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Case Report Open Access

Intra-abdominal Xanthogranulomatoses Secondary to Postoperative Bile Leak

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Abstract

Xanthogranuloma is one of the 2 disorders of non-Langerhan cell histocytoses. It is a benign tumor histologically characterized by foamy histiocytes, numerous Touton giant cells and rare eosinophils. The histology differs from Langerhan cell histocytosis disorders in which convoluted histocytes, numerous eosinophils and rare Touton cells are seen. Grossly, the lesion appears as golden yellow papules or nodules. Electron microscope characteristics include presence of cytoplasmic lipid. The etio-pathogenesis is not fully understood, however granulomatous reaction of histiocytes to an unknown stimulus is postulated. Here, we discuss a rare presentation of peritoneal xanthogranulomatoses in a patient caused due to postoperative bile leak.

Keywords: Bile leak; Xanthogranuloma

Abbreviations

CT: Computed Tomography; ERCP: Endoscopic Retrograde Cholangiopancreatography; HIDA: Hepatobiliary Iminodiacetic Acid; JXG: Juvenile Xanthogranuloma; MRI: Magnetic Resonance Imaging

Introduction

Xanthogranuloma is a rare benign tumor characterized macroscopically by golden yellow nodules and microscopically as multicentric, mass-like accumulation of non-Langerhans lipid-laden histiocytes [1]. The etio-pathogenesis is not fully understood, however granulomatous reaction of histiocytes to an unknown stimulus is postulated [2]. Here, we discuss a rare presentation of peritoneal xanthogranulomatoses in a patient caused due to postoperative bile leak.

Case Presentation

A 71 year old female patient with past medical history of hypertension, hyperlipidemia and hypothyroidism was referred to our facility for persistent abdominal pain for past 6 months. The pain was associated with poor appetite and progressive loss of weight. On further questioning, it was revealed that she had undergone laparoscopic cholecystectomy for acute cholecystitis prior to the onset of these symptoms. Postoperative course was complicated by bile leak which was diagnosed by hepatobiliary iminodiacetic acid (HIDA) scan. Subsequently, she underwent endoscopic retrograde cholangiopancreatography (ERCP) with sphincterotomy with resolution of bile leak. No stents were placed. For the persistent abdominal pain, the patient underwent a computed tomography (CT) scan of abdomen which showed 1.7 cm nodule in the omentum surrounding the gastric antrum. In addition, she also had multiple peritoneal nodules surrounding the liver. To further delineate these lesions, she underwent magnetic resonance imaging (MRI) of the abdomen which showed similar lesions suspicious for peritoneal carcinomatosis (Figure 1). The patient underwent CT guided biopsy of the lesions. Pathology revealed mixed inflammatory cell infiltrate composed of histiocytes, lymphocytes, plasma cells, eosinophils and giant cells suggestive of xanthogranulomatous inflammation. The giant cells contained bile pigment which suggested that the most likely etiology was postoperative bile leak. Further, a diagnostic laparoscopy was done. Intraoperative findings included yellowish 1-2 cm masses in omentum near the gallbladder fossa and greater curvature of stomach. Surgical pathology confirmed the diagnosis of xanthogranulomatous with bile pigment deposits.

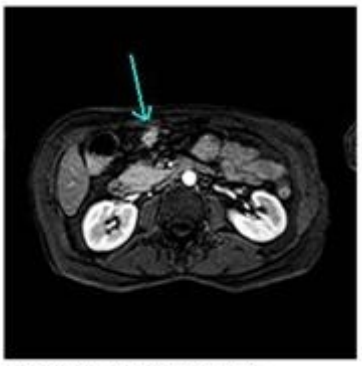

Figure 1: MRI showing peritoneal nodule.

Discussion

Xanthogranuloma is one of the varaints of non-Langerhan cell histocytoses [3]. It is a benign tumor histologically characterized by foamy histiocytes, numerous Touton giant cells and rare eosinophils [4]. The histology differs from Langerhan cell histocytosis disorders in which convoluted histocytes, numerous eosinophils and rare Touton cells are seen. [5] Grossly, the lesion appears as golden yellow papules or nodules [6]. Electron microscope characteristics include presence of cytoplasmic lipid [7]. Further, Birbeck granules characteristic for Langerhans cells are absent [7]. When xanthogranuloma affects

multiple organs; it is called Erdheim-Chester disease, which occurs more commonly in men than women, with a peak incidence in the sixth decade of life [8]. Xanthogranuloma mainly affects juveniles and occasionally adults [9]. Etiopathogenesis is poorly understood. The rarity of the disease has led to the paucity of literature regarding the etio-pathogenesis of the disease [10]. Juvenile xanthogranuloma (JXG) presents as single skin lesion or multiple skin lesions [10]. Occasionally JXG may affect visceral organs [11]. Skin lesions are usually self-limiting and may spontaneously resolve. However, adult form of the disease may not resolve spontaneously [12]. Non-resolving severe cutaneous adult form may require excision or symptomatic treatment [12].

A variant of xanthogranulomatous inflammation called as necrobiotic xanthogranuloma has a chronic, indolent, and progressive course with predilection for periorbital region, trunk and extremities [13]. It is commonly associated with monoclonal gammopathy and multiple myeloma. Chemotherapy and systemic steroids are used to alter the course of this disease [14].

Occasionally, xanthogranulomatous inflammation may affect visceral organs [15]. Although, the etio-pathogenesis is still unclear, infectious or inflammatory process or abnormalities in lipid metabolism or physical insult are thought to be inciting factors.[1] Few cases of cholecystitis and pyelonephritis due to xanthogranulomatous inflammation has been reported in the literature [15,16]. Further, one report described xanthogranulomatous panniculitis in a patient after laparoscopic cholecystectomy attributed to gall stones [17]. Usually benign, co-existence with malignancy has been reported [18]. Additionally, xanthogranulomatous inflammation of the sigmoid colon and urinary bladder has also been reported in the literature [19,20]. However, literature on xanthogranulomatous inflammation is either case reports or case studies. Furthermore, till date, no definitive etiopathogenesis has been explored.

Our case report describes xanthogranulomas distributed in the peritoneal cavity due to post-operative bile leak. The history of laparoscopic cholecystectomy prior to the onset of the symptoms were key in identifying the etiology of this case. Further, histology of the nodules revealed bile pigments which further supported our assumption. We opted to do laparoscopic surgery and removal of the tumor due to the persistence of her symptoms. Long term follow up may be needed to see if the patient had recurrence of symptoms. Though excision could be the treatment of choice, currently no definitive or alternative treatment plan is available in the literature.

In conclusion, intra-abdominal xanthogranuloma is an extremely uncommon condition, the etiopathogenesis of which is not yet known. Given the presence of bile pigments in the histology, post-operative bile leak was the inciting factor for this patient. Further, though rare, intra-abdominal xanthogranuloma secondary to postoperative bile leak should be considered as one of the differentials while managing patients with complains of abdominal pain post cholecystectomy. Also, those patients may present with symptoms months after the procedure. Hence, a good history is key in diagnosing these patients.

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