Abstract
A thirty-year-old woman was transferred to the United Kingdom two weeks after bowel resection for superior mesenteric vein thrombosis. She was dehydrated, malnourished and complained of abdominal pain. Computerized Tomography revealed no intra abdominal collection and she was managed conservatively. She settled without intervention and was sent home after 30 days.

Fourteen days later (fifty-nine days since her initial operation), she represented with abdominal pain and discharge from two small wound sinuses on the anterior abdominal wall. A sinogram was performed and contrast was seen to pass into the common bile duct, the cystic duct and into the duodenum. A cholecysto-cutaneous fistula was demonstrated. She underwent an ERCP and biliary stenting. The discharge settled and her abdominal pain resolved. Spontaneous cholecysto-cutaneous fistulae are rare due to the early diagnosis and treatment of biliary tract disease. This case is particularly unusual as the patient had no evidence of biliary tract disease and the cause of the fistula still remains unclear.

Introduction
Spontaneous cholecysto-cutaneous fistulae are uncommon, with the majority of those described in the literature occurring secondary to pre-existing biliary tract disease1-4. We present a case of a spontaneous cholecysto-cutaneous fistula presenting forty-four days after bowel resection for superior mesenteric vein thrombosis. No biliary tract disease was detected.

Case History
A thirty-year-old woman presented to an Emergency department in Israel with abdominal pain, confusion, hypotension and hypothermia. Computerized Tomography demonstrated air in the portal vein consistent with portal pyaemia. Laparotomy revealed gangrenous distal ileum and ascending colon with suspected superior mesenteric vein thrombosis. One and a half metres of small and large bowel were resected and an end ileostomy and mucous fistula were fashioned in the right iliac fossa. Re-look laparotomy twenty-four hours later revealed no further bowel ischaemia, but due to her unstable condition a bowel anastomosis was not attempted at that time. Two weeks later she was transferred to the United Kingdom (where she normally resides).

Shortly after her arrival she presented to the Emergency department dehydrated, malnourished and complaining of generalised abdominal pain with marked tenderness around the stoma site. Her blood results revealed hyponatraemia and deranged liver function tests, (Sodium 126 mmol/l, Alkaline phosphatase 386 IU/l, Bilirubin 5 mmol/l, Gamma-glutamyl transpeptidase 674 IU/l, Albumin 20g/l, Alanine aminotransferase 8 IU/l).

There was no reason for immediate intervention so she was rehydrated and nasogastric enteral feeding instituted. She began to make a good recovery. A few days later, she became pyrexial and developed marked right flank tenderness with a white cell count of 17 x 109/L. Computerised tomography of the abdomen revealed an enlarged liver with patent portal, superior mesenteric, splenic and hepatic veins. There was no evidence of portal gas or venous collaterals and no intra-abdominal collection was identified.

Subsequently, her liver function tests improved and she settled with broad-spectrum antibiotics. After 30 days she was sent home on warfarin having had a weakly positive serum antibody test for anti-phospholipid syndrome. No cause for her abdominal pain was found.

Fourteen days later she was readmitted with abdominal pain and a bile stained serous discharge from two small sinuses along the abdominal wound (figure 1):

Figure 1. Anterior abdominal wall, demonstrating two small sinuses with the stoma on the right side.

An ultrasound of the abdomen demonstrated no intra-abdominal collection and a non-obstructed biliary tree. A sinogram was performed (figure 2) and contrast was seen to pass from the gall bladder to the cystic duct, common bile duct and into the duodenum. A cholecysto-cutaneous fistula was demonstrated:

Figure 2. Two small radio-opaque tubes have been passed through the two sinuses on the abdominal wall at the bottom of the picture. Contrast is seen to fill the tract from the catheters. Contrast is then seen to pass from the tract into a convoluted duct that is the cystic duct. This then drains into the common bile duct and into the small intestine. The gall bladder itself is not outlined.

The bile stained discharge continued despite there being no evidence of distal obstruction. An endoscopic retrograde cholangiopancreatogram showed the cholecysto-cutaneous fistula and normal calibre biliary ducts with no calculi (figure 3):
Perforation of the gallbladder is a well recognized complication of acute cholecystitis, occurring in up to 10% of cases. Spontaneous cholecysto-cutaneous fistulae are an uncommon complication of gall bladder perforation, but have been previously reported in the literature. Perforation may occur into the peritoneal cavity, adjacent bowel or through the skin. Cutaneous fistulae are thought to arise as a consequence of repeated attacks of cholecystitis leading to adhesion of the gall bladder wall to the abdominal parietes. It is usually seen in the geriatric or psychogeriatric populations, as these patients are less likely to undergo surgery. They characteristically have a history suggestive of longstanding biliary tract disease leading to distension and inflammation of the gall bladder predisposing them to fistula formation.

In 1670, Thileus reported the first case of biliary fistula and in 1890 Courvoisier published 169 cases of spontaneous fistulae through the abdominal wall. In the majority of these cases the cystic duct was either occluded by a stone or less commonly a carcinoma. Perforation of the gall bladder is nearly always at the fundus and has been seen to be more common in patients with systemic disease, such as diabetes and athero sclerosis. The fistulae are usually single and exit in the right upper quadrant of the abdomen or at the umbilicus. In rare cases the cutaneous opening has been seen as far away as the anterior chest wall or the back.

With the modern treatments of biliary tract disease these complications rarely arise. There are now only eleven published cases in the last 50 years. In all these cases the pathogenesis of the fistula can be attributed to underlying biliary disease. In the case we present there was no history of cholelithiasis or malignancy. On presentation the liver function tests were abnormal, but no evidence of obstruction to the biliary tree was found. At the time it was suggested that the deranged liver function tests might represent a cholestatic picture secondary to short bowel syndrome. No actual cause was identified and they returned to normal soon after admission. She also had a history of alcohol abuse, but this is unlikely to be directly related to the fistula formation. Her hepatitis serology was negative.

It is possible that there may have been trauma to the gall bladder at the time of surgery, although it seems unlikely, as the fistula did not appear until 59 days after the initial surgical procedure. Another consideration is fundal infarction of the gall bladder secondary to anti-phospholipid syndrome. Perforation of the gall bladder is more common in patients with arteriosclerotic disease and the fundus is the least well vascularised area of the gall bladder. This has not been previously reported as a cause of cholecysto-cutaneous fistula formation.

**Conclusion**

Cholecysto-cutaneous fistulae are usually a result of chronic biliary disease. In this case it is likely to be due to a combination of her poor nutritional status and a pro-thrombotic condition.

**Conflicting Interests** - None declared.

**References**