

# Choledochal Cyst in Children in Dakar: Diagnostic and Therapeutic Aspects

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## Abstract

**Context:** Choledochal cyst is a rare malformation that mainly affects girls. **Aims:** The aim of this work is to report the diagnostic, therapeutic and evolutionary aspects of bile duct cyst in children in Dakar. **Subjects and Methods:** we conducted a prospective monocentre descriptive study from 1 July, 2016, to 30 June, 2019, in the Pediatric Surgery Department of Albert Royer Children's Hospital in Dakar. The studied parameters featured clinical, biological, radiological, therapeutic and evolutionary data. Overall, ten patients, including eight girls and two boys, were selected. **Results:** Two patients presented the classic triple-syndrome complex featuring an abdominal pain, an abdominal mass and jaundice. Most of the patients presented a symptomatology associating abdominal pain and vomiting. Biological cholestasis syndrome was present in six cases and biological cytolysis syndrome in five cases. The abdominal ultrasound-computed tomography (CT) scan confirmed the diagnosis and helped to set the Todani classification with a predominance of Types 1 and 4. Surgical treatment by laparotomy consisted of total excision of the cyst followed by a Y-en-Roux hepatico-jejunal anastomosis in nine cases, whereas we performed a drainage of the cyst in one case. Two patients presented a morbidity of anastomotic suture release and evisceration and had a good outcome after surgical repair. There was no mortality. **Conclusions:** The diagnosis of choledochal cyst dilatation (CCD) can be suspected in the postnatal period on the basis of a gastro-intestinal symptomatology with or without associated to jaundice. The combination ultrasound-abdominal CT-scan helps set the diagnosis. The laparotomic approach always has its place for the management of CCD, especially in our context.

**Keywords:** Bile ducts, child, dilation, malformation, surgery

## INTRODUCTION

Choledochal cyst dilatation (CCD) is an uncommon congenital abnormality. Its prevalence varies according to populations with a higher frequency in Asians.<sup>[1,2]</sup> It mainly occurs in females and constitutes the second cause of biliary duct congenital malformation after biliary atresia.<sup>[3]</sup> In developed countries, prenatal diagnosis allows an early management of this condition. In developing countries, the diagnosis is set after birth with sometimes a broad clinical spectrum. The aim of this paper is to outline the therapeutical and diagnostic aspects of choledochal cyst in children in Dakar.

## SUBJECTS AND METHODS

We conducted a multicentre prospective and descriptive study ranging from 1 July 2016, to 30 June 2019. All children who had a CCD and who underwent a surgical treatment at

the Pediatric Surgery Department of Albert Royer Children Hospital in Dakar were included.

The studied parameters comprised the clinical course, biological and radiological findings, Todani classification, surgical time limit, technical approach, surgical exploration data, procedures, morbidity and mortality. Our series accounted for eight girls and two boys, meaning a sex ratio of 0.25. The mean age of our patients during their admission was 85.3 months (7.1 years), with extremes ranging from 6 months to 15 years.

## RESULTS

The clinical course mainly featured abdominal pain and vomiting that, respectively, occurred in nine and eight patients. Physical

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examination revealed a mass on the right hypochondrium and/ or epigastrium on five patients and jaundice in four patients with two cholestatic patterns. The triple-syndrome complex comprising pain, jaundice and abdominal mass was present in two patients. Figure 1 shows the various clinical signs presented by the patients. The most commonly associated signs were abdominal pain and vomiting, as shown in Table 1.

Biological cholestatic syndrome was present in six cases, with total bilirubin levels ranging from 2 to 29 times the normal rate, direct bilirubin levels from 2 to 55 times the normal rate and alkaline phosphates from 2 to five times the normal levels. Biological cytolysis syndrome was noted in five cases, with transaminases levels reaching 38 times the normal rate.

The ultrasound, performed in all patients, allowed to evoke the diagnosis in all cases. Once an ultrasonographic diagnosis of choledochal cyst was provided, abdominal computed tomography CT-scan was performed and showed a choledochal cyst Type 1 in six cases, Type 4 in three cases

Symptoms	n (%)
Abdominal pain + vomiting	4 (40)
Abdominal pain + jaundice + abdominal mass	2 (20)
Abdominal pain + abdominal mass + vomiting	2 (20)
Abdominal pain + vomiting + jaundice	1 (10)
Jaundice + vomiting + abdominal mass	1 (10)

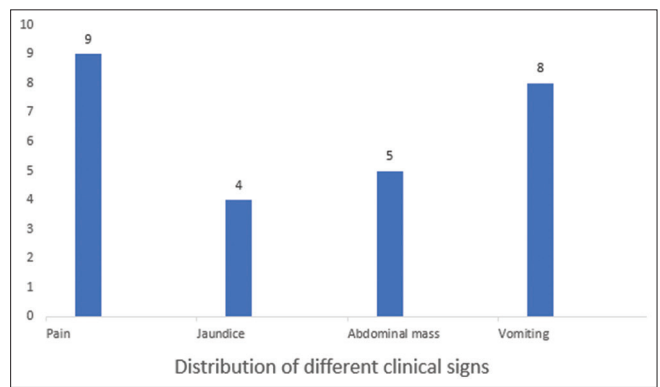


Figure 1: Distribution of clinical signs

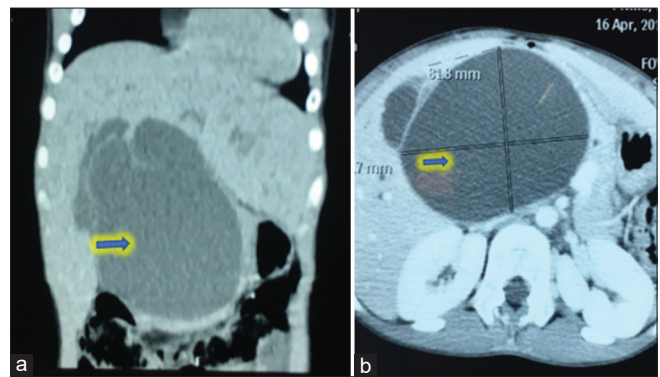


Figure 3: Choledochal cyst Todani type 4 (arrow) (a): Sagittal section. (b): Transverse section

and Type 3 in one case, according to Todani classification. Figures 2 and 3 show the most common types.

The mean surgical time limit was 3 months and 4 days, with extremes ranging from 1 day to 11 months and 19 days. Open surgery was realised for all patients. Right sub-costal approach was performed in eight cases, whereas the median sub-umbilical approach was performed in two cases. Surgical exploration revealed a macroscopic healthy liver in eight cases, a macroscopic cholestatic liver in one patient and a macroscopic cirrhotic liver associated with ascites in one patient, and helped confirm the types according to Todani. Figures 2 and 3, respectively, show Types 1 and 4 that represent the most common patterns.

Nine patients underwent a total cyst excision with Roux-en-Y hepatico-jejunal transmesocolic anastomosis [Figures 4 and 5]. A patient had an external derivation of the cyst.

Two patients went through a morbidity, indicating a surgical repair, for an anastomotic suture release and evisceration. They both had a good outcome after surgical reintervention. We did not record mortality.

**DISCUSSION**

In general, in CCD condition, the triple-syndrome complex features abdominal pain, jaundice and abdominal mass. Nevertheless, it is not steady in children.<sup>[4-6]</sup> In fact, out of our ten patients, only two had a complete triad. The most common clinical courses included abdominal pain and vomiting.

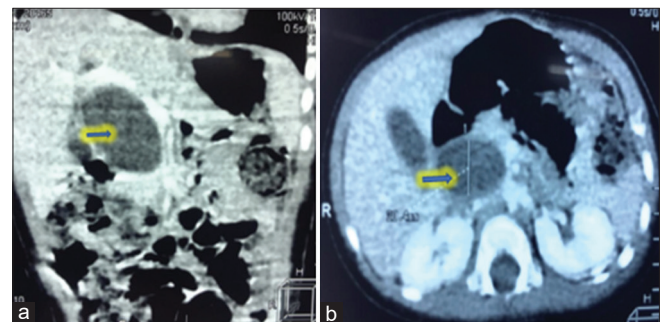


Figure 2: Choledochal cyst Todani type 1 (arrow) (a): Sagittal section. (b): Transverse section

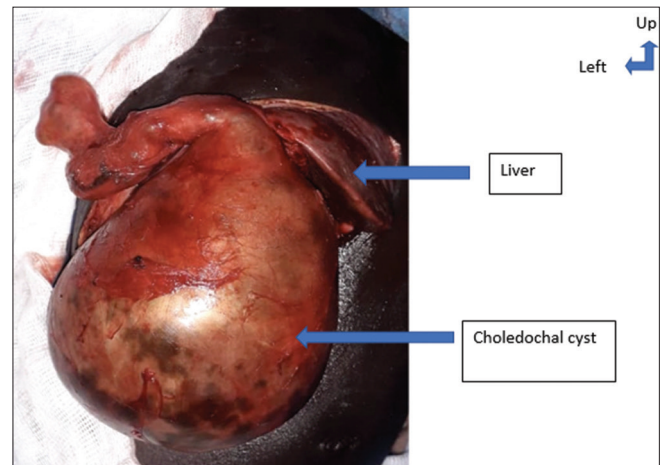
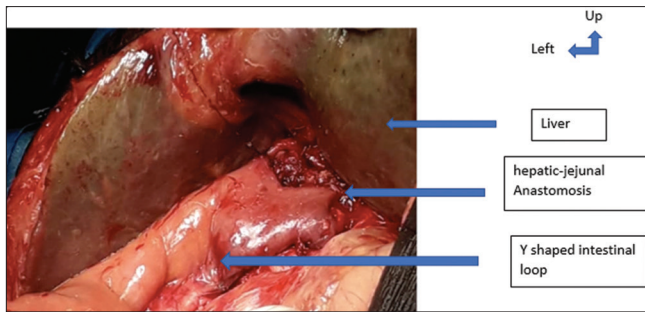


Figure 4: Pre-operative picture of choledochal cyst type 1



**Figure 5:** Image after cyst excision and hepatic-jejunal anastomosis

The ultrasound-abdominal CT-scan combination allowed to set both the diagnosis and Todani classification. However, besides the exploration of large cysts, the combination of ultrasound and magnetic resonance imaging is a rational strategy and helps reduce radiations.<sup>[7]</sup> Most of the authors recommend early management during the 1<sup>st</sup> months of life to avoid complications because hepatic cirrhosis can occur precociously.<sup>[8]</sup> A case of liver failure was reported in an 5-month-old toddler.<sup>[9,10]</sup> Our data suggest a management delay for our patients compared to that of the literature.<sup>[2,9]</sup> This could be explained by the absence of prenatal diagnosis in our context, late admission and lack of knowledge of this condition by some practitioners.

The treatment of CCD is surgical.<sup>[11,12]</sup> The approach may be through laparotomy or laparoscopy with similar results.<sup>[11]</sup> In fact, Cherqaoui *et al.*<sup>[13]</sup> reported in a series of 19 patients that a laparoscopic approach of CC is possible and safe, even on toddler who had a prenatal diagnosis, with a low morbidity and identical operative suites to laparotomy. In our series, laparotomic surgical exploration was performed for all patients. It allowed to confirm the predominance of Type 1 of Todani classification, in consonance with that of the literature data.<sup>[4,6,11]</sup>

Several surgical treatments have been proposed for the management of choledochal cyst. Historically, Types 1, 2 and 4 underwent a cysto-enterostomy, resulting in a high incidence of recurrent cholangitis and stenosis.<sup>[14]</sup> Furthermore, this procedure does not withdraw the cyst mucosa which can lead to biliary duct carcinomas.<sup>[14]</sup>

Currently, the most admitted surgical approach is total excision of the enlarged portion of the biliary duct followed by a hepatico-duodenal or hepatico-jejunal anastomosis.<sup>[15-17]</sup> For many authors, the technique of choice remains the anastomosis of the biliary duct with the jejunum through a Y-shaped loop.

Our complication rate is similar to the data from the literature, witnessed by the series provided by Khmekhem *et al.*<sup>[11]</sup> and Cherqaoui *et al.*,<sup>[13]</sup> who, respectively, reported a complication rate of 37.5% and 5.2%.

The mortality varies in the literature but has essentially decreased these last years and has almost become zero in a recent series.<sup>[13,18,19]</sup> After a follow-up of 24 months, no deaths were recorded.

## CONCLUSIONS

The diagnosis of CCD can be suspected in the postnatal period on the basis of a gastro-intestinal symptomatology featuring abdominal pain and vomiting with or without associated to jaundice. Ultrasound-abdominal CT-scan combination helps set the diagnosis. Laparotomic approach always has its place for the management of CCD especially in our context. Bilio-digestive derivation through Roux-en-Y performed after total excision of cyst yields good outcomes.

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## Conflicts of interest

There are no conflicts of interest.

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