

SPONTANEOUS COMBINED INTERNAL AND EXTERNAL BILIARY FISTULAE IN ASSOCIATION WITH GALLSTONES AND GLIOMATOSIS OF THE GALLBLADDER*

Fístulas Biliares em Combinação Espontânea Interna e Externa Associadas a Cálculos e Glimatose de Vesícula Biliar

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ABSTRACT

Spontaneous combined internal and external biliary fistula is a very rare complication of biliary tract disease. Only two previous cases have been reported so far, and all of them with previous episodes of acute cholecystitis, treated without surgery. We describe a 27-year-old female patient (the younger reported to date) who presented with a long history of neglected gallbladder inflammation complicated by neuroglial implants on the peritoneum and on the gallbladder wall due to an ovarian teratoma. She had spontaneous cholecystocutaneous fistula and cholecystocolic fistula that were successfully treated by cholecystectomy and excision of the fistulous tracts.

Key words: fistula; gliomatosis peritonei; cholecystitis; gallstones.

RESUMO

As fistulas combinadas internas e externas são complicações muito raras das patologias biliares. Apenas dois casos foram previamente reportados, ambos com episódios prévios de colecistite aguda, tratada clinicamente, sem cirurgia. Os autores descrevem, neste estudo, uma paciente de 27 anos de idade (a mais jovem relatada na literatura), que se apresentou com uma longa história de colecistite calculosa, complicada por implantes neurogliais no peritônio e na parede da vesícula biliar, originários de um teratoma ovariano maduro. Ela apresentou fistula colecistocutânea e colecistocólica, que foi tratada com sucesso pela colecistectomia e excisão do trajeto fistuloso.

Palavras-chave: fistula; gliomatose peritoneal; colicistite; vesícula biliar.

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INTRODUCTION

Spontaneous biliary fistula is a rare complication of gallstones, occurring in 3 per cent of all patients undergoing biliary surgery.¹ Such fistulae usually occur between the biliary tree and the gastrointestinal tract (internal fistulae). Spontaneous external biliary fistula was common in the last century, but became rare because of early diagnosis of biliary tract disease.² We report the third case of simultaneous external and internal biliary fistulae in a 27-year-old female patient. This is the younger patient reported to date, and the first with the association of long time biliary tract disease, gallstones and gliomatosis peritonei with deposits on the gallbladder wall due to an ovarian teratoma.

CASE REPORT

A 27-year-old woman was admitted for the first time six years ago with an episode of acute cholecystitis and an ultrasound scan confirmed the presence of gallstones. At that time the patient refused surgical treatment. After that first admission she had 3 more episodes of biliary pain. The second previous admission was two years ago when she was submitted to an exploratory laparotomy due to a 30 cm partially cystic mass replacing the left ovary. Ascites and multiple peritoneal deposits were also present. A left salpingo-oophorectomy and omental biopsy were performed and histological examination showed, at that time, a well differentiated solid teratoma with omental deposits of benign neuroglial tissue. No evidence of extra-abdominal disease was found at that time. On her third and last admission she presented with a one year history of discharging sinus affecting her right subcostal region. A sinogram revealed cholecystocutaneous fistulae, multiple large gallstones, cholecystocolic fistulae; the remaining of biliary system appeared within normal limits (Figure 1). Another laparotomy was then performed. A thick walled contracted gallbladder containing several large stones was

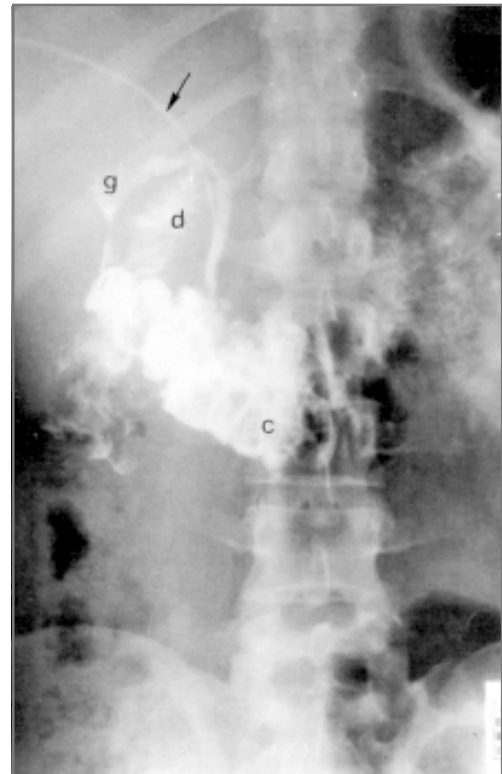


Figura 1 - Contrast sinogram showing: the catheter introduced in cutaneous fistulae (arrow), gallbladder (g), colon (c) and duodenum (d).

adherent to the anterior portion of the abdominal wall, and there was a further communication with the transverse colon. The fistulae were divided, and the defect in the transverse colon was then surgically closed. There was no mass, but the peritoneum persisted with multiple deposits. Postoperative recovery was uneventful. Histological examination of the gallbladder showed extensive fibrosis of the wall with deposits of well differentiated circumscribed neuroglial tissue (Figure 2). In the fistula there was only fibrous tissue. At present, 28 months after her third admission, she is doing well, and the wound and fistulae healed completely.

DISCUSSION

Spontaneous external biliary fistulae was a common complication of biliary tract disease in the last century. The first description is attributed to Thilesus in 1670 while Courvoisier in 1890 described further 169 cases. After these initial reports, Naunyn

in 1896 reported 184 cases, and Bonnet (1897) 122 cases. However only 16 cases have been described in the last 50 years, and only two cases of spontaneous combined internal and external biliary fistulae have been reported to date.^{3,4}

Spontaneous biliary fistulae are usually a complication of acute suppurative cholecystitis associated with cholelithiasis, and are uncommon in biliary disease without gallstones (0,6%). Perforation in such cases occurs probably because of bacterial dissemination, steroids, polyarteritis nodosa, typhoid, and trauma.² The necrosis of the gallbladder wall and perforation leads to drain throughout the abdominal wall to the surface of the skin (external fistulae), or to the abdominal viscera such as the duodenum (77%), colon (15%), or rarely into the bronchial tree, stomach, and even urinary tract (internal fistulae).⁴

The gliomatosis peritonei in association with ovarian teratoma is also a rare condition, with few cases reported in the literature. Robboy and Scully reviewed 12 cases of gliomatosis peritonei in association with ovarian teratoma and showed good prognosis of patients with this kind of secondary deposits due to the benign behavior of mature implanted tissues. The implants of the ectopic glial tissue are probably due to capsular defects in the primary ovarian tumor, although lymph node spread has also been described.^{4,5} The behavior of ectopic neural tissue is not completely understood.

Cytogenetic analysis of benign ovarian teratomas usually reveals normal karyotypes (46,XX). There are very few data reporting chromosome abnormalities in this type of tumor. Surti in 1990 found 7% of the numerical alterations (trysomy, triploidy and mosaicism) in 102 ovarian teratomas analysed.⁶

So far this seems to be the first case in the literature describing the association of gliomatosis peritonei in the wall of the gallbladder with cholelithiasis and further complicated by spontaneous combined internal (colon) and external biliary fistulae. The two previous cases affected old women (81 and 84-year-old) with previous episodes

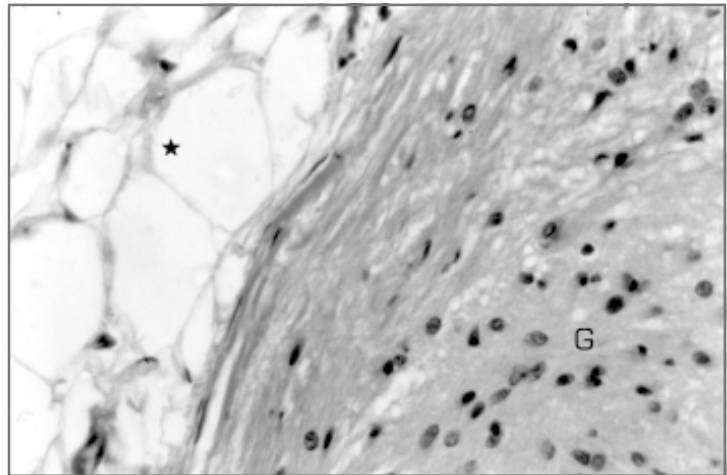


Figura 2 - Histological section of gallbladder showing serosal adipose tissue (*) and nests of glial cells (G) (HEX100).

of acute cholecystitis treated without surgery because of their poor health conditions.^{3,4} In the present report the development of the fistulae could be due to the long history of neglected gallbladder inflammation complicated by the association with the presence of ectopic neural tissue implants on the wall of gallbladder, which contributed to a poor contraction and stasis.

The treatment for such fistulae is cholecystectomy with the excision of the fistulous tract. However, in high risk patients, percutaneous removal of the stones should be also considered, so that the cholecystocutaneous fistula might close spontaneously.³ The treatment of choice to gliomatosis peritonei is the simple excision of any symptomatic masses.^{7,8}

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