Scholars Journal of Medical Case Reports
Abbreviated Key Title: Sch J Med Case Rep
ISSN 2347-9507 (Print) | ISSN 2347-6559 (Online)
Journal homepage: https://saspublishers.com

OPEN ACCESS

Visceral Surgery

Gallbladder Agenesis Discovered Intraoperatively: About a Case

Hicham Krimou[1,2*], Samir Hasbi[1,2], Nizar Errabi[1,2], Mohammed Cisse[1,2], Mohammed Laaroussi[1,2], Abdessamad El Kaoukabi[1,2], Belhamidi Said[1,2], Mohammed Menfaa[1,2], Fouad Sakit[1,2], Abdelkarim Choho[1,2]

[1]Service of Visceral Surgery, Military Hospital Moulay Ismail, Meknes, Morocco
[2]Faculty of Medicine and Pharmacy of Fez, Fez, Morocco

DOI: https://doi.org/10.36347/sjmcr.2024.v12i08.019 | **Received:** 16.06.2024 | **Accepted:** 24.07.2024 | **Published:** 19.08.2024

***Corresponding author:** Hicham Krimou
Service of Visceral Surgery, Military Hospital Moulay Ismail, Meknes, Morocco

Abstract **Case Report**

Gallbladder agenesis is a rare, little-recognized congenital anomaly, presenting in the form of hepatic colic in 23% of patients. The ultrasound assessment often describes a scleroatrophic vesicle, which can lead to unnecessary and potentially dangerous surgery. We report the case of a 64-year-old man, with no history of cholecystitis, presenting with hepatic colic and whose ultrasound diagnosis of scleroatrophic vesicle was questioned. patient was operated on by the traditional route for gallbladder lithiasis. The absence of a gallbladder was discovered intraoperatively. In order to confirm the diagnosis postoperatively, we performed magnetic resonance imaging (cholangio-MRI) which made the diagnosis of agenesis of the gallbladder.

Keywords: Agenesis, Gallbladder, Congenital, Gallbladder Imaging.

1. INTRODUCTION

Agenesis of the gallbladder is a rare congenital anomaly. A retrospective autopsy study reported a prevalence of 0.03%, most often associated with other malformations. Only 23% of patients exhibit hepatic colic. Routine radiological assessments rarely suggest this diagnosis, often interpreting the observed images instead as a sclerotic-atrophic gallbladder. This can lead to unnecessary and potentially dangerous surgical interventions. Therefore, it is important to consider this diagnosis in cases of unusual presentations of hepatic colic. We report the case of a men in whom the diagnosis was established intraoperatively.

2. OBSERVATION

A 64-year-old patient without a pathological history has been presenting with paroxysmal right hypochondrium pain of gravity type for 2 months, unrelated to food intake. The biological balance is normal; in particular, there is no cholestasis, no cytolysis, and no leukocytosis. An objective abdominal ultrasound reveals a dense echo with a posterior shadow cone measuring 13 mm wide in the gallbladder bed, indicating a lithiasic scleroatrophic gallbladder. There is no dilation of the intra- and extra-hepatic biliary tract. A CT scan was not performed.

Given the appearance of the scleroatrophic gallbladder, a cholecystectomy by laparoscopic approach was planned, but it was later decided to proceed with a cholecystectomy via the right subcostal approach. Intraoperatively, the hepatic pedicle was well individualized, but the gallbladder was not visualized in its usual position, nor in the sites of gallbladder ectopy (Figure 1). The calcification described on ultrasound was not found. Intraoperative cholangiography was not deliberately performed.

During follow-up, the patient underwent an abdominal CT scan post-surgery to confirm gallbladder agenesis. No other associated anomalies were found intraoperatively or on the scan. He received a cholangio-MRI, which conclusively showed gallbladder agenesis with the absence of the cystic duct and no other associated biliary abnormalities (Figure 2).

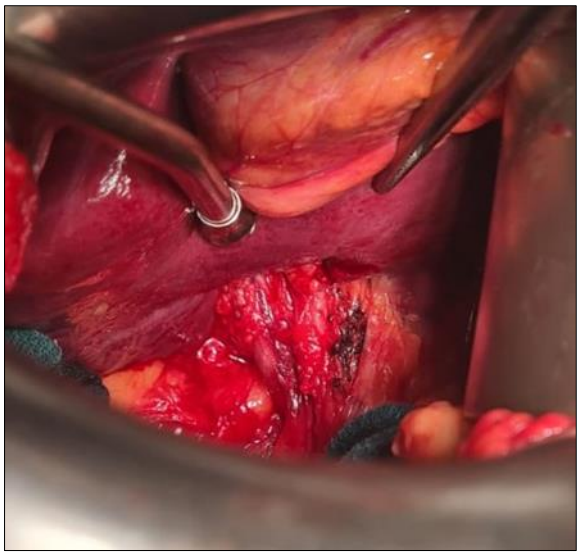

Fig 1: Intraoperative image showing absence of gallbladder in position

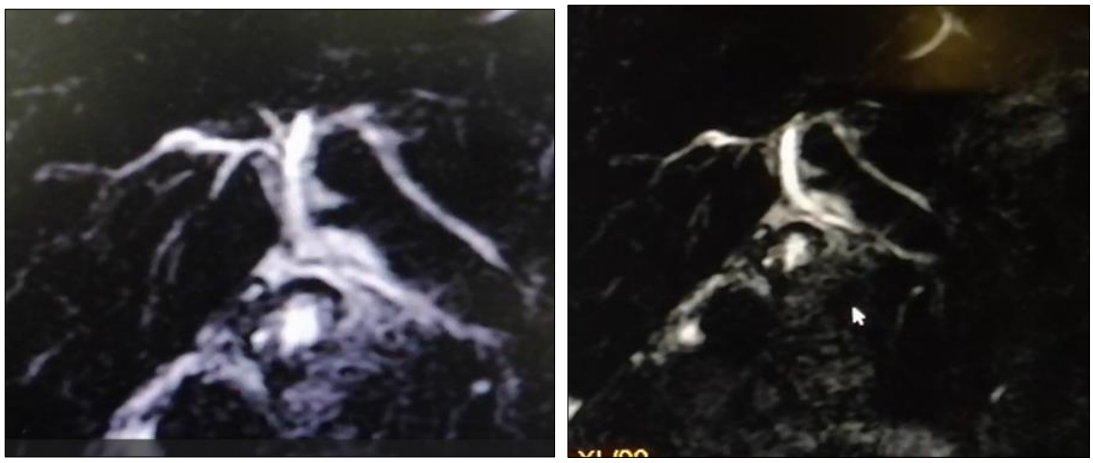

Fig. 2: From a cholangio-MRI conclusive to gallbladder agenesis with absence of cystic duct

3. DISCUSSION

Gallbladder agenesis is an extremely rare embryological aberration with an incidence ranging from 0.01% to 0.075% (10 to 75 per 100,000 population) [2, 3]. The prevalence is estimated between 1 and 10 per 10,000 people in the general population [4]. The sex ratio is 1 for cases discovered at autopsy [1], but the female-male ratio is 3:1 in clinical studies, probably because the symptomatology is similar to that of biliary lithiasis which is more common in women [5], in our case, we have a man at 64-year-old.

From an embryological point of view, extrahepatic bile ducts develop from the anterior intestine, in the ventral pancreas. The first pathophysiological hypotheses suggested an anomaly of vascularization of the gallbladder outline, explaining its absence [6]. More recently, genetic transcription factors have been shown to be necessary for gallbladder development: Sox17+, Pdx1+ [7], HNF6 [8], and HNF1b [9], located respectively on chromosomes 8, 13, 15, and 17. Their absence leads to gallbladder agenesis and other biliary tract abnormalities [7-10].

It should be noted that gallbladder agenesis is usually accompanied by cystic duct agenesis [11].

Patients with an intraoperative diagnosis were wrongly operated on for gallbladder lithiasis [14, 15]. These patients can be operated on either conventionally or laparoscopically. During the procedure, the different ectopic sites of the gallbladder should be thoroughly examined, ideally with the help of an intraoperative ultrasound if available [13]. Intraoperative cholangiography by puncture of the choledochus will also not be carried out deliberately due to the high risk of biliary tract or portal vein injury during cholangiography [16]. Its necessity is also questioned. In all cases, it must be supplemented by bili-MRI postoperatively to confirm the diagnosis [17]. It can detect gallbladder agenesis [12-14]. Bili-MRI is a non-invasive imaging examination that allows the detection of a gallbladder in an ectopic site (e.g., in the falciform ligament, the small omentum, at the level of the pancreas, behind the duodenum, in the

pyloric digestive wall, or even intrahepatic). If no gallbladder is found, it can make the correct preoperative diagnosis of agenesis.

For asymptomatic patients, no treatment is required. In patients with biliary pain related to Oddi sphincter dysfunction, treatment includes either medical treatment primarily represented by smooth muscle relaxants used to relieve discomfort [18], or endoscopic sphincterotomy.

4. CONCLUSION

Gallbladder agenesis is a very rare condition caused by an aberration in embryological development. It can be associated with other congenital anomalies but is most often discovered in isolation in adulthood. It should be considered when there is an absence of gallbladder visualization on ultrasound, or more commonly, when a scleroatrophic appearance is observed. Biliary MRI allows for a definitive diagnosis and avoids surgical intervention, which could pose unnecessary risks to the patient. There is no specific treatment for gallbladder agenesis itself, but treatment is directed towards associated lesions, particularly choledocholithiasis and sphincter of Oddi dysfunction**.**

Conflicts of Interest: The authors declare no conflicts of interest.

REFERENCES

1. Turkel, S. B., Swanson, V., & Chandrasoma, P. A. R. A. K. A. R. M. A. (1983). Malformations associated with congenital absence of the gall bladder. *Journal of Medical Genetics*, *20*(6), 445-449.
2. Serralta de Colsa, D., Arjona Medina, I., Quirós Higueras, E., Sanz Sánchez, M., Díaz, P., & Turégano Fuentes, F. (2006). Gallbladder agenesis: diagnosis of a case without surgical intervention. *Spanish Journal of Digestive Diseases*, *98* (12), 964-965.
3. Singh, B., Satyapal, K. S., Moodley, J., & Haffejee, A. A. (1999). Congenital absence of the gall bladder. *Surgical and Radiologic Anatomy*, *21*(3), 221-224.
4. Muguruma, N., Okamura, S., Ichikawa, S., Tsujigami, K., Suzuki, M., Nakasono, M., ... & Ito, S. (2001). Asymptomatic case of congenital absence of the gallbladder. *JOURNAL OF MEDICAL INVESTIGATION*, *48*(1/2), 118-121.
5. Lamah, M., Karanjia, N. D., & Dickson, G. H. (2001). Anatomical variations of the extrahepatic biliary tree: review of the world literature. *Clinical Anatomy: The Official Journal of the American Association of Clinical Anatomists and the British Association of Clinical Anatomists*, *14*(3), 167-172.
6. Fiaschetti, V., Calabrese, G., Viarani, S., Bazzocchi, G., & Simonetti, G. (2009). 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*(1), 674768.
7. Zong, Y., & Stanger, B. Z. (2011). Molecular mechanisms of bile duct development. *The international journal of biochemistry & cell biology*, *43*(2), 257-264.
8. Clotman, F., Lannoy, V. J., & Reber, M. (2002). The onecut transcription factor HNF6 is required for normal development of the biliary tract. *Development*, *129*, 1819–28.
9. Coffinier, C., Gresh, L., & Fiette, L. (2002). Bile system morphogenesis defects and liver dysfunction upon targeted deletion of HNF1beta. *Development*, *129*, 1829–38.
10. Keplinger, K. M., & Bloomston, M. (2014). Anatomy and embryology of the biliary tract. *The Surgical clinics of North America*, *94*(2), 203-217.
11. Cockbain, A. J., Watson, J., & Toogood, G. J. (2011). Hepatobiliary and Pancreatic: Agenesis of the gallbladder. *Journal of Gastroenterology & Hepatology*, *26*(5).
12. Fiaschetti, V., Calabrese, G., Viarani, S., Bazzocchi, G., & Simonetti, G. (2009). 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*(1), 674768.
13. Chowbey, P. K., Dey, A., Khullar, R., Sharma, A., Soni, V., Baijal, M., & Mittal, T. (2009). Agenesis of gallbladder—our experience and a review of literature. *Indian Journal of Surgery*, *71*, 188-192.
14. Malde, S. (2010). Gallbladder agenesis diagnosed intra-operatively: a case report. *Journal of medical case reports*, *4*, 1-5.
15. Gupta, N., Gupta, S. K., & Kapoor, H. S. (2010). Gallbladder and cystic duct agenesis diagnosed laparoscopically. *Hepatobiliary & Pancreatic Diseases International: HBPD INT*, *9*(1), 107-108.
16. Cano-Valderrama, O., Talavera, P., Dominguez-Serrano, I., Sanchez-Pernaute, A., & AJ, T. G. (2011). Gallbladder agenesis. Presentation of a case. *Cirugia Espanola*, *89*(7), 471-472.
17. Balakrishnan, S., Singhal, T., Grandy-Smith, S., & El-Hasani, S. (2006). Agenesis of the gallbladder: lessons to learn. *JSLS: Journal of the Society of Laparoendoscopic Surgeons*, *10*(4), 517.
18. Kwon, A. H., Yanagimoto, H., Matsui, Y., & Imamura, A. (2006). Agenesis of the gallbladder with hypoplastic cystic duct diagnosed at laparoscopy. *Surgical Laparoscopy Endoscopy & Percutaneous Techniques*, *16*(4), 251-254.