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Case Report

Choledocele, A Rare Entity of Congenital Dilatation of the Bile Duct: A Case Report

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Abstract

Introduction: Choledocele, first reported by Wheeler in 1940, is classified as type 3 of congenital biliary dilatation according to Todani's classification. It is considered the rarest form. We report a case of choledocele in a 10-year-old boy.

Observation: A 10-year-old boy with no history of pain or jaundice was admitted with acute pancreatitis. Clinical examination revealed no palpable mass. Radiological findings revealed an intraduodenal cystic image with a digestive wall, initially suggestive of a digestive duplication. However, given the location of the lesion, the hypothesis of congenital dilatation of the main bile duct (DCVBP) was also considered. After treatment of the pancreatitis, surgical exploration via duodenotomy confirmed the diagnosis of choledochocele. The operation revealed a cystic formation on the inner wall of the second duodenum, centred by a bile-flowing orifice. Treatment consisted of a simple incision of the choledocele. A biliodigestive bypass was not considered necessary due to the low risk of degeneration associated with this type of DCVBP. Postoperative follow-up was straightforward. After 6 months, the patient was asymptomatic and his pancreatic work-up was normal.

Conclusion: This rare case of choledocele illustrates the diagnostic and therapeutic challenges of this pathology, underlining the importance of an individualized approach. Treatment by simple incision proved effective, but long-term follow-up remains essential to monitor possible late complications.

Keywords: *Congenital Dilatation of the Bile Ducts; Choledocele*

Introduction

Congenital dilation of common bile duct is a rare malformation of the bile ducts. Several types have been described, the most recognized classification being Todani's, which includes five types of dilations. Choledococele represents type 3 of congenital dilatations of the main bile duct (DCVBP). However, its clinical presentation, pathophysiology and management make it a distinct pathological entity, justifying a specific approach. We report one case of choledococele in a 10-year-old boy whom we took care of in our department.

Observation

A 10-year-old boy, with no significant medical history, including no history of intermittent pain or jaundice, was admitted with acute pancreatitis. The main symptoms were epigastric pain associated with elevated pancreatic enzymes. Physical examination did not reveal any palpable mass. Imaging exploration included ultrasound and computed tomography (CT). These examinations revealed an intraduodenal cystic image with a digestive wall (Figure 1) and a swelling of the pancreas (Figure 2). These results initially led to suspicions of digestive duplication. However, given the location of the lesion on the inner wall of the second duodenum, the differential diagnosis of DCVBP has been suggested, despite the absence of visible dilation of the bile ducts.

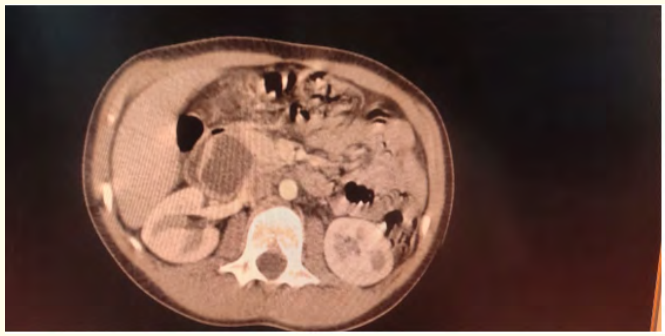

Figure 1: *Abdominal CT showing infraduodenal cystic formation.*

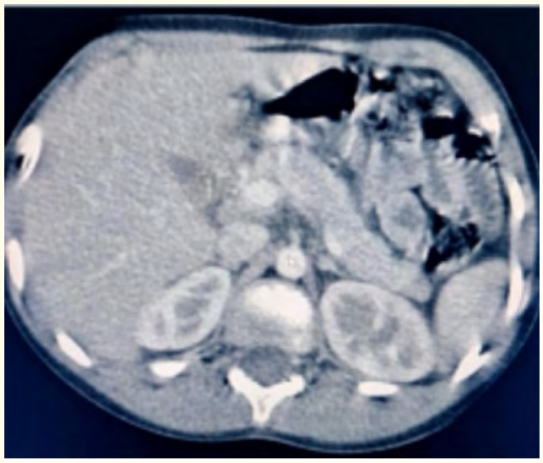

Figure 2: *Abdominal CT showing Balthazar's stage C pancreatitis.*

After the initial treatment of pancreatitis, surgical exploration was performed: duodenotomy approach, discovery of a cystic formation on the inner wall of the second duodenum (Figure 3), identification of a central orifice giving outlet to bile (Figure 4), confirming the diagnosis of choledococele. Surgical treatment consisted of a simple incision of the choledococele. A biliodigestive diversion was not considered necessary, given the low risk of degeneration associated with this specific type of BPDC. The immediate post-operative effects were simple. At 6 months postoperatively, the patient was asymptomatic and the pancreatic work-up had returned to normal.

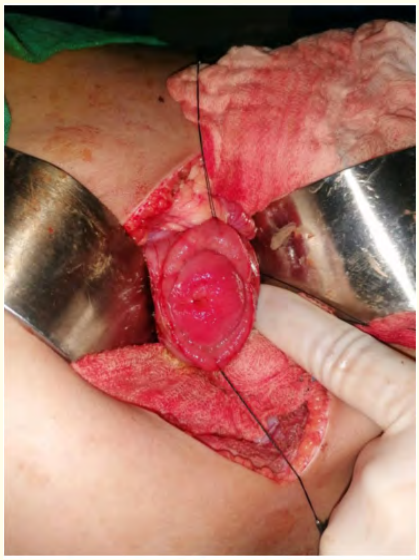

Figure 3: *Intraoperative image of the cystic mass of the inner wall of the duodenum centered by an orifice.*

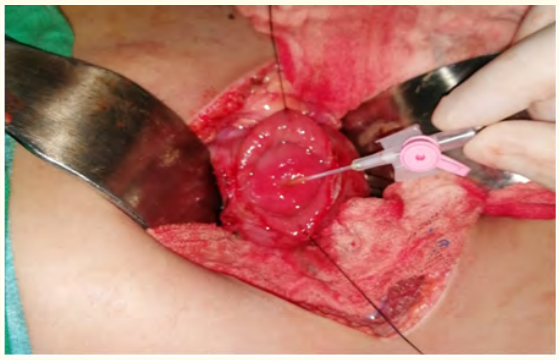

Figure 4: *Catheterization of the orifice giving outlet to bile.*

Patient perspective

The 10-year-old patient, initially admitted for acute pancreatitis, had an unexpected medical experience with the diagnosis and treatment of his choledococele. Although he had no history of pain or jaundice, the sudden onset of epigastric pain must have been a cause for concern for him and his family. The diagnostic process, involving imaging tests and surgical exploration, was likely a stressful time. However, the success of the surgery, with simple postoperative follow-up, undoubtedly brought great relief. Six months after the operation, the patient is asymptomatic and his pancreatic assessment is normal, suggesting satisfaction with the outcome of the treatment and a significant improvement in his quality of life.

Informed consent

Informed consent was obtained from the patient's parents for the publication of this clinical case and associated images. Parents have been fully informed about the nature of their child's condition, the treatment options available, and the potential risks and benefits of

surgery. They also agreed to the anonymised use of medical data and images for educational and scientific publication purposes. This process has been documented in accordance with the institution's policies and ethical standards.

Discussion

Choledococele, although classified as type 3 of DCVBP according to Todani, has distinct characteristics that make it a pathological entity in its own right [1]. These particularities concern: clinical presentation, evolution, diagnosis, pathophysiology and management [2]. Unlike other types of CVDP, choledococele, it is predominantly male and the diagnosis is made in adulthood. A systematic review published in 2017 identified only 13 pediatric cases, highlighting the rarity of this pathology in children [3]. Pain is the main symptom [4]. Other symptoms are less common: vomiting, nausea, fever, and jaundice [3,4]. The pathology is usually revealed by a picture of acute pancreatitis, as in the case presented. Diagnosis is mainly based on imaging, including: ultrasound, computed tomography and bili-MRI. Bili-MRI is considered the key examination, providing accurate mapping of bile duct and pancreatic tract terminations on the second duodenum. In our case, this examination could not be carried out due to a lack of available resources.

The main differential diagnosis to consider is duodenal duplication. This has distinct anatomical characteristics: location generally on the mesenteric edge, common wall with the duodenal loop, variable communication with the duodenal lumen. Specific radiological criteria make it possible to differentiate duodenal duplication from choledococele [5]. According to Sarris, bile mucocele can be classified into two main types, with three subtypes for type A: common opening of the Wirsung duct and the common bile duct in the cyst for type A1, separate openings for the Wirsung duct and the common bile duct in the cyst for type 2, small intramural cyst for type A3. In type B, the ampulla flows directly into the duodenum, and the cyst forms a distal diverticulum protruding into the duodenal lumen.

The treatment of choledococeles is the subject of debate within the surgical community. Two main approaches are currently being used. Some surgeons opt for a treatment similar to that of other types of DCVBP by a biliodigestive diversion with complete resection of the extrahepatic bile duct justified by the risk of degeneration in adulthood. Other practitioners advocate a more conservative approach, taking into account the particularities of choledococele. They perform a simple incision of the choledococele without resection of the bile duct, justified by the low rate of degeneration reported in children in the literature (only 4 documented cases). The risk of degeneration is age-related, with a rate of 1% in children under 10 years of age and 15% in patients over 20 years of age [6].

A recent study demonstrated the normalization of the histological appearance of the choledococele mucosa on two biopsies performed less than six months after the cyst incision [2]. These results suggest that a simple incision of the choledococele, without resection of the bile duct, may be sufficient, making biliopancreatic shunt unnecessary. Surgical treatment varies depending on the type of choledococele. For types A1 and A2, excision of the intraluminal portion of the cyst, preserving the part in contact with the papilla, is considered the best treatment. In the case of type A3, sphincteroplasty may be sufficient, while for type B, excision of the cystic wall combined with sphincteroplasty is often necessary. During surgery, it is crucial to precisely locate the papilla and the accessory pancreatic duct to avoid any damage. Intraoperative cholangiography is usually performed before and after cyst excision to assess the extent of the lesions and check for associated lithiasis [7]. The surgical approach generally involves a duodeno-pancreatic detachment, followed by the opening of the second duodenum with regard to the swelling. Choledococele is then identified and opened. Hemming from the walls to the duodenal mucosa is often necessary to ensure adequate drainage of the biliopancreatic sectors, thereby reducing the risk of pancreatic reflux to the bile ducts.

In recent years, the management of choledococeles has been revolutionized by the advent of endoscopic treatments. Since the first endoscopic sphincterotomy described by Dehyle., *et al*. In 1974 [8], this approach became the standard treatment for many authors [9,10]. Endoscopic techniques include endoscopic sphincterotomy, excision of the cyst at the diathermic loop, or a combination of these two [9,11]. Endoscopic treatment has several advantages: it is a minimally invasive method that saves the patient from major surgery,

and it allows simultaneous diagnosis and treatment, especially for the extraction of common bile duct stones, present in 20% of cases. In the case presented, surgical treatment was chosen, consisting of an incision of the choledococele with hemming of the banks (Figure 5) to ensure effective drainage of bile and pancreatic juice into the duodenal lumen. Postoperative follow-up generally includes a clinical assessment to check that the pain has disappeared, a laboratory assessment to confirm that pancreatic enzymes have returned to normal, and an ultrasound to ensure that the cystic lesion has disappeared. Long-term follow-up is essential to detect any potential complications early, including the development of hepatobiliary cancer. In general, the aftermath of the operation is simple, but long-term monitoring is still required to ensure that there is no silent development of hepatobiliary cancer. This comprehensive approach, combining targeted intervention and rigorous follow-up, allows for optimal management of patients with choledococele, minimizing the risk of short- and long-term complications.

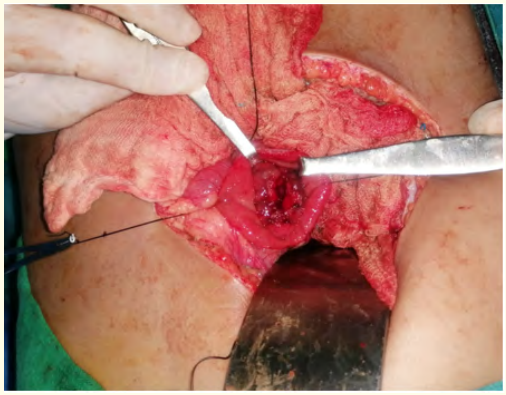

Figure 5: *Appearance of choledococele after hemming of the banks.*

Conclusion

Choledococele, although classified as a form of congenital dilation of the bile ducts, is distinguished by its unique clinical, pathophysiological, and therapeutic features. The case of a 10-year-old boy illustrates the rarity of this pathology in paediatrics and underlines the importance of an accurate diagnosis, particularly in the face of atypical presentations such as acute pancreatitis. Diagnosis is mainly based on imaging, with a preference for bili-MRI when available. Therapeutic management is the subject of debate, opposing traditional surgical approaches to less invasive endoscopic techniques. In the present case, a conservative surgical approach was chosen, consisting of a simple incision of the choledococele, reflecting the current trend to favor less aggressive interventions in younger patients, given the low risk of malignant degeneration. The evolution of endoscopic techniques offers promising prospects for the treatment of this pathology, often allowing simultaneous diagnosis and treatment. However, regardless of the approach chosen, long-term follow-up remains essential to monitor for possible late complications.

What is known on this subject:

- Choledococele is a rare form of congenital dilation of the bile ducts, classified as type 3 according to the Todani classification.
- Diagnosis is mainly based on imaging, with a preference for bili-MRI when available.

What is new in your study:

- This case illustrates the atypical presentation of choledococele in a 10-year-old child in the form of acute pancreatitis, highlighting the importance of including this pathology in the etiological diagnosis of pancreatitis in children.

- The study demonstrates the efficacy of a conservative surgical approach (simple choledococele incision) in a young patient, reflecting the current trend towards less aggressive interventions given the low risk of malignant degeneration at this age.

Conflicts of Interest

The authors do not declare any conflicts of interest.

Authors' Contributions

Khadija ElBied: Data collection, bibliographic research and manuscript writing. Fadwa Idrissi Fawzi: Manuscript validation. Anas Abouelkheir Zineb Hammoumi, Nadir Ferram, Mounia Al Zemmouri: Proofreading of the manuscript. All authors have read and approved the final version of the manuscript.

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