Gallbladder agenesis intraoperatively diagnosed: a case report

The agenesis of the gallbladder is a rare anomaly with an incidence varying between 0.01 and 0.05% [1]. It may be associated with other digestive, cardiovascular or genitourinary tract malformations, which are sometimes incompatible with life. This could explain the low prevalence of this anomaly in the surgical series (0007% - 0027%), compared to autopsy reports (0.04 - 0.13%) [2]. An association with stones in the bile duct has been reported in 25-50% [2].

Misleading symptoms and false ultrasound images bring patients to unnecessary surgery. Magnetic resonance cholangiography (MRC- MRI) is the key examination to confirm the diagnosis preoperatively because it allows a biliary cartography without injection of contrast material [3].

We report a case of gallbladder agenesis associated with an absence of cystic duct diagnosed intraoperatively.

Case report

A 50-year-old man, presented to our hospital for episodes of right upper quadrant pain, not punctuated by meals, evolving for the last two months.

He was a smoker and had a history of a duodenal ulcer treated 16 years ago and a fatty food intolerance for which he was on diet. The patient was in good general condition, anicteric and afebrile. Abdominal examination found mild tenderness of the right upper quadrant and epigastrium. Laboratory tests were within normal values.

Figure 1: Intra-operative cholangiography showing absence of the gallbladder



Abdominal ultrasound showed hyperechoic shadows suggestive of multiple gallstones, with a shrunken gallbladder. The bile ducts were of normal caliber. Based on the clinical and ultrasound features, a cholecystectomy was decided. The intraoperative exploration by a conventional laparoscopic approach found an empty gallbladder fossa. Conversion to an open procedure was decided. A thorough manual exploration failed to locate the gallbladder. Intraoperative cholangiography was performed via catheterization of the bile duct. It showed a common bile duct (CBD) of normal caliber with a normal

intrahepatic biliary tree, but did neither visualize the cystic duct nor the gallbladder (fig.1 and 2). Besides, a perforated clogged duodenal ulcer of intraoperative incidental discovery was sutured. The postoperative course was uneventful.

Figure 2: Intra-operative cholangiography after massive injection confirming agenesis of the gallbladder



An abdominal CT made two weeks after surgery, showed an infiltration of pre-antral and mesenteric fat, secondary to ulcer perforation, with an empty gallbladder fossa (fig.3a and 3b). Moreover, this examination didn't found the gallbladder in any ectopic location and revealed no other abdominal or pelvic abnormalities.

Figure 3a et 3b: Abdominal CT showing an empty gallbladder fossa





Conclusion

The gallbladder agenesis is certainly a rare entity, but has to be kept in mind in doubtful situations in order to further investigations before surgery. MRC-MRI can confirm the diagnosis and rule out a gallstone disease. Unnecessary surgery and its risks, would be avoided.

References

- Toufeeq Khan TF, Baqai FU. Agenesis of the gall bladder with duplication cysts
 of the hepatic flexure A case report and literature review. Singapore Med J
 1991:34:181-82.
- N. Peloponissios, M. Gillet, R. Cavin, and N. Halkic, "Agenesis of the gallbladder: a dangerously misdiagnosed malformation," World Journal of Gastroenterology 2005;11:6228-31.
- Valeria Fiaschetti, Giovanna Calabrese, Silvia Viarani and al. Gallbladder agenesis and cystic duct absence in an adult patient diagnosed by magnetic resonance cholangiography: report of a case and review of the literature. Case Reports in Medicine, 2009.

Noomene Haoues, Sarra Zairi, Haithem Zaafouri, Anis Ben Maamer, Rabii Noomene, Abdelaziz Oueslati, Ahmed Bouhafa, Abderraouf Cherif

Departement of surgery, Habib Thameur Hospital, Tunis

An exceptional association: ulcerative colitis, amyloidosis and type-2 diabetes mellitus

The association of inflammatory colitis (Crohn's disease and ulcerative colitis) to diabetes let us think of suspected autoimmune disorders, the association to amyloidosis is recognised and this is due to the inflammatory state.

The association of ulcerative colitis, amyloidosis and diabetes was not described until now in the literature.

Islet amyloid has been implicated as a pathological entity in type 2 diabetes since the turn of the century. It has as its unique component the islet beta-cell peptide islet amyloid polypeptide (IAPP), or amylin, which is co-secreted with insulin (1, 2).

The progressive accumulation of IAPP-containing fibrils allows the eventual replacement of beta-cell mass by amyloidal and contributes to the development of hyperglycaemia (3, 4).

The aim of this study is to discuss if the association of ulcerative colitis, diabetes and amyloidosis is fortuitous and to focus on the role of IAPP in the occurrence of type-2 diabetes.

Case Report

We report the case of a 49 years old man having an ulcerative colitis diagnosed since the age of thirty years when the patient had diarrhoea and rectal haemorrhage.

The diagnosis was confirmed by the anatomo-pathological exams. The treatment consisted of salazopyrine and then total large bowel resection.

A type-2 diabetes was revealed ten years after. The patient was treated by oral anti-diabetics then insulin.

Six years after, he was hospitalized to explore generalised oedema. The physical examination found orthostatic hypotension and oedema of inferior legs.

He had also nephrotic syndrome and renal insufficiency. The diagnosis of amyloidosis was confirmed by accessory

salivary glands biopsy. The type of amyloidosis was AA.

The patient refused to practice a biopsy of the pancreas.

The outcome of the patient was characterised by disturbance of renal function implicating extra-renal purifying, a high persistent proteinuria, and an activated ulcerative colitis.

After necessary liquid supply, arrest of conversion enzyme inhibitor and treatment by colchicine and salazopyrine; the digestive signs, the renal function and the glycaemia were improved.

Nevertheless, the patient dead in terminal renal stage insufficiency.

The pancreatic biopsy wasn't done in our case. Nevertheless, the literature results permit to us to think about the hypothesis of pancreatic amyloid deposits explaining the type 2 diabetes installation. Then, in our case the association of ulcerative colitis to amyloidosis is classic, but the occurrence of a type 2 diabetes let us think about its physiopathology.

Conclusion

Beta-Cell dysfunction is an important factor in the development of hyperglycemia of type-2 diabetes mellitus, and pancreatic islet amyloidosis has been postulated to be one of the underappreciated contributors to impaired insulin secretion. The comprehension of the mechanism(s) involved in islet amyloidogenesis allow the development of therapeutic agents that inhibit amyloid fibril formation, with the goal being to preserve beta-cell function and improve glucose control in type 2 diabetes.

References

- Höppener JW, Nieuwenhuis MG, Vroom TM et al. Islet amyloid and diabetes mellitus type 2. Ned Tijdschr Geneeskd. 2000; 144:1995-2000.
- 2- Jaikaran ET, Clark A. Islet amyloid and type 2 diabetes: from molecular misfolding to islet pathophysiology. Biochim Biophys Acta. 2001; 1537: 179-203
- Powell DS, Maksoud H, Chargé SB et al. Apolipoprotein E genotype, islet amyloid deposition and severity of Type 2 diabetes. Diabetes Res Clin Pract. 2003: 60: 105-10.
- 4- Butler AE, Janson J, Soeller WC et al. Increased beta-cell apoptosis prevents adaptive increase in beta-cell mass in mouse model of type 2 diabetes: evidence for role of islet amyloid formation rather than direct action of amyloid. Diabetes. 2003; 52: 2304-14.

Mahfoudhi Madiha, Ounissi Mondher, Hariz Anis, Ben Abdelghani Khaoula, Boubaker Karima, Turki Sami, Kheder Adel.

Department of Internal Medicine A - Charles Nicolle Hospital Tunis - Tunisia

Métastase testiculaire bilatérale d'un adénocarcinome prostatique

Les métastases testiculaires sont rares, ayant le plus souvent pour origine la prostate [1]. Les métastases testiculaires du cancer de la prostate sont caractérisées par leur latence clinique et leur découverte est habituellement fortuite [2,3]. Les atteintes