figure 1) and biopsy was taken from the lesion. Intense vascular structures, increased inflammation and fibroblastic proliferation were observed on pathological evaluation. Computed tomography showed a polyploidy mass (5.5 cm x 3.9 cm x 3.7 cm) in the greater curvature of the stomach (Figure 2). Laparotomy was decided. On laparotomy, intraluminal mass (6 cm x 4 cm) in the greater curvature of the stomach was palpated. Polypoid mass was reached through gastronomy. Macroscopically well-defined, solitary mass was removed via gastric wedge resection (Figure 3 and 4). Histopathologically fusiform and plasma cells was observed (Figure 5 and 6). It was reported as inflammatory myofibroblastic tumor. The patient was discharged from the hospital on the tenth postoperative day. Radiograph of esophagus, stomach and duodenum was taken on the ninth-postoperative month (Figure 7) and showed no pathological findings. The patient was followed-up via ultrasonography for two years and no recurrence was observed.

Discussion

Inflammatory Myofibroblastic Tumor (IMT) in stomach is extremely rare in childhood. The most frequent symptoms of gastric masses include abdominal pain, weight loss, anorexia,

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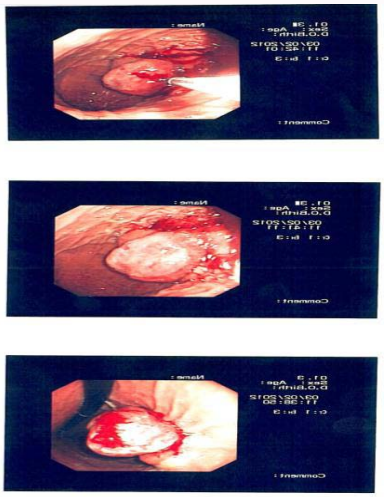

Figure 1: On gastroduoendoscopy, polypoid mass was detected in the gastric corpus and biopsied. Examination of the mass revealed intense vascular structures, increased inflammation and fibroblastic proliferation.

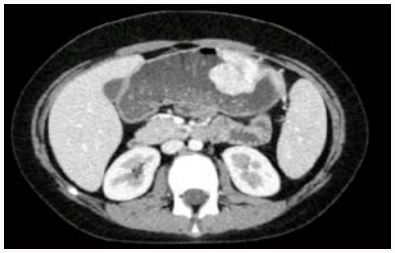

Figure 2: Abdominal computed tomography; mass (5.5 cm x 3.9 cm x 3.7 cm) protruding into the lumen of the stomach in the greater curvature of the stomach.

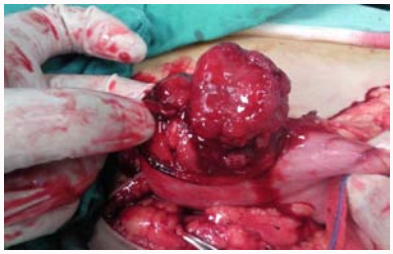

Figure 3: On laparotomy; intraluminal polypoid mass (4 cm x 5 cm) in the greater curvature of the stomach.

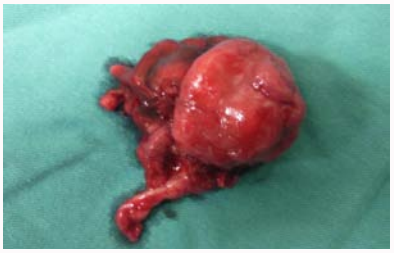

Figure 4: The mass and the gastric tissue were excised via wedge resection.

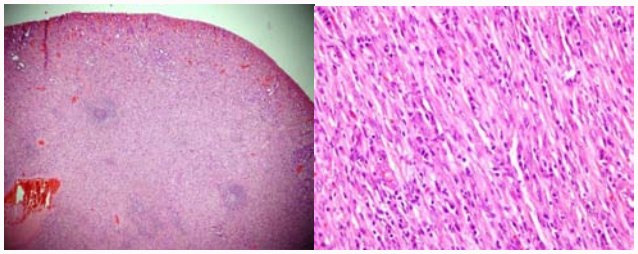

Figure 5 and 6: Histological appearance of the polypoid mass (4x, H&E), fusiform and plasma cells (40x, H&E).

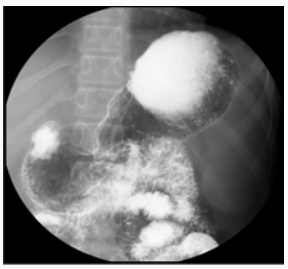

Figure 7: Postoperative 6th-month radiograph of esophagus, stomach and duodenum.

epigastric burning [2,5,7]. In our case, the patient was admitted to the clinic with the complaint of bloody vomiting and a history of anorexia and epigastric pain. IMT is a rare tumor characterized by unsteady proliferation of inflammatory cells. These lesions are also called as inflammatory pseudo tumor, fibrous xanthoma, plasma cell granuloma, pseudo sarcoma, lymphoid hamartoma, myxoid hamartoma, inflammatory myofibrohistiocytic proliferation and benign myofibroblastoma. Due to its rarity, the pathogenesis and treatment are still controversial. Main cell is known to be the myofibroblast in the development of pseudo tumor [1,7,8]. They are more frequent in children and young adults [2,3]. Although lung involvement is more common, airway, gastrointestinal tract, pancreas, tonsil, liver, gall bladder, duodenum, kidney, bladder, mesentery, spleen and epididymal involvement have also been reported [1,9-12]. Extra pulmonary involvement of IMT is more frequent in the first two decades of life [7]. Although the pathogenesis of IMT is not clear, there are publications asserting that it result from infections, multifactorial, vascular disorders, immune system disorders [8,9]. Pulmonary involvement of IMT has been attributed to pneumonia agents whereas infections such as gram-positive cocci and E Coli have been considered to be the cause of liver involvement [12,13]. In the etiology of inflammatory myofibroblastic tumor (IMT) of the stomach, helicobacter pylori and EBV virus infection were investigated but no effect was reported [5]. Although inflammatory myofibroblastic tumor (IMT) is considered as a benign lesion, there are cases in the literature which display recurrence, local invasion and distant metastasis [3,10,14]. Considering the literature are IMT of the stomach has a low risk of metastasis [2,4,14]. In our literature review, we have encountered limited number of gastrointestinal involvement in children and our case is the eighteenth pediatric patient presenting gastric inflammatory tumor. Localization of the lesion varies; cardia, antrum and pylorus localized cases have been identified. As in other gastric tumors, the most common causes of admission to the hospital are weight loss, anemia, epigastric pain, vomiting, anorexia, and upper gastrointestinal bleeding, as in our patient. The size of IMT of the stomach varies between 3 cm and 10 cm [5]. In cases presenting stomach localization of IMT, establishing diagnosis via preoperative radiological imaging is difficult. Radiological imaging does not indicate specific diagnostic findings. They usually appear as well-defined, hard masses. In patients with postoperative

histopathological diagnosis, radiological imaging may be useful to follow-up other organ involvement and recurrence. Pathologic evaluation is also important in differential diagnosis [13,14]. In our case, the boundaries of mass lesion were clearly imaged and excision was performed. The size of the extracted mass was 6 cm x 4 cm. No other organ involvement was observed on CT examination. Although conservative follow-up has been considered as an alternative treatment option due to the presence of spontaneously regressing cases, in cases presenting recurrence or local invasion, surgical excision of the mass is preferred. Steroids and chemotherapy options can be applied in cases presenting recurrence or incomplete resection [1,8,10]. Because they are nodular and solid masses and they invade the tissue which they originate from, inflammatory myofibroblastic tumors are difficult to distinguish from malignancy. Therefore, the surgical approach may be more radical. Because the recurrence rate is very low in cases where the whole mass is removed, we can conclude that the treatment is administered when the pathological diagnosis is made. In our case, because the macroscopic appearance of the mass is well-defined, and the whole mass is completely removed and surgical margins are clean on microscopic examination and there is no evidence of metastases on radiological evaluation, the patient did not receive add-on-therapy. In a study evaluating 13 adult cases with IMT of the stomach, the initial complaint in majority of the patients was abdominal pain and mass size ranged from 1.5 cm to 10 cm. All patients underwent surgery and additional treatment was not administered. No pathology was observed during postoperative follow-up except recurrence in one patient and peritoneal invasion [7].

While recurrence was not observed after total excision in one pediatric case with IMT of the stomach, gastric tumor extending esophagus and meditational mass extending to the left pulmonary hilar were observed in the second case during four-year follow-up. The patient received chemotherapy after surgery but the tumor spread to the abdomen and right lung. The patient died 11 months after diagnosis [5].

Conclusion

IMT should also be considered in children. In most cases, complete surgical resection of the mass is sufficient for treatment. However, patients should be followed-up due to the postoperative risk of recurrence, local invasion and distant metastasis.

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