Xanthogranulomatous Cholecystitis: A Case Report

Ksantogranülomatöz Kolesistit: Bir Vaka Sunumu

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Abstract

Xanthogranulomatous cholecystitis (XGC) is a rare variant of chronic cholecystitis that occurs as the result of destruction of the mucosa of the gallbladder and infiltration of macrophages inside the gallbladder wall. It was first described in 1970 as ''fibroxanthogranulomatous cholecystitis'' by Christensen and Ishak. The major radiologic finding of XGC is thickening of the gallbladder wall (>4mm). Importance of this disease lies in the fact that as it shows neither clinical nor radiologic unique feature to be differentiated from gallbladder carcinoma, it may lead to unnecessary enlarged resections due surgeries. The final diagnosis is usually made during microscopic examination in pathology. This case report describes clinical, radiologic and pathologic findings of a patient diagnosed with XGC. . **(Sakarya Med J 2019, 9(3):554-557)**

Keywords xanthogranulomatous cholecystitis; cholelithiasis; gallbladder

Öz

*Ksantogranülomatöz kolesistit (KGK) ender görülen ve iyi huylu kronik bir safra kesesi enflamasyonu olup, genelde mukoza zedelenmesi sonrasında makrofajların safra kesesi duvarına infiltrasyon göstermesi sonucu meydana gelir. İlk olarak 1970 yılında 'fibroksantogranülomatöz kolesistit' olarak Christenser ve Ishak tarafından tanımlanmıştır. Radyolojik olarak safra kesesinde duvar kalınlaşması (>4 mm) görülmesi nedeniyle, safra kesesi tümörlerini yapı ve morfolojik görüntüsü ile taklit etmesi açısından tanısı önem taşımaktadır. KGK'e spesifik bir radyolojik veya klinik bulgu bulunamadığı için cerrahi işlemler esnasında gereksiz ve genişletilmiş organ rezeksyonları yapılmaktadır. Kesin tanı patolojide mikroskopik inceleme sonucu ortaya konur. Bu olgu sunumunda KGK tanısı almış bir vakanın klinik, radyolojik ve patolojik bulguları sulumuştur. **(Sakarya Tıp Dergisi 2019, 9(3):554-557)***

Anahtar kelimeler *ksantogranulomatoz kolesistit; kolelityazis; safra kesesi*

INTRODUCTION

Xanthogranulomatous cholecystitis (XGC) also known as cholecystic granuloma, is an uncommon variation form of chronic cholecystitis.[1] It is characterized by infiltration of foamy cells inside the gallbladder wall, often as a result of the rupture of Rokitansky–Aschoff sinuses that leads to destructive fibrosis and thickening of gallbladder wall. As this benign lesion age, it becomes densely fibrotic so that it may be confused with cancers of the gallbladder.[2] Sometimes XGC also shows a tendency to adhere to adjacent organs or even form fistulas.[3] It causes difficulties during cholecystectomy and most of the patients are misdiagnosed preoperatively as having gallbladder carcinoma. Some extreme surgeries, such as segmental resection of liver and pancreaticoduodenectomy, may be avoided if a correct preoperative diagnosis of XGC may be done. The final diagnosis is usually made by pathological examination after cholecystectomy.[4]

This case report describes clinical and pathological findings in one patient diagnosed with XGC.

CASE REPORT

A 32-year-old woman with one-month history of abdominal pain, associated with abdominal swelling and vomiting, has been admitted to our surgery polyclinic. Laboratory examinations show an increase in the levels of CRP and GGT. Abdominal ultrasound revealed features of chronic cholecystitis characterized by thickness of the gallbladder wall (7mm) and 4 gallstones found inside the gallbladder (largest one with 2.2 cm in dimension). Laparoscopic cholecystectomy has been performed and due to operation, gallbladder was found to be stiff and edematous. As the gallbladder wall was found to be thick, it was sent to pathology department for further macroscopic and microscopic examinations. Due to macroscopic examination, gallbladder was found to be 8x6 cm in dimensions; after opening of gallbladder, wall thickness was 1.6 cm and inside of gallbladder 4 gallstone were found the largest one with 2.2 cm in dimension. Microscopic examination of gallbladder wall revealed multiple mucosal ulcers and infiltration of polymorphonuclear type inflammatory cells inside the gallbladder wall. Pigment laden macrophages infiltrations and fibroblastic proliferation were also seen (Figures 1 and 2). To support the XGC diagnose, CD68 and pancytokeratine immunohistochemical analyses were done. CD68 stain was expressed positive in macrophages, while pancytokeratine stain was not (Figure 3). As the result of the pathological examinations, XGC diagnose has been confirmed.

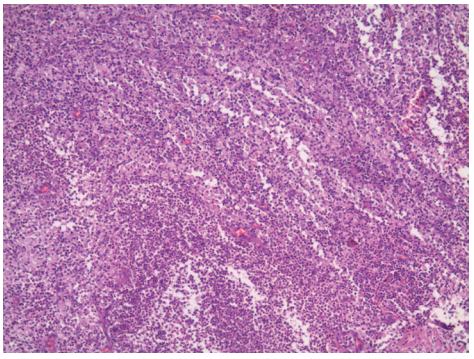

Figure 1: Pathologic view of the gallbladder wall and polymorphonuclear leukocyte infiltration inside the gallbladder wall (H&E-X100).

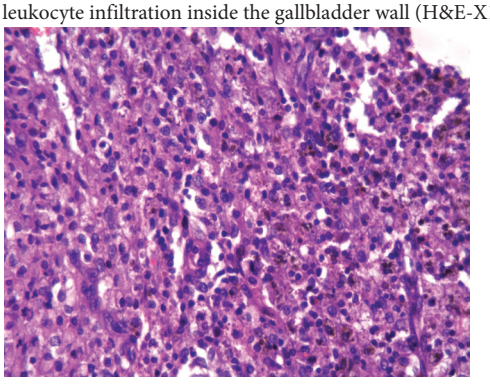

Figure 2: Pigment laden macrophages inside the gallbladder wall (H&E-X400).

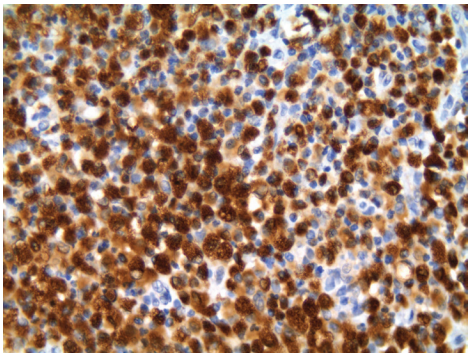

Figure 3: CD68 stained foamy histiocytes (CD68-X400).

DISCUSSION

XGC was first described by Christensen and Ishak in 1970 as "fibroxanthogranulomatous inflammation".[5] Its incidence remains low of all inflammatory diseases of gallbladder and may vary from 0.7% to 13.2%.[6] This benign disease is highly misdiagnosed as malign lesion because there is no specific symptom or any radiologic finding to differentiate this lesion from carcinomas of gallbladder. Though, CT findings as mucosal line or cholelithiasis, are more specific for XGC. While cholangiocarcinoma progress mostly asymptomatically, patients with XGC mostly have a positive Murphy's sign in acute phase. CEA and CA19-9 tumor markers may be found high in both malignancies and XGC.[4] Cholelithiasis and XGC correlation has been reported to be 70 % in literature.[7] In this case report, the major intraoperative findings include gallbladder wall thickening and cholelithiasis.

Proliferating cell nuclear antigen (PCNA), p53, and beta-catenin were studied for XGC, gallbladder cancer, chronic cholecystitis and cholelithiasis. P53 mutation and PCNA were present in 52% and 60% of gallbladder carcinoma and only 3% and 11% of XGC respectively. In chronic cholecystitis and cholelithiasis no mutations were detected. The inflammatory component of XGC does not show any evidence of premalignant condition.[1]

One of the major findings of XGC is diffuse or focal gallbladder wall thickening. Difficulties during laparoscopic cholecystectomy were reported in most of the surgical cases as occurred in our case's surgery.[7] Though, laparoscopic cholecystectomy can be successfully performed in majority of the cases with diffuse thick-walled gallbladder.[8]

In conclusion, XGC is a rare, uncommon destructive form of chronic cholecystitis and in most of the cases has to be differentiated from gallbladder carcinoma as it shows similar preoperative radiologic properties, but no association has been found. In this case the significant finding was cholelithiasis and XGC diagnose was confirmed due microscopic examination of the thickened gallbladder wall.

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