

# Pigment polyp in the common bile duct

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## ABSTRACT

We report here a case of a young female with sickle cell disease and a past history of open cholecystectomy 10 years ago presenting with intermittent upper abdominal pain and jaundice. Abdominal ultrasonography revealed a dilated biliary tree with the possibility of a stone in the proximal common bile duct, but cholangiopancreatography showed a polypoid lesion at the junction of the common hepatic and common bile ducts. Biopsy of the lesion was benign. She underwent bile duct exploration and excision of what proved to be a 'pigment' polyp arising from the cystic duct stump. This is the first reported case of a pigment polyp encountered in sickle cell disease population.

Saudi Med J 2003; Vol. 24 (7): 776-777

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Sickle cell disease (SCD) is prevalent in the Eastern province of the Kingdom of Saudi Arabia. The affected individuals are at an increased risk of developing pigmented gallstones due to repeated hemolysis. Such stones remain silent and rarely cause symptoms. However, in some patients it may cause biliary colic, cholecystitis and occasionally obstructive jaundice or pancreatitis. Cholesterol polyps have been described before<sup>1,2</sup> but there has been no previous report of pigmented polyps. We report here a pigment polyp arising from the cystic duct remnant of a cholecystectomized sickle cell disease patient. We believe this could be the first ever reported pigment polyp in the literature.

**Case Report.** A 26-year-old girl, who is known to have SCD, presented with a year history of upper abdominal pain that occurs intermittently and was compressing in nature and associated with jaundice, dark urine but no history of pale stools or itching. The pain was aggravated by fatty meals but improves with simple analgesia. She gave a past history of an open cholecystectomy 10 years ago and was taking folic acid tablets regularly. On examination, she was thin, pale and

jaundiced but the vital signs were normal. The abdomen was soft, non-tender and with no palpable masses or organomegaly. Routine blood investigations showed a low hemoglobin of 6.9g%, raised bilirubin (total: 2.25 mg/dl, direct: 1.57mg/dl), slightly raised alkaline phosphatase and lactate dehydrogenase, but normal transaminases. The hemoglobin electrophoresis showed hemoglobin (Hb) A 10.7%, HbA<sup>2</sup> 2.8% and HbS 86.5%. Ultrasonography (US) showed a dilated common hepatic duct (CHD) of 11mm with possible stone at the proximal part of the common bile duct (CBD). Endoscopic retrograde cholangiopancreatography (ERCP) revealed a pedunculated polyp at the distal part of the common hepatic duct with the dilatation of the common hepatic and the right and left hepatic ducts. (**Figure 1**) After partial exchange transfusion to bring down HbS to less than 50%, the patient was taken for laparotomy. A dilated common hepatic duct (CHD) and normal size CBD were found. There was a 1.5cm long cystic duct remnant containing a hard nodule, which was believed to be the polyp. Common bile duct exploration revealed a polyp, which was arising from and adherent to the cystic duct stump. It fragmented into tiny pigment stones upon mobilization. Choledochoscopy revealed no

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Received 19th January 2003. Accepted for publication in final form 31st March 2003.

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Figure 1 - Cholangiopancreatography showing dilated common hepatic duct and a polypoid filling defect at the junction of common hepatic and common bile ducts.

other biliary abnormalities. The cystic stump was excised and the choledochotomy was closed over a test tube, which was removed at 7th postoperative day, and the patient was discharged home. She remained symptom free at 12-month follow up.

**Discussion.** Intraluminal polypoid lesions in the CBD and large intrahepatic ducts consisting of granulation tissues were seen at ERCP in 26% of patients with AIDS-related sclerosing cholangitis.<sup>3</sup> The presence of these polyps was not associated with any particular infective agent. Similarly, hyperplastic polyp of the CBD was also reported to be mistaken for adenocarcinoma that have had necessitated a major surgical procedure.<sup>4</sup> Cook et al<sup>5</sup> reported an unusual case of biliary obstruction secondary to benign adenomyoma of the CBD, which are found infrequently in the biliary tree and preoperative investigations are difficult to distinguish it from malignant or other lesions. Histological examination is often essential to establish the diagnosis. Although sometimes radiological studies and ERCP may sometimes detect bile duct polyps, exact diagnosis before surgery is very unusual and therefore, the diagnosis is usually surgical.<sup>6</sup> The radiological signs

that suggest presence of a bile duct polyp in the ERCP are the presence of repletion defects fixed unilaterally to the biliary conduit, without meniscus and without circumferential stenosis of the affected conduit.<sup>6</sup> The most frequently found polyps are fibroinflammatory, and less frequently adenomatous.<sup>6</sup> Cholesterol polyp causing CBD obstruction has been reported.<sup>1,2</sup> We believe that repeated hemolysis in this sickle cell patient has caused pigmented sludge and stones to be deposited in the cystic duct remnant that have eventually given rise to an inflammatory polyp covered with granulation tissue as confirmed histologically. Such polyp can be called 'pigment polyp' and sickle cell patients are at an increased risk of developing such polyps. If this were the case, one would expect to see more similar cases in SCD patients in the future. This can be explained by the fact that in laparoscopic cholecystectomy, it is customary to leave a longer cystic duct stump to avoid CBD injury.

To our knowledge, this could be the first inflammatory pigmented polyp arising in the cystic duct stump in a patient with SCD ever reported in the literature.

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