Ciliated cyst of the gallbladder: A new case and literature review

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A B S T R A C T
INTRODUCTION: The ciliated cyst is a rare anomaly that develops from the anterior primitive intestine. Its localization is essentially supra-diaphragmatic. It’s localization in the gallbladder is very uncommon.

CASE REPORT: We report the first case in Tunisia of a ciliated cyst of the gallbladder in a 34 years old woman who was operated for a gallbladder stone and in whom the discovery of a cystic mass attached to the neck of the gallbladder was preoperatively. The pathologic study did conclude to a ciliated cyst.

DISCUSSION: The ciliated cyst of the gallbladder is a benign congenital lesion that develops from the anterior primitive intestine. The most frequent clinical symptom is abdominal pain and the median age is 45 years old. The most frequent location is the neck. This pathology affects women more than men. Abdominal ultrasound is not very specific exam and describes the ciliated cyst as a cystic lesion often anechogenic and sometimes hyperechogenic. The CT-scan as well as the abdominal MRI are very helpful of the diagnosis. The positif diagnosis of ciliated cyst is histological.

CONCLUSION: The recommended current treatment for this rare pathology is surgery and it consists of a celioscopic cholecystectomy. The place of conservatory treatment hasn’t been established due to the rarity of described case and the possibility of degeneration.

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1. Introduction

The ciliated cyst is a rare congenital lesion developed from the primitive anterior intestine who is the origin of the oropharynx, the bronchio-pulmonary trectus, the esophagus, the stomach, the duodenum, the liver, bile ducts and the pancreas. The ciliated cyst is essentially supra-diaphragmatic and located in the bronchial tree, the esophagus, the mediastinum and sublingual. When the cyst is infra diaphragmatic, it’s usually located in the liver especially in medial segments (IV, V, VIII). In can be located in the pancreas as well. It’s location in the gallbladder is exceptional. Therefore, few cases were published. To our knowledge, only 15 cases were described in literature. We report the first case diagnosed in Tunisia and the 16th in the world with a review of literature. This work has been reported in line with the SCARE criteria [1].

2. Observation

A 34 years old patient with no medical history was admitted for an upper right quadrant pain. The clinical examination found right upper quadrant tenderness. Blood test showed no anomalies and the abdominal ultrasound showed a gallbladder stone with thin intra and extra hepatic bile and hiliar adenopathy. The abdominal computed tomography revealed unilocular cystic lesion attached to the neck of the gallbladder (Fig. 1). The patient was operated via a right subcostal incision. The paraopeartively exploration found a gallbladder with thin walls and a 2 cm round cystic mass attached to the neck of the gallbladder. She had a cholecystectomy and the follow ups were simple. The anatomoopathologic study found a cystic lesion with thin walls covered with ciliated epithelium which lays on a connective tissue that contains smooth muscle fibers (Figs. 2 and 3). The content of the cyst was a mucoid liquid and there was no communication between the cyst and the gallbladder lumen. There was no sign of degeneration.

3. Discussion

Cystic lesions of the gallbladder are rare and they can originally congenital, acquired or neoplastic [2]. The ciliated cyst of the gallbladder is a benign congenital lesion that develops from the anterior primitive intestine. Cysts are essentially supradiaphragmatic in the bronchial tree, the esophagus, the mediastinum and...
Sublingual. Under the diaphragm, the ciliated cysts are in the liver and develop from intrahepatic bile ducts. Its location in the gallbladder is exceptional.

There is many hypotheses that describes the pathogenesis of ciliated cysts [3]. Kakitsubata described in 1995 the first case of gallbladder cyst who was covered with cylindrical ciliated epithelium and he named it “Epithelial cyst of the gallbladder” [4]. In 2000, another case was reported by Nam under the title of “Ciliated foregut cyst of the gallbladder” [5]. Afterward, only twelve cases were published.

Tables 1 and 2 present a resume of literature review that permits to deduce the epidemiological and pathologic characteristics related to this entity.

This pathology affects women more than men (10 women to 5 men) with a sex ratio of 2. The median age is 45 years old with extreme ages of 9–75 years old. Only one case of an infant was noted. The median size of the cyst is 2.5 cm with extremes of 0.7–3.5 cm. The most frequent location is the neck, it was observed in 6 cases followed by the body with 4 observed cases. In three patients the cyst was located in the fundus. The location wasn’t mentioned in 2 cases. The most frequent clinical symptom is abdominal pain which was noted in 6 patients. In 5 cases, the discovery of the gallbladder cyst was incidental. On patient had cholecystitis and another one had epigastric pain and vomiting. In two observations, clinical symptoms weren’t mentioned. All patients underwent radiologic exams preoperatively. In two thirds of cases (11 cases), ultrasound revealed a cystic lesion. In one case, ultrasound revealed gallbladder stones and in another an acute cholecystitis. The aspect of diverticula or duplication of the gallbladder was observed in one case. In our patient, the gallbladder lesion was mistakenly interpreted as an adenopathy while CT-scan confirmed the presence of an aspect of a cyst. In 100% of cases, the cyst was unilocular. In the majority of cases, the cyst content was mucoid (9 patients) while gelatinous in 2 patients, dark brown in 2 others and milky in one patient.

The positif diagnosis of ciliated cyst is histological. On an anatomicopathologic level, the cystic lesion is characterized by a thin wall covered with a cylindrical ciliated mucus-secreting epithelium which lays on connective tissue containing smooth muscular fibers. There is now communication between the cyst and the gallbladder lumen.

Concerning imaging techniques, abdominal ultrasound is the fist most sought exam in exploring hepatic and biliary diseases. Although it is not very specific, it describes the ciliated cyst as a
cystic lesion often anechoic and sometimes hyperechogenic. In our patient, the abdominal ultrasound didn’t establish the diagnosis. When the lesion is hyperechogenic the diagnosis of a neoplastic origin can be evoked [6] especially when the cysts size is exceeds 1 cm. In this case the CT-scan as well as the abdominal MRI are very helpful of the diagnosis. The ciliated cyst is often observed in hyper-intensity T1 and T2 and certain cysts can appear in hypointensity T1. In fact, the contrast depends in parts on the viscosity of the cyst content and in other part on the presence or absence of calcium and cholesterol crystals.

Treatment is based on the cholecystectomy [7]. Although, Tuncyurek proposed a close surveillance when the cyst is silent [6]. The systematic cholecystectomy seems the most wise attitude since the distinction between benign and malignant lesion is difficult on imaging techniques and all the more that cases of malpigien metaplasia and epidermoid carcinoma that occurred on hepatic ciliated cysts were observed [8]. Even though no case of malignant transformation of the ciliated gallbladder cyst was described, it’s convenient to look thoroughly into the resection specimen.

### 4. Conclusion

The ciliated cyst of the gallbladder is a congenital rare cyst. The preoperative diagnosis is still difficult to establish despite the progress of imaging techniques. The positive diagnosis is established on anatomopathologic data. The recommended current treatment is surgery and it consists of a celioscopic cholecystectomy. The place of conservatory treatment hasn’t been established due to the rarity of described case and the possibility of degeneration.

### Conflicts of interest

There aren’t any conflicts of interest for all authors.

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Ethical approval

Ethical approval has been exempted by our institution.

Consent

Our study is a case report. The consentment of the patient has been obtained.

Author contribution

Wissem Triki: development of the idea and writing of the article.
Oussema Baraket and Ahmed Itami: bibliographic search
Ines Marzouk: provision and interpretation of radiological images.
Abdelmajid Baccar: provision and interpretation of anatomo-pathological images.
Sami Bouchoucha: final revision.

Registration of research studies

None.

Guarantor

Triki Wissem.

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