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Concurrent Eosinophilic Cholangiopathy and Pancreatitis in a Child Due To Eosinophilic Gastroenteritis: A Rare Entity

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Abstract

Background: Eosinophilic gastroenteritis is an increasingly recognised condition with variable presentations. Eosinophilc involvement of the biliary tract and the pancreas is extremely rare and usually occurs in the setting of idiopathic hypereosinophilic syndrome.

Case report: We report a case of 12 years old male child who was receiving steroids after a diagnosis of eosinophilic gastroenteritis related ileocaecal obstruction. While the dose of steroids was tapered, he developed abdominal pain and jaundice with elevation in absolute eosinophil count. The diagnosis of eosinophilic pancreatitis with cholangiopathy was eventually established and the patient improved with steroids and biliary stenting.

Conclusion: We report this case for its rarity and the unusual presentation as concurrent cholangiopathy and pancreatitis in a child and were preceded by eosinophilic gastroenteritis with intestinal obstruction.

Keywords: Eosinophilic gastroenteritis; Pancreatitis; Cholangiopathy; Child

Introduction

Eosinophilic gastroenteritis (EG) is the abnormal accumulation of eosinophils in the gastrointestinal tract (GIT) which may result in a myriad of presentations. While luminal involvement is now well recognised in EG, the involvement of biliary and pancreatic systems is reported less frequently. Also, the concomitant involvement of the GIT, the bile duct and the pancreas has been reported only rarely [1]. We present the case of a 12 years old male child with EG causing intestinal obstruction who presented with eosinophilic cholangiopathy and pancreatitis while reduction in dose of steroid was attempted.

Case Report

A 12-years old male child presented to with abdominal pain and jaundice for a week. The pain was epigastric, radiating to the back and was associated with non-bilious vomiting at its onset. Around the same time the patient also noted yellowish discoloration of his sclera and passage of dark urine but no preceding prodrome, associated pruritus or clay coloured stools. He had a previous history of acute intestinal obstruction and underwent an emergent surgery two years back. Intraoperatively an ileocaecal mass was detected and patient underwent right hemicolectomy and ileocaecal anastomosis. The histopathology revealed eosinophilic infiltration of the caecum and resected lymphnodes were reactive. Further evaluation revealed multiple axillary, cervical and abdominal lymphnodes and elevated absolute eosinophil count (AEC; 1880/μL). Serum IgE levels were elevated (>10,000 IU/mL). The workup for cause of eosinophilia was non-contributory and the counts did not improve with diethylcarbamazine. The patient was initiated prednisone 1.5 mg/kg body weight for 8 weeks with subsequent reduction in eosinophil counts and reduction of lymphadenopathy.

Patient became symptomatic when reduction in steroids was attempted with an increase in AEC from 113 to 3500/μL associated with pain and jaundice. Laboratory evaluation revealed serum amylase 480 U/L (normal 0-70 U/L), total bilirubin: 6.5 mg/dL (normal 0-1 mg/dL) with conjugated bilirubin 5 mg/dL (normal 0-0.3 mg/dL), aspartate aminotransferase 53 IU/L (normal 2-40 IU/L), alanine aminotransferase 95 IU/L (normal 2-41 IU/L), alkaline phosphatase 480 IU/L (normal 42-128 IU/L), albumin 4.1 g/dL (normal 3.4-4.8 g/dL), total protein 7.5 g/dL (normal 6.4-8.3 g/dL), prothrombin time 9.8 s (normal 9.0-11.5 s), international normalized ratio 1.0, and activated partial thromboplastin time 26 s (normal 22-34 s). Stools were negative for cysts, ova and parasites. Contrast enhanced computed tomography (CECT) was suggestive of dilated intrahepatic biliary radicles and dilated common bile duct (CBD) with thickened walls of the CBD (Figure 1A). The pancreatic head was bulky (Figure 1B). Magnetic resonance cholangiopancreatography (MRCP) revealed dilated lower-mid CBD with narrowed lower and proximal CBD (Figure 1C). For relief of the obstruction, endoscopic retrograde cholangiopancreatography (ERCP) was undertaken, infiltrated duodenal mucosa was visualised and the MRCP findings were confirmed (Figure 1D). Biliary decompression was undertaken with insertion of a 7 French biliary double-pigtail stent with relief in jaundice. During ERCP brushing from distal CBD was performed and cytological examination revealed eosinophilic infiltrates apart from bile ductular cells. The steroids were escalated to 2 mg/kg with reduction in AEC to 195/μL. The child was doing well on follow-up but unfortunately while returning to home after follow-up consultation he met road-side traffic accident and died on spot.

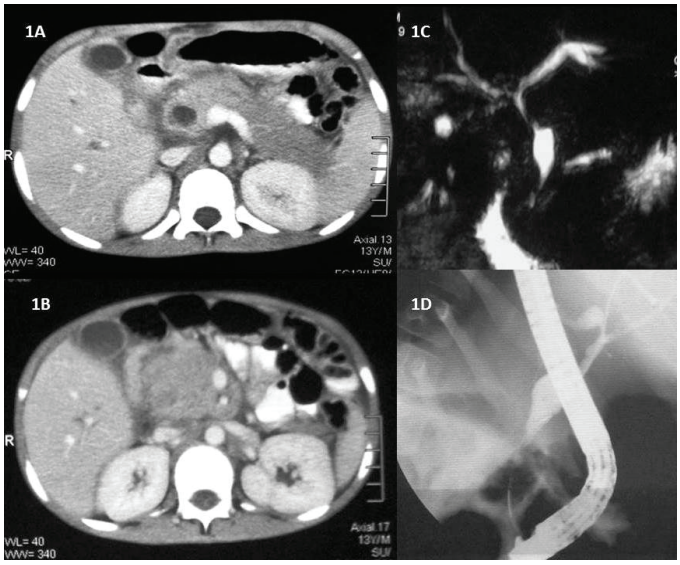

Figure 1: (1A) CECT showing dilated intra hepatic biliary radicles and dilated CBD with thickened wall;
(1B) CECT is showing bulky pancreatic head;
(1C) MRCP image showing dilated lower-mid CBD with narrowed lower and proximal CBD;
(1D) ERCP: Fluoroscopic image also showing dilated lower-mid CBD with narrowed lower and proximal CBD. Endoscope is visible.

Discussion

Eosinophilic infiltration of tissues is common in idiopathic hypereosinophilic syndromes (IHES) as was noted in our patient [2]. Involvement of GIT has been noted in 14-53% of cases of IHES while hepatic involvement is noted in around 30% of patients [3]. Eosinophilic involvement of the GIT usually manifests as involvement of the luminal organs [4]. Involvement of the biliary and pancreatic system is unusual. In the present case, we report a paediatric patient with combined involvement of the biliary system and the pancreas which was managed with biliary stenting and therapy targeted at reducing the eosinophil count. Eosinophilc cholangiopathy is an uncommon condition which is confused with more common biliary diseases like cholangiocarcinoma, primary sclerosing cholangitis, autoimmune (IgG4 related) sclerosing cholangitis etc [4]. The commonest presentation is pain abdomen and jaundice. Two-thirds of the patients have associated peripheral eosinophilia as in the index case. The management includes use of steroids and surgery in some patients [4]. Earlier, eosinophilic cholangiopathy was reported in a 13 years old child but concurrent eosinophilic pancreatitis in paediatric age has not been reported in the literature [5,6]. Although eosinophilia may occur in relation to any pancreatitis, eosinophilic pancreatitis is usually the result of infiltration of the pancreatic tissue with eosinophils resulting in pancreatic injury [7]. Eosinophilic pancreatitis usually manifests in the form of a pancreatic mass and may present with jaundice and abdominal pain [8]. On occasion the presentation may be in the form of focal pancreatic involvement without an elevated peripheral eosinophil count [9].

In conclusion, eosinophilic cholangiopathy and pancreatitis are rare conditions which manifest with abdominal pain and jaundice. The clue to the diagnosis usually comes from an elevated AEC. In cases where a diagnosis is made early, surgical interventions can be avoided as the response to steroid is excellent. This case also highlights that caution needs to be exercised while tapering steroids as an increase in AEC may result in clinical manifestations.

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