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SPONTANEOUS EXTERNAL BILIARY FISTULAE-IS IT A REVISIT?

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ABSTRACT

Spontaneous biliary fistulae are encountered, not very rarely, in one's surgical practice. These fistulae are of three types, Internal, External and Combined. Internal spontaneous biliary fistulae are commonest. External fistulae could be spontaneous or because of Therapeutic, Iatrogenic or Traumatic reasons and are very very rare. Spontaneous cholecystocutaneos fistula (SCCF), secondary to calculous cholecystitis is an extremely rare presentation in the present day scenario. It used to be quite common before the year 1900, but is very rare now because of better management of cholecystitis and cholelithiasis. SCCF is, usually, a complication of neglected chronic cholelithiasis. This is seldom seen today because of the early diagnosis and better management made feasible by ultrasound as first line investigation, broad spectrum antibiotics, and effective surgical management of biliary tract diseases. We hereby present two reports including a very rare case of 35 years old female patient presenting in the outpatient department with the multiple stones carefully preserved, which she had been extruding through the fistulous opening in the umbilicus, for the last one year. She was investigated and was operated for the same condition. Though the entity is very rare yet clinician should keep this condition in mind while examining any case of chronic discharging sinus or fistula on the abdominal wall, particularly the wound extruding stones in which case the diagnosis is self-revealing. Though the early diagnosis and improvement in the management of gallbladder disease has improved tremendously yet the possibility of this condition arising mostly from the neglected gallbladder disease should always be kept in mind as such cases are again being reported from all over the world. In the absence of positive history of expelling stones the diagnosis can be confirmed by computerized tomogram [CT] fistulography. The literature of this extremely rare and interesting entity is also reviewed in this article.

INTRODUCTION

Spontaneous Internal biliary fistulae are commonest amongst the biliary fistulae (Avital, 1998). These could be Spontaneous [as a result of Intrahepatic abscess, necrosis or perforation of the gall bladder and inflammation of the biliary tree], Therapeutic, Traumatic or Iatrogenic. Most of these Internal fistulae communicate with duodenum (77%), colon (15%), and stomach (6%) (Glenn, 1981). Rarely it can communicate with urinary system or bronchial tree (Henry, 1949). Spontaneous external biliary fistulae are very rare and were first described in 1670 by Thilesus (Horhammer, 1916). In the world literature only 65 cases had been described, since the year 1900 (Henry, 1949) and Fitchett, 1970).

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Prior to 1900 AD, Three large series were published in quick succession, including a detailed report by (Courvoisier, 1890) in1890, which described 169 of 499 cases of gallbladder perforation, and (Naunyn, 1892), in 1896 reported a series of 184 cases, whereas another series by (Bonnet, 1897), in 1897 comprising 122 cases. The incidence of SCCF has reduced dramatically; from the years 1890 to 1949, only 37 cases were identified in the published literature (Henry, 1949). No data exists in United States for the incidence of this condition (Cherry, 1972). In a retrospective study of this condition in Greece, out of 210 cases of internal biliary fistulae over a period of 22 years only one was due to spontaneous cholecystocutaneous fistulae (Dadoukis, 1998). A review of literature published in the last 50 years from 1961 to 2011 reveals fewer than 50 cases. The decreasing incidence is furtherer established by the large series published prior to the 20th century, in contrast to the recent literature, which consists

mainly of individual case reports. Less than 20 cases prior to year 2006 has been reported in the last 50 years (Cruz.,2006). Now, most biliary fistulae are postoperative complication of liver and biliary tract surgery or trauma. Biliary fistulae can be divided into external and internal biliary fistulae. External biliary fistulae, can further be subdivided into Spontaneous, Therapeutic, Traumatic and Iatrogenic fistulae. External spontaneous cholecystocutaneous fistula is very rare surgical complication of neglected calculous biliary disease that has become even increasingly rarer because of easy and early diagnosis and expedient surgical intervention for gallstones disease. External biliary fistulae sometimes occur spontaneously as a result of intrahepatic abscess (Pyogenic or parasitic), Necrosis or perforation of the gall bladder, or other inflammatory processes involving the biliary tree (Nicholson, 1999) and (Nayman, 1963). Though the entity had almost vanished yet recently, a few cases are being reported from all over the world (Cherry, 1972). In spite of early diagnosis and better management of gallbladder disease, it is feared that this may not be a revisit by this, once not so uncommon entity, and a clinician should arouse a suspicion of spontaneous cholecystocutaneous fistula in the patient having chronic discharging sinus or fistula on the abdominal wall whether the history of the gallbladder disease is forthcoming or not. A rare case of spontaneous cholecystocutaneous fistula and its management is also discussed here. The available literature is also reviewed.

Case -1

A 35 year old female patient presented in the outpatient department, with history of passing multiple stones, repeatedly, from her umbilicus for the last one year. There was no history of episodes of fever, chills and rigors and anorexia. On examination, thinly built, asthenic patient had an umbilical fistula, having serous discharge from the umbilicus. The patient presented the carefully preserved stones which she used to frequently expel from her umbilicus (Fig.1).

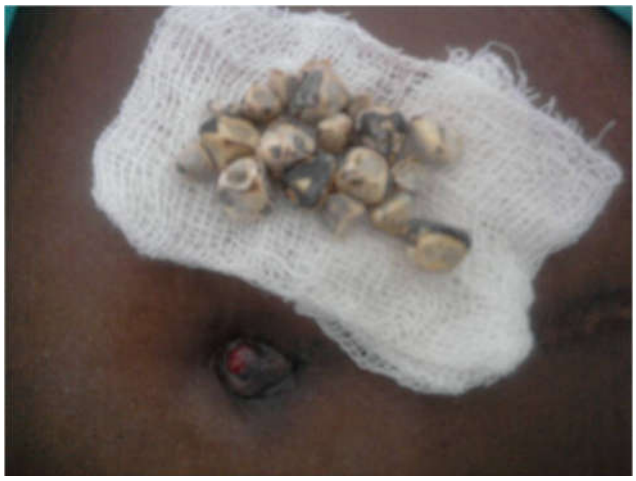

Fig. 1. Spontaneous external Cholecysto-umbilical fistula, extruding gall stones from umbilicus

The fistula was chronic and painless. Clear history of passing large, faceted, multiple stones repeatedly through the umbilicus was the mainstay of the clinical diagnosis which was confirmed by the CT fistulogram, as it delineated the tract and demonstrated the multiple stones present in the fistulous tract leading to the umbilicus (Fig.2). There was no history of abdominal trauma or previous surgery in this patient. Patient was posted for exploratory laparotomy after all requisite pre-operative investigations were done. Cholecystectomy of the chronically inflamed and shrunken gall bladder was done along with excision of the fistulous tract. The fistulous tract containing multiple stones, extending from gall bladder to umbilicus and traversing anterior abdominal wall was dissected and excised from anterior abdominal wall. The tract was identified emerging from the gall bladder, which was shrunken and fibrosed. There were no stones in the gall bladder. Postoperative period remained uneventful.

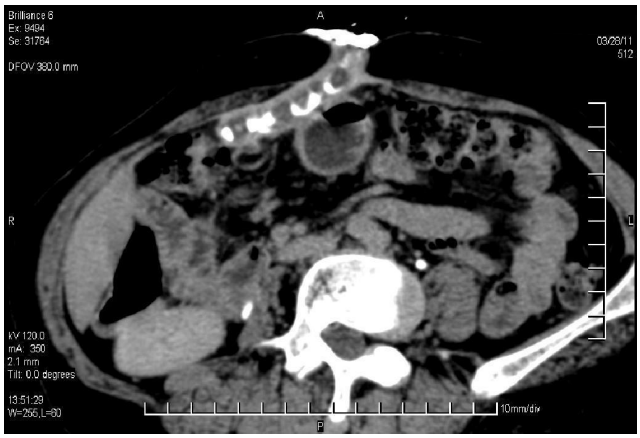

Fig. 2. CT Fistulogram demonstrating Cholecysto-umbilical fistulous tract containing multiple gall stones

Case-2

A 50 years female patient presented with multiple episodes of pain abdomen and history of recurrent/sub acute intestinal obstruction. The patient was diagnosed to have cholecystogastric fistula on CT examination (Fig. 3) and a wide fistulous tract, communicating gall bladder with the pre-pyloric region of the stomach ,containing large multiple stones stuck up in he tract was excised and repaired (Fig.4), alongwith cholecystectomy. Patient recovered well post operatively.

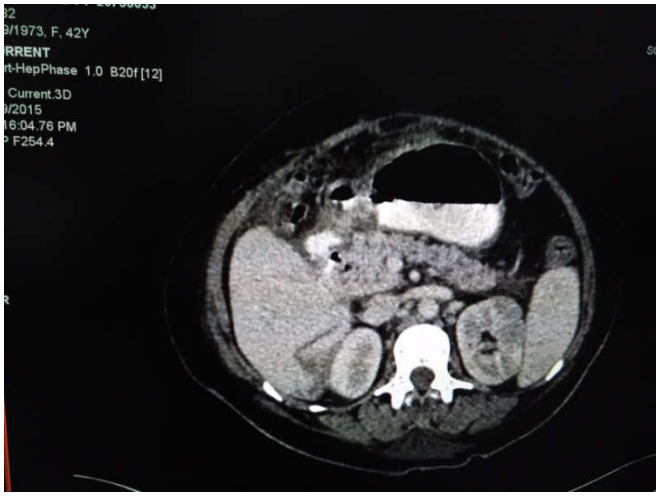

Fig. 3. CT scan showing Cholecysto-gastric fistula

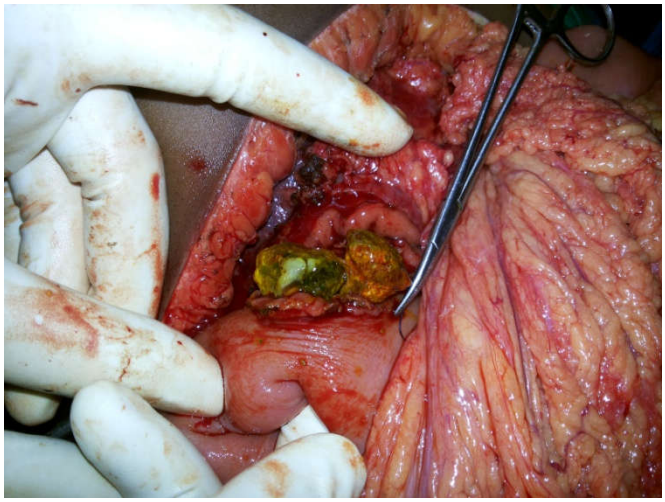

Fig. 4. Cholecysto-gastric fistula containing large, multiple gall-stones

DISCUSSION

Spontaneous biliary fistulae may be external or internal, most being internal (Avital, 1998). The internal fistulae communicate with the enteric lumen mainly to the duodenum (77%) and to a lesser extent to the colon (15%) and stomach (6%) (Fig. 3), (Glenn, 1981). These fistulae may remain symptomless or can cause varied symptoms as pain abdomen, as in the second case reported above. These fistulae may cause Gall stone ileus, suspected as the cause of positive history of recurrent bouts of intestinal obstruction in the second case, as a large fistula communicating gall bladder with the distal end of the stomach containing large gall stones was seen (Fig.3,4). These fistulae may also cause cholangiitis and jaundice, when they impinge upon common bile duct, or rarely are of ulcerous (duodenal) origin (Abbey, 2003). Other rare types of internal spontaneous biliary fistulae, such as those terminating in urinary system or bronchial tree have also been reported (Henry, 1949). A rare case of spontaneous external and internal (combined) fistula has also been described (Urban, 2001). Spontaneous external biliary fistula discharging into the skin surface, as in the present case, is rare. It is defined as a rupture of the Gall bladder through all layers of the abdominal wall, with the creation of a fistulous tract to the skin, not preceded by any biliary surgery or trauma. The first description of SCCF is credited to Thelesus (Horhammer, 1916) in 1670. In 1890 Courvoisier (6) published a large series describing 169 patients with spontaneous external biliary fistulae draining to the abdominal wall. Other unusual sites of the external opening in SCCF are, right inguinal region, right anterior chest wall, left costal margin, right iliac fossa, right groin and right gluteus region, back (Fitchett, 1970), and abdominal scar (Ulreich,1983). Most external fistulae drain in right upper quadrant (Raffaele, 2010), but the umbilical area is less common. In another series (Henry, 1949), it was found that 47% 0f such fistulae opened at right upper abdominal quadrant whereas 27% fistulae were opening in the umbilical area. In the year 1949 Henry and Orr3 reviewed 37 patients with external SCCF which had been diagnosed since 1890. Since then only sporadic reports of this biliary tract disease complication have appeared, mainly as case reports of patients with neglected biliary disease, (Rudderman, 1975). Causes of external SCCF include, obstruction, and less frequently gall bladder adenocarcinoma occluding the cystic duct (Hoffman, 1982) SCCF has also been described in acalculous cholecystitis, (Birch, 1991), (Khan, 2010). The perforation of the gallbladder without stones is said to complicate 0.6%-1% of all cases of acute cholecystitis. The aetiopathogenesis of the perforation in these cases of acalculous cholecystitis is not very clear, although bacteraemia, polyarteritis nodosa, steroids, trauma and typhoid have all been implicated (Sherlock, 1985). The process of fistula formation is precipitated by obstruction of the cystic duct, which raises the pressure in the gall bladder, impairing the vascular supply and resulting in focal necrosis. This inflammatory process is typically insidious and recurrent. The fistula usually forms through the fundus of the gallbladder (Vasanth, 2004), since this part of the gallbladder is farthest from the cystic artery and thus most likely to be affected by ischemia. Chronic inflammation of the gall bladder can cause the gall bladder fundus to adhere to the abdominal parities triggering the formation of fistulous tract. Underlying pathophysiology is the perforation of gall bladder which may be acute, sub-acute or chronic, it is the chronicity of the diseased gall bladder which is responsible for SCCF (Birch, 1991). Retained gall stones following laparoscopic cholecystectomy may cause biliary fistula or abdominal wall sinuses (Cherry, 1972). This occurs because gall stones can harbor bacteria, which may form localized abscess with localized sinus, in an attempt to discharge the foreign body (Lau, 1996), Weiler, 2002). Salmonella typhi, which has a predilection for the gallbladder can cause chronic cholecystitis and may predispose to spontaneous SCCF (Birch, 1991). A perforation may occur alongwith concomitant adhesions with the internal abdominal wall and formation of an abdominal wall abscess may be initiated. Spontaneous or surgical drainage of the abscess can lead to the formation of a fistula. The spontaneous cholecystocutaneous abscess is walled off by the abdominal wall and finally penetrates it. The cholecystic abscess may initially cause a tender area in the abdominal wall and spontaneous rupture forming a fistula draining in to the skin. Communication to the umbilicus may be through the falciform ligament, (Davies, 1989). However the fistula formation may not always be preceded by an episode of acute cholecystitis, and that may be one of the reasons that the patient ignores the mild symptoms as it may be painless or the patient may not be having any noticeable symptoms. Painless cholecystoumbilical fistula has been reported in the literature (Avital, 1998 and Davies, 1989). Sometimes the only manifestation may be the passing of the stones and discharge, from the fistula which is the case in the present patient also. Some other similar reports are also available in the literature Davies, 1989). Most of these patients have a history of biliary disease, with mild symptoms, depending on the stage of progression (Raffaele, 2010). The patient may present with empyema of the gall bladder or a discharging sinus (Nayman, 1963). Systemic symptoms like fever, sweating, chills and rigors, anorexia and a tender or painful lump may be present. An erythematous skin lesion may be the only presenting feature. The fistula itself may be painless as in the present case. The majority of the patients having external biliary fistulae are elderly females (Rosario, 1990), though our patient was of comparatively younger age i.e.35 years. One of the reasons for this younger age of our

patient may be that the gall stone disease is now seen in younger age group. These SCCF has been reported as early as the third decade of life also (Cherry, 1972) and a case has been reported at the at the young age of 24 years (Andley, 1996). Younger patients may neglect their symptoms for a comparatively longer time or these patients have to be ruled out neuropathy, causing altered sensation, (Hoffman, 1982). Another case of spontaneous cholecystocutaneous fistula has been described where the patient was neither having pain nor there were any symptoms of gallbladder disease (Leela, 2010). Females are affected more than males, reflecting the higher incidence of gall bladder disease among females. The psychogeriatric population having cholelithiasis is more likely to have this problem as cerebral atherosclerosis may obscure the acute symptoms (Davies, 1989). The fistulous discharge varies depending upon whether the fistulous passage is obstructed or not. The discharge may be purulent if empyema is the source, mucoid if the source is mucocele or bilious in the absence of obstruction. Passing of the stones through the fistula with the discharge confirms the diagnosis clinically as in the present case. Various conditions which can be considered for differential diagnosis are, metastatic carcinoma, tubercular sinus, pyogenic granuloma, chronic osteomyelitis of ribs with sequestrum and infected epidermal inclusion cyst.

The other main causes of SCCF are, abdominal trauma, previous abdominal surgery and therapeutic percutaneous cholecystotomy used to treat cholecystitis or empyema of the gallbladder, generally reserved for patients unfit for surgical intervention. The patient in the case repot no.1 did not have any history of these procedures performed on her. Possibility of the SCCF should always be considered in any patient who has a chronic discharging sinus in abdominal wall or umbilicus, moreover the typical history of the patient is self revealing and diagnostic. The other less common causes of SCCF incude retained stones, enteric infection, polyarteritis nodosa and steroids (Avital, 1998), the conditions which were excluded in this patient by careful history and clinical examination. A SCCF fistula may be a rare presentation of the underlying carcinoma of the gall bladder. The association of adenocarcinoma with chronic wounds, sinuses and scars is very well known but only one case of associated adenocarcinoma in cholecystocutaneous fistula in the gall bladder could be traced (Gifford, 1981), (Vasanth, 2004), (leela, 2010).

Another case of associated carcinoma in SCCF has been reported in the recent literature also (Sodhi, 2012), though an another mention of gall bladder adenocarcinoma blocking the cystic duct and causing spontaneous cholecystocutaneous also exists in the literature. (Hoffman, 1982). Gall bladder cancer is an aggressive malignancy and has dismal prognosis, (Barlette, 1996 and Gibson, 1987). It has remarkable tendency to seed and grow in the peritoneal cavity, as well as along the needle biopsy site and in the laparoscopic port sites. It has the propensity of spreading early by direct extension into the liver and other adjacent organs. The presence of right upper quadrant mass in relation to gall bladder cancer reflects an advanced disease and unresectability in more than 90% of cases. Palliative Chemotherapy is the mainstay of treatment in such cases. However, the histopathology of the gall bladder and the fistulous tract tissue sent for examination in the present case revealed the benign nature of the disease. Presentation of SCCF in the presence of carcinoma of gall bladder usually means that the disease is in advanced stage and beyond surgical salvage. Palliative treatment is the aim and overall prognosis is grave.

INVESTIGATIONS

Haematology: Leukocytosis and raised CRP levels indicate an inflammatory and or infective process. ALP (Alkaline phosphate) may be elevated if extrahepatic ductal obstruction is there. However jaundice is not common even if choledocholithiasis is present (Hoffman, 1982). Analysis of the fistulous content may reveal the nature of the discharge, whether it is bilious and may provide the bacteriological results aiding in initiating the treatment. Eschcherichia coli and proteus species are commnonly detected organism from the fistulous discharge. CT scan will be helpful in demonstrating abnormal position of the gall bladder in case it is adherent to the anterior abdominal wall. A heterogenous mass may be apparent in case of malignancy. CT fistulogram can demonstrate the fistulous tract and its contents, making a definitive diagnosis as was done in the present case (Fig. 2 to 4). The contrast will demonstrate the tract and gall bladder. Fistulography also may demonstrate the CBD, permitting evaluation of the biliary anatomy, It may also demonstrate multiple fistulous tracts or communications in some rare cases. Cholangiography will outline biliary anatomy and will exclude the presence of choledocholithiasis which should be simultaneously tackled at the time of the surgery. MRCP has been described as a confirmatory investigation in a case where CT scanning did not establish the fistula until the fistula passed calculi (Leela, 2010). USG is useful as it can demonstrate gallstones, condition and position of the gall bladder whether it is adjacent to the anterior abdominal wall, and an overlying oedematous abdominal wall due to inflammation may also be demonstrated. Occasionally USG can demonstrate a gallbladder herniating in the subcutaneous tissue (Carragher, 1990). Inflammation of the overlying skin can limit examination because of pain. Clinical presentation and radiological imaging provide valuable information in making the diagnosis, of this rarely seen condition. The biopsy from the presenting mass, if any, may not prove helpful (Leela, 2010). The various differential diagnosis of SCCF, which could be considered are, Infected epidermal inclusion cyst, tubercular sinus/fistula, Pyogenic granuloma and Metastatic carcinoma.

Treatment

The initial treatment consists of drainage and antibiotics as an adjunct, followed by evaluation with abdominal USG and CT fistulography. Bacteriological examination of the contents of the fistula, are helpful in deciding antibiotic therapy. Drainage of the abscess prior to the formation of the spontaneous fistula turns the abscess into a fistula and may allow control of sepsis. Many methods have been described to keep the tract patent including the dilatation of the fistulous tract (Cherry,1972). Drainage of the cholecystocutaneous abscess also offers temporary relief and buys time to prepare the patient for definitive surgery. Surgery is required most of the times and includes cholecystectomy with the excision of the tract, as

both the gallbladder and the fistulous tract need to be excised to achieve a cure, further the surgical options may be rationally used depending upon the co morbid conditions usually present in such patients. In the present case open cholecystectomy with the excision of the tract was done and a subhepatic drain was placed which was removed on the third day. Drain should always be placed if a sub hepatic collection is found/ suspected, an infective process is still present or difficult dissection has taken place with even a minor degree of suspicion regarding any hepatobiliary injury. The gallbladder was shrunken and fibrosed whereas the fistulous tract contained multiple gall stones and the tract was adherent with the anterior abdominal wall as well with the surrounding tissue. The tract was opening into the umbilicus, externally. Laparoscopic approach which is less invasive has also been used in these cases. First such case dealt by laparoscopic approach has been described (Kumar,1998), however the conversion rate may be high depending upon the intra abdominal findings and the condition of the gall bladder, common bile duct and the fistulous tract. More adhesions are expected in such cases. Port placement may also need to be improvised depending upon the adhesions and the course of the fistulous tract. another approach has been described (Malik, 2007), Leela, 2010), in which gall bladder is laparoscopically removed but the excision of the fistula from the abdominal wall is not done. An open approach should be used if malignancy is suspected.

However spontaneous closure of the fistula is a possibility, as recorded in few cases, this can occur provided there is no distal obstruction, (Henry, 1949), (Davies, 1989). In the review of 37 patients in their series in 1949 by Henry and Orr [3], 6 [16%] healed spontaneously. Incision and drainage of the cholecystic abscess without definitive excision of the tract or gall bladder lead to the spontaneous healing in three more patients in a different series (Cherry,1972). Spontaneous closure of the fistula in a patient has been described by (Davies, 1989), in whom extraction of the gall stones from the fistulous tract was done, on direct exploration. Spontaneous healing as an option may be tried in patients who are at very high risk of surgery (Hoffman, 1982) or in patients with significant co morbidities Leela, 2010), as 20% of all external SCCF may heal spontaneously (Birch, 1991). Staged procedures may be required, in the form of drainage of abscess with cholecystectomy and excision of the tract, or drainage of the abscess followed with definitive surgery. Whether to include the external opening in the excision, and method of closure will have to be decided on case to case basis. With modern day advancements in surgery and anaesthesia there are hardly any contraindications for definitive surgery in these cases and the same is recommended. Other procedures as ERCP balloon retrieval and sphincterotomy have been tried with success in morbid patients, more conservative approach may be adopted in elderly patients having co morbidities. The diagnosis of this rare entity often proves challenging if the clear history of passing the calculi per fistula is not there as a significant proportions of these patients present with non specific symptoms, ideally the treatment should include broad spectrum antibiotics, drainage of the abscess and elective cholecystectomy with excision of the fistula (Leela, 2010).

Complications

Spontaneous cholecystocutaneous fistula is itself a complication of the neglected gall bladder disease. septicaemia may ensue prior to fistula formation which may subside septicemia. Necrotising fasciitis of the anterior abdominal wall due to sepsis has been reported (Chang, 2002). Bilious discharge can cause skin irritation and dermatitis. Dysplasia and subsequent malignant change similar to marjolin ulcer has been described as a result of chronic untreated fistula. Only one case of adenocarcinoma arising from a biliary fistulous tract has been reported (Gifford,1981). Mailgnant change in the fistulous tract is rare and may occur after a long period of 10-20 years (Gifford,1981).The ERCP and stenting has aso been used in the management of these cases (Avital,1998). Conservative treatment of SCCF has also been reported with success (Raffale, 2010), even cauterization of the tract leading to spontaneous closure has been described (Rudderman,1975), Expulsion and or extraction of stones from the fistulous tract may lead to the Spontaneous closure of the fistula (Davies,1989). Only cholecystectomy leading to spontaneous closure of the fistula has also been reported (Avital.,1998). (Henry, 1949), reported spontaneous closure in 8 out of 36 cases during a period between the years 1890-1948. Even staged procedures were used to be done earlier for this condition (Davies, 1989). Only laying open of the fistulous tract has also been reported with cure of the condition (Avital, 1998).

Conclusion

The possibility of External Spontaneous cholecystocutaneous fistula though very rare in present day scenario, should be kept in mind, in a patient having discharging sinus over abdomen or lower chest wall. In the patient passing stones through these fistulae, the diagnosis is obvious. Though with the advent of newer and efficient investigative and operative modalities the diagnosis and management of gall bladder disease has been made easier yet the entity may be the result of neglected gall bladder disease as quite a few of these cases have been recently observed, as the modern day advances in the treatment of gall stone disease are still not available in some pockets of the poorer population who prefer to bear or ignore the disease for the socioeconomic reasons, or is it a revisit by this entity?

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