

Original Research Report

PRIMARY HEALTHCARE PROVIDERS' KNOWLEDGE ON THE EARLY DETECTION OF BILIARY ATRESIA

Rendi Aji Prihaningtyas¹ , Bagus Setyoboedi^{1*} , Ni Nyoman Metriani Nesa², Melinda Masturina¹ 
 , Martono Tri Utomo¹ , Sjamsul Arief¹ 

¹Department of Child Health, Dr. Soetomo General Academic Hospital; Faculty of Medicine, Universitas Airlangga, Surabaya, Indonesia

²Department of Child Health, Faculty of Medicine, Universitas Udayana, Denpasar, Indonesia

ABSTRACT

Primary healthcare providers frequently fail to recognize biliary atresia because it closely mimics physiological jaundice. Early detection plays an important role in ensuring the optimal treatment of biliary atresia. Delays in referring to biliary atresia cases remain a problem around the world. The objective of this study was to analyze the knowledge of primary healthcare providers regarding prolonged jaundice as an early sign of biliary atresia. A cross-sectional study was conducted at primary health facilities using a self-administered questionnaire. The data were analyzed using the descriptive method with the assistance of IBM SPSS Statistics for Windows, version 21.0 (IBM Corp., Armonk, N.Y., USA). A total of 271 respondents participated in this study, including midwives (63.8%), nurses (24.4%), and medical doctors (8.5%). It was found that 73.8% of the respondents agreed that infants with prolonged jaundice need to undergo a serum bilirubin level assessment. However, 40.2% of the respondents believed that pale stools and dark urine in yellow infants were symptoms of physiological jaundice. Concerning the definition of prolonged jaundice, 209 (77.1%) respondents provided a response indicating a duration of more than two weeks. A total of 137 (50.6%) respondents stated that biliary atresia is a disease that must be ruled out first in cases of prolonged jaundice. Nevertheless, 86 (31.7%) and 87 (32.1%) respondents also mentioned that breastfeeding jaundice and breast milk jaundice must be considered as potential causes that need to be ruled out, respectively. These results indicated that primary healthcare providers still had limited knowledge regarding cholestasis. Education on prolonged jaundice in primary health facilities requires improvement to enhance the early detection of biliary atresia.

Keywords: Biliary atresia, primary healthcare provider, education, early detection, child mortality

***Correspondence:** Bagus Setyoboedi, Department of Child Health, Dr. Soetomo General Academic Hospital; Faculty of Medicine, Universitas Airlangga, Surabaya, Indonesia. Email: bagus.setyoboedi@fk.unair.ac

Article history

• Submitted 7/8/2023 • Revised 29/9/2023 • Accepted 27/11/2023 • Published 10/3/2024

How to cite: Prihaningtyas RA, Setyoboedi B, Nesa NNM (2024). Primary Healthcare Providers' Knowledge on the Early Detection of Biliary Atresia. *Folia Medica Indonesiana* 60 (1), 85-93. doi: <https://doi.org/10.20473/fmi.v60i1.48446>



Copyright: © 2024 Folia Medica Indonesiana.

This is an open-access article distributed under the terms of the Creative Commons Attribution License as stated in <https://creativecommons.org/licenses/by-nc-sa/4.0/deed.id>.

pISSN:2355-8393, eISSN: 2599-056x

Highlights:

1. An evaluation is necessary to verify that the knowledge of healthcare providers is sufficient for the early detection of biliary atresia.
2. It is crucial to promote education on prolonged jaundice to ensure early detection of biliary atresia in primary health facilities, as some primary healthcare providers have insufficient knowledge on the subject.

INTRODUCTION

As described by Mitra & Rennie (2017), jaundice is a medical condition characterized by elevated amounts of bilirubin in the bloodstream, resulting from the breakdown of erythrocytes. Physiological jaundice is a common occurrence among neonates. The condition of jaundice that manifests or continues beyond 14 days after birth is referred to as

prolonged jaundice. It is possible that there is an association between prolonged jaundice and the etiology of diseases. Therefore, further assessment is necessary in cases of prolonged jaundice (Menz et al. 2019).

Breast milk jaundice is recognized as the prevailing etiology of prolonged jaundice, which is characterized by an extended duration of

unconjugated hyperbilirubinemia. Breast milk jaundice is generally considered a benign condition and is observed in approximately one-third of breastfed newborns (Tan et al. 2019). However, chronic jaundice can also be caused by various other conditions, such as infectious diseases and hematological, hepatobiliary, metabolic, endocrine, and genetic abnormalities, including biliary atresia. Primary healthcare professionals sometimes struggle to identify cases of biliary atresia, as it can be easily mistaken for physiological jaundice. The diagnosis of breast milk jaundice can only be established by ruling out other pathogenic causes. It is necessary to assess the concentration of conjugated bilirubin in infants who experience prolonged jaundice (Mitra & Rennie 2017).

Additional research is warranted to analyze instances of persistent jaundice resulting from conjugated hyperbilirubinemia. The incidence of conjugated hyperbilirubinemia is estimated to be one in every 2,500 term newborns (Fawaz et al. 2017). This particular state has consistent pathological characteristics, which serve as an indicator of hepatobiliary dysfunction. Elevated concentrations of conjugated bilirubin in neonates may be an indication of biliary atresia (Mitra & Rennie 2017). Early detection of biliary atresia by primary healthcare providers is a significant factor in ensuring the most effective treatment and prognosis. This study aimed to examine the knowledge of primary healthcare providers regarding prolonged jaundice as an early indicator of biliary atresia.

MATERIALS AND METHODS

This study was carried out using a cross-sectional research design at some primary health facilities. This study assessed the variations in the diagnosis and management of infants with jaundice by using a self-administered questionnaire. The questionnaire comprised nine items regarding neonatal jaundice, prolonged jaundice, and biliary atresia (Setyoboedi et al. 2022b). The inclusion criteria encompassed health workers (n=271) who provided services to patients in primary health facilities. However, if they did not provide direct patient care, they were deemed ineligible.

A self-administered questionnaire was created by referencing our prior research. The survey was utilized to assess the respondents' understanding of prolonged jaundice as an early indicator of biliary atresia (Setyoboedi et al. 2022b). Descriptive data were processed and presented in the form of tables consisting of frequencies and percentages. The data analysis was performed using the descriptive analysis technique with the assistance of IBM SPSS

Statistics for Windows, version 21.0 (IBM Corp., Armonk, N.Y., USA). The conduct of this study fully adhered to the Declaration of Helsinki of the World Medical Association. Additionally, approval for this study was obtained from the Sidoarjo District Health Office, Indonesia, and consent was obtained from all of the primary health care facilities. The ethical clearance for this study was issued by the Health Research Ethics Committee of the Faculty of Medicine, Universitas Airlangga, Surabaya, Indonesia (No. 60/EC/KEPK/FKUA/2022, dated 7/3/2022).

RESULTS

There were 271 respondents who participated in this study, with 66% of them aged 30–50 years. The majority of the respondents in this study were midwives (63.8%). A total of 79% of the respondents had been working in a primary health facility for more than five years. Table 1 presents the detailed characteristics of the respondents.

Table 1. Basic characteristics of the participants (n=271).

	n	%
Sex		
Female	253	93.4
Male	18	6.6
Age (y.o.)		
20–29	34	12.5
30–39	99	36.5
40–49	80	29.5
50–59	54	19.9
>60	4	1.5
Occupation		
Midwife	173	63.8
Nurse	66	24.4
Medical doctor	23	8.5
Others	9	3.3
Working duration		
<3 years	25	9.2
3–5 years	32	11.8
>5 years	214	79.0

There were 40.2% of respondents who answered that infants suffering from jaundice, accompanied by pale stools and dark urine, might be attributed to physiological jaundice. However, 57.9% of the respondents disagreed with this statement. As many as 184 (67.9%) respondents believed that infants who have been suffering from jaundice since birth may not necessarily recover on their own. Regarding infants with prolonged jaundice, a total of 68 (25.1%) respondents indicated that no further examination is necessary. Instead, they suggested that these infants just need to be exposed to sunlight (Table 2).

Table 2. Analysis of the self-administered questionnaire.

Questionnaire items	Answers (n, %)
Infants suffering from jaundice accompanied by pale stools and dark urine are attributed to physiological jaundice.	True (109, 40.2%) False (157, 57.9%) No answer (5, 1.8%)
Infants who have been suffering from jaundice since birth will always improve on their own.	True (76, 28.0%) False (184, 67.9%) No answer (11, 4.1%)
Infants with prolonged jaundice do not necessitate further examination but rather require sunlight exposure.	True (68, 25.1%) False (200, 73.8%) No answer (3, 1.1%)
How long is the duration of jaundice that is defined as prolonged jaundice?	>1 week (10, 3.7%) >2 weeks (209, 77.1%) >3 weeks (16, 5.9%) >4 weeks (30, 11.1%) No answer (6, 2.2%)
What conditions need to be ruled out first in cases of prolonged jaundice?	Biliary atresia (137, 50.6%) Breastfeeding jaundice (86, 31.7%) Breast milk jaundice (87, 32.1%) Bile acid synthesis disorders (67, 24.7%) Kernicterus (50, 18.5%) Obstructive gallstones (37, 13.7%) Choledochal cyst (34, 12.5%)
What examination is required for prolonged jaundice?	Bilirubin test (250, 92.3%) Abdominal ultrasound (62, 22.9%) Scintigraphy (82, 30.3%)
What is the definition of cholestasis?	A serum direct bilirubin levels of >1 mg/dL (101, 37.3%) A serum direct bilirubin level of >20% of the total bilirubin (163, 60.1%)
When is the optimal time to perform Kasai surgery for biliary atresia?	Within 30 days of birth (42, 15.5%) Within 45 days of birth (8, 3%) Within 60 days of birth (119, 43.9%) Within 90 days of birth (42, 15.5%) No answer (60, 22.1%)
What is the frequency of your diagnosis of prolonged jaundice in infants?	Never (106, 39.1%) Seldom (113, 41.7%) Sometimes (44, 16.2%) Often (8, 3%)

When asked about the definition of prolonged jaundice, 209 (77.1%) respondents stated that it refers to a condition that has persisted for more than two weeks. Conversely, 16 (5.9%) respondents indicated that it lasts for more than three weeks, whereas 30 (11.1%) respondents answered that it lasts for over four weeks. A total of 137 (50.6%) respondents suggested that biliary atresia is a disease that must be ruled out first in cases of prolonged jaundice. However, 86 (31.7%) and 87 (32.1%) respondents mentioned that breastfeeding jaundice and breast milk jaundice, respectively, must be prioritized for ruling out in cases of prolonged jaundice in infants (Table 2).

The majority of the respondents, accounting for 250 respondents (92.3%), understood the importance of examining serum bilirubin levels in cases of prolonged jaundice in infants. However, when

questioned about the definition of cholestasis, as many as 101 (37.3%) respondents indicated that cholestasis is diagnosed when the serum direct bilirubin level exceeds 1 mg/dL. Meanwhile, 163 (60.1%) respondents stated that cholestasis can be determined when the serum direct bilirubin level is over 20% of the total bilirubin. Almost half of the respondents (43.9%) responded that the optimal time to perform Kasai surgery for biliary atresia is during the first 60 days after birth. A total of 113 (41.7%) respondents reported that they seldom diagnose prolonged jaundice.

DISCUSSION

Biliary atresia is a relatively uncommon condition that has emerged as a prominent reason for liver transplantation in the pediatric population. This prominence can be attributed to the absence of any

efficacious medicinal interventions capable of impeding the advancement of biliary atresia up until the present (Antala & Taylor 2022). The present understanding of the causes of biliary atresia encompasses various etiological pathways, including developmental, genetic, immunological, and viral factors. Nevertheless, the precise explanation of the underlying mechanism by which cholestasis culminates in fibrosis remains elusive to date. The issue of delays in referring cases of biliary atresia persists globally, affecting several locations such as Asian countries, Saudi Arabia, and Indonesia (Al-Hussaini et al. 2022, Davenport et al. 2023).

Biliary atresia is widely recognized as the primary etiology of newborn cholestasis. It is recommended that infants displaying jaundice persisting beyond the second week of life, also known as prolonged jaundice, undergo testing for both conjugated and unconjugated bilirubin levels. It is recommended to perform universal screening for neonates with elevated levels of direct bilirubin and to provide appropriate monitoring for these neonates (Al-Hussaini et al. 2022).

According Asai et al. (2015), biliary atresia has a variable occurrence depending on the geographical location, with reported rates ranging from 1 in 12,000 to 1 in 19,500 live births. The estimated occurrence of biliary atresia in the state of Texas, USA, is 0.65 cases per 10,000 live births. Meanwhile, the prevalence of biliary atresia in the USA is reported to be 4.47 per 100,000 individuals. Certain demographic factors have been associated with an increased chance of developing this condition. These factors include being female (relative risk [RR]=1.43, 95% confidence interval [CI]=1.27–1.62), residing in the Asia/Pacific region (RR=1.89, 95% CI=1.44–2.47), and being of black ethnicity (RR=1.30, 95% CI=1.06–1.58). There has been a notable rise in the prevalence of biliary atresia, with an average annual increase of 7.9% observed between the years 1997 and 2012 ($p < 0.001$) (Hopkins et al. 2017). A comprehensive examination was conducted by Cavallo et al. (2022) to investigate the correlation between biliary atresia and fetal development. The study found a positive correlation between biliary atresia and female sex (adjusted prevalence ratio [PR]=1.68, 95% CI=1.33–2.12), as well as delivery before 32–37 weeks of gestation (adjusted PR=1.64, 95% CI=1.18–2.29) and delivery before 32 weeks of gestation (adjusted PR=3.85, 95% CI=2.38–6.22). Additionally, a significant association was observed between biliary atresia and maternal diabetes (adjusted PR=2.34, 95% CI=1.57–3.48), with a stronger association reported for pregestational diabetes compared to gestational diabetes.

The prognosis for infants diagnosed with biliary

atresia is typically less than ideal, even in the USA. A delayed diagnosis has been identified as a contributing reason for increased healthcare expenditures due to the subsequent requirement for further medical interventions (Townsend et al. 2018). The optimal prognosis for individuals with biliary atresia remains closely associated with the timely detection and prompt implementation of surgical intervention. Hence, the primary approach for managing biliary atresia is to identify the condition at an early stage. This is particularly crucial in Indonesia, where liver transplantation resources are scarce. Nevertheless, the diagnosis of biliary atresia in neonates poses significant challenges. The initial presentation of biliary atresia typically lacks symptoms, and the infants appear to be in good health based on clinical observations. The primary manifestation that typically presents is frequently limited to jaundice, which has clinical resemblance to physiological jaundice. Physiological jaundice is the predominant etiology of newborn jaundice. Approximately 20–30% of infants who are exclusively breastfed continue to exhibit symptoms of jaundice at the age of one month (Maisels et al. 2014).

According to the findings of this survey, a significant proportion of respondents (73.8%) expressed the belief that newborns experiencing prolonged jaundice should undergo serum bilirubin level assessments. Conversely, a notable percentage (40.2%) of respondents indicated that pale feces and black urine in yellow infants were indicative of physiological jaundice. In instances of chronic jaundice in neonates, a significant proportion of respondents (25.1%, $n=68$) indicated that further diagnostic evaluations were unnecessary and that exposure to sunshine alone sufficed as a potential remedy. The findings suggest that there is a need for comprehensive enhancement of the primary health care providers' understanding of cholestasis. Conversely, Fawaz et al. (2017) recommend that infants who exhibit prolonged jaundice should be assessed for the presence of cholestasis. This assessment should include a measurement of direct serum bilirubin levels. The consideration of elevated levels of serum direct bilirubin is crucial. This is because infants diagnosed with biliary atresia exhibit heightened levels of direct bilirubin in their serum shortly following delivery.

According to Hodgson et al. (2018), the National Institute for Health and Care Excellence (NICE) recommendations suggest that infants with conjugated bilirubin levels exceeding 25 $\mu\text{mol/L}$ should be examined for potential liver illness. Additionally, the North American Society for Pediatric Gastroenterology, Hepatology, and Nutrition (NASPGHAN), in collaboration with the European Society for Paediatric Gastroenterology,

Hepatology, and Nutrition (ESPGHAN), recommends that a serum direct bilirubin level exceeding 1.0 mg/dL (17.2 μ mol/L) should prompt additional investigation and referral to a specialist in pediatric hepatology (Fawaz et al. 2017). The majority of the respondents (92.3%) in this study possessed prior knowledge on the importance of conducting serum bilirubin tests in newborns presenting with persistent jaundice. Nevertheless, when queried regarding the precise characterization of cholestasis, a significant proportion of the respondents, specifically 101 individuals (37.3%), stated that cholestasis is defined by a serum direct bilirubin level exceeding 1 mg/dL. Additionally, 163 respondents, representing 60.1% of the sample, identified cholestasis as a condition characterized by a serum direct bilirubin level surpassing 20% of the total bilirubin concentration.

Conjugated hyperbilirubinemia is a rare but medically significant condition. The findings of prior research indicated the presence of hepatobiliary dysfunction, as reported by Fawaz et al. (2017). Cholestasis refers to a condition characterized by a decrease in the production or movement of bile, resulting in the accumulation of biliary chemicals within the liver. The primary etiologies of cholestasis include biliary atresia (25–40%), monogenic diseases (25%), and multifactorial reasons such as cholestasis due to parenteral nutrition (Fawaz et al. 2017). A study by Dehghani et al. (2015) involving 122 infants diagnosed with cholestasis showed that jaundice was observed at a mean age of 15.6 \pm 16.1 days. The study further revealed that the most common causes of cholestasis were biliary atresia (30, 24.6%), idiopathic neonatal hepatitis (30, 24.6%), and insufficiency of bile ducts (16, 10.3%). In a separate study conducted by Isa & Irshad (2023) in Bahrain, it was shown that 86.4% of biliary atresia patients had clay-colored stools. Furthermore, 81.8% of the biliary atresia patients experienced hepatomegaly.

In this study, 137 (50.6%) respondents stated that biliary atresia is a disease that must be ruled out first in cases of prolonged jaundice. Nonetheless, a notable proportion of respondents, specifically 86 individuals (31.7%) and 87 individuals (32.1%), emphasized the significance of considering breastfeeding jaundice and breast milk jaundice as potential factors to be eliminated when assessing persistent jaundice in infants. Early diagnosis of biliary atresia is of paramount significance since it necessitates prompt specialist evaluation and surgical intervention, both of which significantly impact the prognosis. It is essential to equip primary health care providers with knowledge about the necessity of ruling out biliary atresia in cases of persistent jaundice. This measure is particularly crucial in primary health facilities, which serve as

the initial and principal points of access for health services within the community (Isa & Irshad 2023).

Lin (2015) reported that there was a decreasing trend in the mean age at which biliary atresia was diagnosed in Taiwan. However, this was not evenly distributed throughout the world, including Indonesia. In a previous study, it was found that the median age at which biliary atresia is diagnosed is 60 days. This diagnosis at such an early age is correlated with poor outcomes, such as mortality or the need for a liver transplant (Fawaz et al., 2017). In another study conducted by Apfeld et al. (2021), it was discovered that out of 470 infants with biliary atresia who underwent the Kasai procedure at an average age of 57 days, 45.1% of them had the treatment performed when they were 60 days of age or older. Biliary atresia can lead to liver cirrhosis and ultimately result in mortality if left untreated.

A study conducted by Holdar et al. (2019) in Saudi Arabia revealed that the mean age at which patients with advanced biliary atresia were referred to pediatric gastroenterology was 65 days. In addition, the Kasai procedure was performed in 12 cases, with a mean age of 73 days and a success rate of around 33%. Another study by Khayat et al. (2021) was conducted on 23 biliary atresia patients who underwent the Kasai procedure at the age of 77 \pm 34 days. The study showed that 13 (56%) patients survived with their native livers. Almost half of the respondents (43.9%) in this study indicated that the optimal time for Kasai surgery in cases of biliary atresia is within 60 days after birth. In contrast, 60 (22.1%) respondents were unsure about the optimal time for surgery in cases of biliary atresia. Survival chances with native livers will decline as the age during surgery grows from less than 45 days to 90 days. A study conducted by Qisthi et al. (2020) in Yogyakarta, Indonesia, revealed that the mean age of biliary atresia patients was 102.5 days during the Kasai procedure. The study also found that 33 (66%) patients had liver cirrhosis, while 17 (34%) patients did not have the same condition.

In a retrospective cohort study conducted by Wang et al. (2019) in China, it was shown that out of 139 patients with biliary atresia, the estimated 5-year survival rate of the native liver was 58.0%. However, there was a significant risk of death and the need for liver transplantation after undergoing the Kasai surgery. A systematic review by Jimenez-Rivera et al. (2013) suggests that the ten-year survival rate for individuals with biliary atresia ranges from 66.7% to 89%. The survival rate of the native liver in individuals with biliary atresia who had the Kasai procedure varies from 20.3% to 75.8% within one to three years and from 24% to 52.8% at ten years. The age at which an earlier Kasai procedure was performed was found to be a

predictor of improved survival of the patients' native livers.

In a study conducted by [Liu et al. \(2017\)](#) in Vietnam, 287 children with biliary atresia were involved. Out of these participants, 149 (52%) were treated without the Kasai procedure, while 138 (48%) were treated with the Kasai procedure. The Kasai procedure was not performed on children diagnosed with biliary atresia within specific age ranges: less than 2 months (31%), 2 to less than 3 months (35%), 3 to less than 4 months (15%), 4 to less than 6 months (10%), and 6 months or older (9%). The survival rates of children with biliary atresia who did not undergo the Kasai procedure were 52% and 28% at 1 and 2 years, respectively. In comparison, children with biliary atresia who underwent the Kasai procedure had survival rates of 84% and 71% at 1 and 2 years, respectively. A study examining the outcomes of infants with biliary atresia found that those who receive liver transplantation do not experience a significantly improved quality of life compared to those who manage to survive with their original livers ([Lee et al. 2016](#)).

[Harpavat et al. \(2018\)](#) conducted a study on infants with biliary atresia, involving a total of 65 participants. The study was retrospective and assessed two specific time periods: from birth until specialist referral, and from specialist referral to treatment. The study revealed a correlation between the duration from birth to the first treatment and an individual's race or ethnicity. Conversely, the duration it took to refer infants to a specialist for treatment was found to be influenced by the age at which they were referred. Infants referred after 30, 45, or 60 days of life experienced a reduced waiting period ($p=0.001$). The reason for the delay in referral might be associated with the presence of normal aminotransferase levels.

In a study conducted by [Isa & Irshad \(2023\)](#) on biliary atresia patients, it was found that a delay in diagnosing the condition might lead to significant morbidity and mortality, despite the effectiveness of the treatment provided. The study focused on patients with an average age of 56 days at the time of diagnosis. According to the study, the survival rate of individuals with native livers was relatively low, at 53.8%. Therefore, it is required to promptly report infants with prolonged jaundice through screening, and the referral protocol must be standardized and widely spread in primary health facilities.

A study by [Gu & Matsui \(2017\)](#) demonstrated that the utilization of a stool color card as a means for early diagnosis is linked with increased long-term survival of the native livers. Furthermore, the implementation of a stool color card has been

associated with a significant decrease in both hospitalization and mortality rates among individuals diagnosed with biliary atresia in Taiwan ([Lee et al. 2016](#)). Stool color cards have been used in population-based screening programs in many countries, such as Taiwan, China, and Japan, to facilitate the detection of biliary atresia at an early stage. [Goodhue et al. \(2017\)](#) conducted a study in North America to assess the cost-effectiveness of screening for biliary atresia using stool color cards. The research findings indicated a notable decrease in the prevalence of biliary atresia, accompanied by an improvement in survival rates and a potential reduction in the financial burden associated with treatment expenses. The biliary atresia screening program for newborns in the United States was developed by the American Academy of Pediatrics (AAP).

A case-control study by [Gu & Matsui \(2017\)](#) was performed in Japan to examine infants with biliary atresia who underwent the Kasai procedure and were screened either with or without the use of stool color cards. The research findings revealed a significant difference in the average age at which the Kasai procedure was performed between individuals who used stool color cards and those who did not. Specifically, the mean ages for users and non-users were 59.7 and 68.2 days, respectively, with a statistically significant value of $p<0.05$. The study further showed that patients who used stool color cards had a survival rate of 48.5% after 12.5 years, while non-users exhibited a survival rate of 36.6%. The observed difference was shown to be statistically significant ($p<0.05$).

An investigation focusing on biliary atresia was carried out by [Gu et al. \(2015\)](#) in Japan. From 1994 to 2011, stool color cards were distributed to pregnant women residing in Tochigi Prefecture. Upon postpartum, the women submitted a properly filled-out stool color card to a physician and received a specimen container to collect a sample for further biliary atresia analysis. The study encompassed a cohort of 313,230 live births, among whom 34 infants were identified as having biliary atresia. The study determined that the stool color card screening had a sensitivity of 76.5% (95% CI=62.2–90.7) and a specificity of 99.9% (95% CI=99.9–100.0) during the 1-month assessment. The findings indicated that the review color cards demonstrated favorable sensitivity and specificity in facilitating early detection of biliary atresia cases.

As documented by [Lien et al. \(2011\)](#), the implementation of a stool color card screening program in Taiwan resulted in a notable rise in the number of Kasai procedures performed during early childhood as well as an improvement in the rate of jaundice-free outcomes observed three months

following the surgery. Hence, it is imperative to employ both methods of biliary atresia detection and serum direct bilirubin analysis when dealing with cases of persistent jaundice. A serum direct bilirubin level equal to or more than 2 mg/dL is indicative of a hepatobiliary disease. Laboratory-based screening of the serum bilirubin has the potential to discover suspected cases of biliary atresia early, leading to an increase in the number of newborns who receive early surgical intervention. Matsui (2017) suggests that primary healthcare providers have poor understanding regarding prolonged jaundice as an early indicator of biliary atresia. This highlights the need for more standardized screening guidance to improve the early detection of biliary atresia in newborns. In our earlier study conducted in Surabaya, we designed a stool color card with the aim of facilitating the assessment of stool colors in newborns suffering from chronic jaundice (Setyoboedi et al. 2022a). However, there is still a need for the widespread dissemination of knowledge regarding the timely identification of infants with cholestasis and the utilization of stool color cards among health professionals working in primary healthcare settings. Health education has proven to be an effective approach to enhancing awareness and understanding of biliary atresia, hence aiding in the early identification of this condition in Indonesia (Setyoboedi et al. 2022b). It is noteworthy to pay attention to the trends indicating an increase in the age at which biliary atresia is diagnosed to improve patient outcomes.

Strength and limitations

This study offers an elucidation of primary healthcare providers' level of knowledge regarding prolonged jaundice as an early sign of biliary atresia. The information was obtained from a survey that included a large number of participants. However, this research was exclusively conducted in an urban area, implying that different results could be observed in underdeveloped areas.

CONCLUSION

The lack of knowledge among primary healthcare providers regarding the early detection of biliary atresia continues to be a problem. There is still a need to raise education in primary health facilities concerning prolonged jaundice as an early sign of biliary atresia. Further studies with a larger sample size are necessary to evaluate the level of knowledge in identifying infants with biliary atresia at an early stage in Indonesia.

Acknowledgment

The authors would like to express gratitude to the

Dean of the Faculty of Medicine, Universitas Airlangga, Surabaya, Indonesia.

Conflict of interest

None.

Ethical consideration

The Health Research Ethics Committee of the Faculty of Medicine, Universitas Airlangga, Surabaya, Indonesia, issued the ethical approval for this study, with reference No. 60/EC/KEPK/FKUA/2022 dated 7/3/2022.

Funding disclosure

None.

Author contribution

RAP and BS contributed to the conception and design, analysis and interpretation of the data, drafting of the article, critical revision of the article for important intellectual content, final approval of the article, provision of study materials, statistical expertise, funding, collection and assembly of the data, as well as provision of administrative, technical, and logistic support. NNMN, MM, MTU, and SA contributed to the conception and design, analysis and interpretation of the data, drafting of the article, critical revision of the article for important intellectual content, final approval of the article, provision of study materials, statistical expertise, collection and assembly of the data, as well as administrative, technical, and logistic support.

REFERENCES

- Al-Hussaini A, Abanemai M, Alhebbi H, et al (2022). The epidemiology and outcome of biliary atresia: Saudi Arabian national study (2000–2018). *Frontiers in Pediatrics*. doi: 10.3389/fped.2022.921948.
- Antala S, Taylor SA (2022). Biliary atresia in children. *Clinics in Liver Disease* 26, 341–354. doi: 10.1016/j.cld.2022.03.001
- Apfeld JC, Cooper JN, Sebastião Y V, et al (2021). Variability in age at Kasai portoenterostomy for biliary atresia across US children's hospitals. *Journal of Pediatric Surgery* 56, 1196–1202. doi: 10.1016/j.jpedsurg.2021.02.027.
- Asai A, Miethke A, Bezerra JA (2015). Pathogenesis of biliary atresia: defining biology to understand clinical phenotypes. *Nature Reviews Gastroenterology & Hepatology* 12, 342–352. doi: 10.1038/nrgastro.2015.74.
- Cavallo L, Kovar EM, Aqul A, et al (2022). The epidemiology of biliary atresia: Exploring the role

[ni.v9i4.813](#).

Tan HS, Balasubramaniam IS, HSS AS, et al (2019). Impact of a standardized protocol for the management of prolonged neonatal jaundice in a regional setting: an interventional quasi-experimental study. *BMC Pediatr* 19, 174. doi: [10.1186/s12887-019-1550-3](https://doi.org/10.1186/s12887-019-1550-3).

Townsend MR, Jaber A, Abi Nader H, et al (2018). Factors associated with timing and adverse outcomes in patients with biliary atresia

undergoing kasai hepatoportoenterostomy. *The Journal of Pediatrics* 199, 237-242. doi: [10.1016/j.jpeds.2018.04.001](https://doi.org/10.1016/j.jpeds.2018.04.001).

Wang Z, Chen Y, Peng C, et al (2019). Five-year native liver survival analysis in biliary atresia from a single large Chinese center: The death/liver transplantation hazard change and the importance of rapid early clearance of jaundice. *Journal of Pediatric Surgery* 54, 1680–1685. doi: [10.1016/j.jpedsurg.2018.09.025](https://doi.org/10.1016/j.jpedsurg.2018.09.025).

