Gastric Inflammatory Fibroid Polyp in Children

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ABSTRACT

Inflammatory fibroid polyp (IFP) constitutes as a chronic inflammatory lesion that is generally located in the gastric (gastric inflammatory fibroid polyp). The lesion originates from the submucosal tissue of the gastrointestinal tract and progress into a polypoid mass. Inflammatory fibroid polyp is rarely found in children and the cause is not definitely known. A case of a four-year old boy with recurrent paleness for 3 months has been reported. At the beginning, the patient was suspected of haemolytic anaemia. Except the hemoglobin level (< 5 g/dL), other laboratorium examination did not support the working diagnosis. During the last one months period, the patient also complained of recurrent abdominal pain and vomiting. On the abdominal ultrasonography (USG) and endoscopy, a mass that covered most of the gastric lumen was detected. Based on that finding, gastrectomy and mass removal were conducted. The pathologic anatomy examination proved the presence of an inflammatory fibrous polypoid. Follow-up untill 12 months showed no any complaint and abnormalities on endoscopic examination. Since there was no consensus regarding the follow-up period following the procedure, the evaluation of this patient would be conducted every 2-3 years.

Keywords: *inflammatory fibroid polyp, tumor, gastric, children*

INTRODUCTION

Inflammatory fibroid polyp (IFP) constitutes as a chronic inflammatory lesion that is rarely encountered in children and it is generally located in the gastric (gastric inflammatory fibroid polyp).[1-3] The lesion originates from the submucosal tissue progress into a polypoid mass.[4-9] The cause of IFP is not definitely known and various factors are suspected of triggering the onset. The prevalence of IFP was very low and only scattered cases have been reported in the literature.[10-13]

The diagnosis of IFP is quite difficult since the profile of this lesion is not specific clinically, endoscopically, and on the imaging findings. The definite diagnosis of IFP is based on the results of histopathological examination.[14-21] This anomaly can lead to gastrointestinal tract (GIT) complications such as abdominal pain, GIT obstruction or intususception.[3,22] Most polyps are small in size so that they can be removed by endoscopic polypectomy, but in other cases they can grow into big masses (tumors) so that surgery is required.[23-32]

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CASE

A boy, four years of age was referred by the Division of Hematology-Oncology, Cipto Mangunkusumo hospital with the chief complaint of gastric tumor. From the history of illness, patient had been monitored by Division of Hematology-Oncology because of recurrent pale since 3 months before admission. During that period, patient was suspected of suffering from hemolytic anemia. Some laboratory examination were done to prove the working diagnosis. The HbF level was 0.4% (normal), HbA2 0.6% (normal), and hemoglobin analysis revealed HbA1F. The bone marrow aspiration examination showed erithropoesis hyperactive whereas the Coombs test revealed blood type B rhesus positive, no sign of in-vivo sensitization and neither free antibodies/ irregular antibodies in the serum. There had been no bleeding detected in oral cavity, nose, gingivae, anus as well as bluish spots on the skin. He also suffered from intermittent fever. No complaint on defecation and micturation.

Two months later, the patient complained of recurrent abdominal pain, nausea, and vomiting.

Two weeks after, he also noticed a lump on his upper belly and it was somewhat distended after meals. The abdominal ultrasonography (USG) was done, and showed a thickening of a part of the gastric lining and a pylorus in the form of a soft tissue mass causing the narrowing of gastric outlet. Based on the laboratory and USG examination, the diagnosis of hemolytic anemia was excluded. Anemia of this patient might be caused by occult bleeding from the tumors. The patient was then referred to Gastro-Hepatology Division for further investigation.

The patient was the fourth out of the four siblings and there was no similar symptoms in other brothers, sister, or parents. There was no also history of cosanguity in the parents married.

On the physical examination, the patient looked conscious, pale, and had neither dyspnea nor cyanosis. Normal vital sign. His body weight (BW) was 11.8 kg, body height (BH) 97.5 cm. The nutritional status based on BW/BH was 78.7% (<P_5 NHCHS). Heart and lungs were normal. The abdomen was tender, with a painless well-defined mass of 17 x 8 x 1 cm in the epigastric area. The turgor was adequate, the liver and spleen were no palpable, ascites was absent and bowel sound was normal. No lymph node enlargement. No abnormality on extremities.

Endoscopic examination was done and showed a reflux of the gastric fluid into the esophagus and the gastric was filled with milk which received ten hours before. After aspiration of the gastric fluid, the profile of eroded esophagus and proximal gastric corpus was seen as well as a mass on the lining of the distal gastric corpus that almost filled up the whole gastric lumen. The mass surface was lumpy, eroded, and not ulcerated. The endoscopic tip could not be further inserted owing to the narrowed lumen caused by the mass obstruction. The impression then was a gastric tumor that caused obstruction and gastro-esophagitis. Considering that the mass had almost caused total obstruction, the patient was prepared for the surgical removal of the tumor and pathologic-anatomy examination from the removed tumor tissue.

During the operation, two extra-luminal tumors were spotted from the gastric corpus to the pylorus (each 4 x 3 x 3 cm in size) and 1 intra-luminal tumors was palpated from the pylorus to the corpus with the size 7 x 6 x 3 cm. At that point, a subtotal gastrectomy was decided on. The removed tumor tissue was then sent to The Department of Pathologic Anatomy with the result relevant to the profile of inflammatory fibrous lesion/polypoid and there was no sign of malignancy. In the post-operative care, the general condition of the patient was good and complications were not found. He weaned off the liquid food to solid food. Patients also received omeprazole 1 x 10 mg for the esophagitis and gastritis. One week and one month after hospitalization, the general condition was good, no vomiting as well as a lump in the abdomen. The body weight increased to 13 kg. Three month after hospitalization, endoscopic re-evaluation was performed and showed no abnormalities in the esophagus, gastric or duodenum mucosa. After one year clinical evaluation, there was no signs of recurrence.

DISCUSSION

Inflammatory fibroid polyp constitutes a chronic inflammatory lesion that can manifest in all parts of the gastrointestinal tract (GIT), but it is most frequently found in the antrum (80%).[8,11,16,17] Nevertheless, in some cases, it is also encountered in the ileum and colon.[9,14,15,19,29] In a retrospective review on gastric polyps, 3-4% of the cases were IFP.[13] Another literature review by Chongsrisawat et al reported six IFP cases in 4-8 years old children from 1966 until 2004 with the locations of the lesions in the rectum, colon, ileum and jejunum in one child respectively whereas two children were found to have the lesions in the gastric. This lesion originated from the submucosa of the GIT that contains a lot of fibrotic tissue, highly vascularized connective tissue with a great number of eosinophils. The resultant polyp is usually small in size and could be either pedunculated or non-pedunculated.[4,5,6] IFP can be encountered at any age. Nevertheless, it is rarely found in children and is more frequently found in the 6-7 decades. IFP is more frequently spotted in females than males (1.6 : 1).[4,9]

The patient which reported here is the first case in The Department of Pediatric of University Indonesia, Cipto Mangunkusumo Hospital Jakarta. The patient was 4 years old with the different shape and location of mass, unlike what has been reported in literature. The mass was of considerable size that does not resemble a polyp and it was found from the corpus to the gastric pylorus.

IFP clinical symptoms are various depending on the location and size of the polyp. Most IFP are small in size and asymptomatic. Big polyps could lead to ulceration and obstruction of the GIT. The location of gastric inflammatory fibrous polypoid is generally at the antrum causing gastric outlet obstruction with such clinical symptoms as abdominal distension, nausea, quick sensation of fullness and vomiting.[3] In some cases, clinical symptoms such as recurrent epigastric pain and bleeding of the GIT secondary to erosion/ulceration of the polyp or gastric mucosa could also be found.[1,3,11] Another clinical sign that might be manifested is body weight loss owing to the lack of food and fluid intake.[10] Approximately 37% of the cause of gastric outlet obstruction is benign gastric tumor.[3,11] Fever has ever

been reported in IFP patients. This condition is correlated with the inflammatory reaction that occurs in IFP; this tumor is suspected of releasing cytokines that can cause fever.[4]

The initial clinical symptom of merely anemia without bleeding and other clinical signs in this case is not typical for gastric tumors. So why the patient once was suspected of suffering from hemolytic anemia but in the clinical monitoring and by further supporting examination it was proven otherwise. The suspicion for gastric tumor rose on the patient's presentation with the complaint of a lump in his belly accompanied by the clinical symptoms of GIT obstruction and it was proven by the findings of the abdominal USG and upper GIT endoscopy. The anemia of the patient could be attributed to the occult bleeding that occurred chronically due to occult bleeding of the polyp or gastric mucosa.

This kind of bleeding is not conspicuous but an occult blood fecal test is required. That examination was not performed on this patient because GIT bleeding was not suspected by the Hemato-oncologist at that time. Despite the hemoglobin level, the serum iron level and iron binding capacity needed to be assessed at the first hospitalization due to the chronic bleeding at that condition could cause iron-deficiency anemia. The intermittent fever in this patient might be a clinical symptom of an inflammatory process.

All layers of the GIT have the potential for tumor growth. In benign gastric tumors, 40% of the lesions are located in the mucosa, 40% in the muscular layers, and the rest are vascular, endocrinal or neurogenic tumors.[1] Various types of tumors are encountered in the gastric among, such as hyperplastic polyps, adenomatous polyps, fundic gland polyps, inflammatory fibroid polyps, juvenile polyps, familial polyposis syndromes, Peutz-Jeghers syndrome, non-mucosal intramural tumors, leiomyoma, fibroma and fibromyoma, lipoma, neurogenic, vascular tumors, cystic tumor, and mucocele.[1] The type of tumor in this patient was inflammatory fibroid polyp.

The etiology and pathogenesis of IFP have not been definitely elucidated up to now.[4] Some hypotheses have been proposed to explain about it. Irritants (chemical, mechanic and biologic) are regarded as the substances destroying the mucosa of the GIT and stimulating polyp formation. Infection has also been reported as the cause of IFP and in some cases it has been correlated with *Helycobacter pylori* infection and parasytes.[4] In other reports, IFP is correlated with such autoimmune diseases as sarcoidosis, rheumatoid arthritis and ankylosing spondylitis.[16-18] In this patient, the cause of IFP has been difficult to be proven. The role of *Helicobacter pylori* infection has been disproved.

The suspicion of a tumor can be advocated by various supporting examination, such as abdominal plain photo, USG, CT-scan, MRI or endoscopy. They can help prove the presence of a mass in the GIT; however they cannot differentiate the types of mass.[22,30,32] In this patient, the abdominal USG examination delineated thickening of part of the gastric lining and pylorus in the form of a soft tissue mass causing the outlet narrowing from the gastric into the duodenum. These findings were supported by a barium meal examination. Since USG examination had already yielded clear information, CT-scan or MRI were not needed.

The definite diagnosis was established based on the histopathological examination. Based on this fact, the biopsy technique to have the right tissue sample should be accurate. It has been reported that accurate diagnosis can not be made if the tissue sample is obtained through endoscopic biopsy it might be caused by the location of the lesion that is between the submucosa and the mucosal bed. That is the reason for taking a sample by endoscopic polypectomy (small polyps) or surgery (big polyps/ polypoid masses) for histopathological examination.[4]

The main therapy for IFP is removal of the polyps/ masses because IFP has the potential to grow and cause obstruction. Polyp removal is performed by incising big polyps/masses or by endoscopic polypectomy for small polyps. It has been reported about the recurrence of IFP following endoscopic polypectomy whereas it has not been found if the polyps are removed surgically.[5,27] In circumstances in which endoscopic polypectomy is the choice, it is suggested that endoscopic examination be conducted periodically to detect the recurrence.[4,5,23] In this patient, the mass size was very bulky so that the removal of the polyp mass was performed by surgery followed with gastrectomy. Buyske et al[24] reported the use of endoscopic surgery in the removal of polyps with < 6 cm of size and the recurrence was not found during three years observation following the procedure.[24] Ke et al, reported the use of laparoscopic polypectomy to remove submucosal polyps located on the posterior of the gastric fundus.[25] Recently, a new technique has been developed, namely endoscopic-USG.[26] Considering that the IFP of t he patient was non-neoplasmic and there has been not yet agreement among the experts regarding the follow-up period following the procedure, so that the evaluation of recurrence in this patient would be conducted every 2-3 years.

CONCLUSION

Gastric inflammatory fibroid polyp can occur in young children with non-specific clinical manifestation. Removal tumor by surgery was the treatment of choice

for larger lesion and gave a satisfactory result without recurrence in one year follow up. Further follow up is still needed to evaluate the recurrence.

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