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Neonatal Gastrointestinal Emergencies

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Fatima Safira Alatas

## Original Article

# Duration and Sociodemographic Factors Associated with Exclusive Breastfeeding Among Mothers in Urban and Semi-Rural Areas of Libreville and Lambaréné in Gabon

Steeve Minto'o<sup>1</sup>, Fifi Claire Loembe<sup>1</sup>, Midili Thècle Larissa<sup>1</sup>, Mireille Mensan Pemba<sup>1</sup>, Koumba Maniaga Raïssa<sup>1</sup>, Mylène Mimbila-Mayi<sup>1</sup>, Yolande Nzame<sup>1</sup>, Essomo Murielle<sup>1</sup>, Eliane Kuissi-Kamgaing<sup>1</sup>, Jean Koko<sup>1</sup>, Simon Ategbo<sup>1</sup>

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**Abstract:**

**Background:** Exclusive breastfeeding from birth to six months of age has an unparalleled benefit on a child's growth and development. This survey aimed to assess the practice of exclusive breastfeeding (EBF) in Libreville and Lambaréné.

**Method:** This is a prospective study, including mother-child pairs of infants taken to vaccination centers in Libreville (urban area) and Lambaréné (semi-rural area). The data collected included sociodemographic aspects of families, the children's characteristics, and the duration of EBF in months.

**Result:** We included 552 mother-child pairs, 58% of whom were recruited (n = 320) in Libreville. The average age of mothers was  $26.5 \pm 6.4$  years, while fathers were  $33.3 \pm 7.8$  years. The mean age of children was  $4.2 \pm 3.1$  months. Male participants were accounted for 52.2% (n = 288), while females 47.8% (n = 264). The mean duration of exclusive breastfeeding in the whole was 0.9 months. Six-month EBF was generally performed at 9.2% (n = 51, 95% CI = 7.1%-11.9%). The factors influencing the EBF were father's level of education (all classes combined) (p = 0.025), marital status (p = 0.011), and residential area (OR = 3.40, p < 0.001).

**Conclusion:** The duration of exclusive breastfeeding in the two studied is lower than the WHO recommendations. The associated factors found are areas of work to be explored to encourage this important practice for our infants.

**Keywords:** associated factors, children, exclusive breastfeeding, gabon, prevalence



## Introduction

Exclusive breastfeeding (EBF) is defined as giving only breast milk to infants. When performed during the first 6 months of life, EBF is a crucial aspect of optimal breastfeeding practices to prevent infant morbidity and mortality. It is important to remember that EBF for six months reduces gastrointestinal infections in young children and helps the mother lose weight while preventing a new pregnancy. It also has long-term impacts on allergic diseases, growth, obesity, cognitive ability, and behaviour. By encouraging this practice, the World Health Organization (WHO) estimates that the lives of one million children under-five could be saved annually in developing countries. Despite its efficacy and cost-effectiveness, EBF during the first six months of an infant's life is insufficiently practiced in several parts of the world.<sup>1</sup> This is particularly evident in sub-Saharan African (SSA) countries, where a significant number of infants (55%) and under-five mortality (75%) are attributed to inadequate breastfeeding practices.<sup>2</sup>

In 2013, the WHO and the United Nations Children's Fund (UNICEF) set a global nutrition target of achieving an EBF prevalence rate of  $\geq 70\%$  by 2030.<sup>3</sup> The rate of EBF in SSA is estimated to be 35%, compared to 39% in other developing regions. Only 18 of the 49 African countries are on track to meet the World Health Organization's (WHO) 2025 global nutrition targets, aiming to increase this rate to 50%.<sup>4</sup> However, UNICEF's efforts to raise awareness among young mothers about the real benefits of EBF on child development are observable. Exclusive breastfeeding rates worldwide increased from 36% in 2011 to 43% in 2014. Niger, for example, documented a surge in the exclusive breastfeeding rate from 4% in 2011 to nearly 23% in 2014, as did Sierra Leone, which rose from 11% to 32% over the same period.<sup>4</sup>

In the same period, Gabon's 2012 Demographic and Health Survey (DHS) noted an EBF rate of 6%, with an average EBF duration of 1.1 months.<sup>5</sup> Therefore, our study aimed to investigate whether UNICEF's all-out advocacy yielded similar results in our country. The purpose of our study was to participate in improving EBF practices in our country by assessing the duration of EBF in Gabon and identifying the sociodemographic factors associated with its inadequacy.

## Method

This prospective, longitudinal and analytical study was conducted in Libreville (urban) and Lambaréné (semi-rural) from Oct 1 2019 to Mar 31 2020. It involved infants aged 0 to 11 months and their mothers.

We included infants aged 0 to 11 months living in Libreville and Lambaréné and their mothers, both in apparent good health, seen during weighing or vaccination sessions in health centres in both localities. All the children included were recruited voluntarily from the families. They had to have a health record.

We did not include infants over 11 months of age, those whose parents had refused to participate in the study, or those who were only passing through the city. We also excluded mother-infant couples who were following a protocol for the prevention of mother-to-child transmission of HIV and those whose parents had decided to leave the study project.

The calculation of the sample size using Statcalc from Epi Info 7.2 gave a minimum number of subjects of 420, with an expected frequency of 70% (which corresponds to the WHO and UNICEF targets for 2030 in EBFs), a margin of error of 5%, with a 95% confidence interval; a mitigating factor of our study type of 1.3. Four hundred twenty subjects for 210 per cluster (Libreville and Lambaréné) were recruited. We then accepted a weighting adjustment, bringing the representation of Libreville cluster to nearly 60% and Lambaréné cluster to 40% to reflect the demographic distribution of Gabon.<sup>6,7</sup>

The active phase consisted of collecting data concerning the children: date of birth, sex, rank of the child among the siblings, number of children in the house, and practice of EBF at the time of the interview. When EBF was underway, the mother entered a database of subjects to be contacted again one week after the interview and then every two weeks to find out when she had stopped the EBF. The variables collected for parents were age in years, education and occupation of the mother and father.

Quantitative variables were expressed as mean or median. Qualitative variables were expressed in terms of frequency. The frequency of EBF was expressed in 95% confidence interval. The univariate analysis assessed the association between a sociodemographic characteristic and EBF  $\geq 6$  months by calculating the odd ratio with a 95% confidence interval according to the Miettinen method. This analysis made extracting the variables of interest possible for the multivariate analysis performed in logistic regression. The p-value selected as significant was  $<0.05$  for a two-tail Chi-square test.

In the absence of a National Ethics Committee during the investigation, the Gabon Ministry of Health approved the study before it started. Parents gave their written consent by signing the informed consent on the day of inclusion. Data confidentiality was fully respected.

Result

We included 552 mother-child couples, 320 (58%) in Libreville and 232 (42%) in Lambaréné. **Table 1** illustrated the characteristics of children and families.

**Table 1.** Characteristics of the families

	Total (n=552)		Libreville (n=320)		Lambaréné (n=232)	
Mothers' age (mean ± SD)	26.5 ± 6.4		27.2 ± 6.1		25.6 ± 6.8	
Fathers' age (mean ± SD)	33.4 ± 7.9		33.8 ± 7.8		32.8 ± 7.8	
Mother's educational level						
Primary	125	22.6%	63	19.7%	62	26.7%
Secondary	331	60.0%	170	53.1%	161	69.4%
University	96	17.4%	87	27.2%	9	3.9%
Father's educational level						
Primary	59	10.7%	18	5.6%	43	18.5%
Secondary	341	61.8%	173	54.1%	167	72.0%
University	152	27.5%	129	40.3%	22	9.5%
Household's monthly income						
< 250 USD	176	31.9%	59	18.4%	117	50.4%
USD 250-500	216	39.1%	137	42.8%	79	34.1%
USD 501-1000	137	24.8%	107	33.4%	30	12.9%
USD 1001-1500	16	2.9%	11	3.5%	5	2.2%
> 1500 USD	7	1.3%	6	1.9%	1	0.4%
Mother's occupational status						
Not Working	351	64%	179	55.9%	172	74.1%
Actively Working	133	24%	94	29.4%	39	16.8%
Student	68	12%	47	14.7%	21	9.1%

**Children's characteristics**

Males accounted for 52.2% (n = 288) and females 47.8% (n = 264). The mean age of the children was 4.2 ± 3.1 months. The median rank of siblings was 2.

**Parents' age and education level.**

Overall, the mother's mean age was 26.5±6.4 years, with mothers from Libreville averaging 27.2±6.1 years and mothers from Lambaréné averaging 25.6±6.8 years. The

majority of mothers had a secondary education (60.0%), followed by primary (22.6%) and university (17.4%). In Libreville, 53.1% of mothers also had a secondary educational level, while 19.7% had a primary education, and 27.2% went to university. Similarly, mothers from Lambaréné mainly had secondary education (69.4%), while 26.7% had a primary educational level and 3.9% had university education level.

The overall mean age for fathers was  $33.4 \pm 7.9$  years, with the fathers from Libreville averaging  $33.8 \pm 7.8$  years and those from Lambaréné averaging  $32.8 \pm 7.8$  years. Fathers mainly had secondary education (61.8%), with 10.7% having primary education and 27.5% receiving a university education. Fathers from Libreville also primarily graduated from secondary education (54.1%), followed by primary education (5.6%) and university (40.3%). In Lambaréné, 72.0% of fathers had a secondary education, 18.5% had a primary education, and 9.5% went to university.

### Characteristics of the households

Parents lived as a couple in 67.2% of cases ( $n = 371$ ). Mothers had a median number of pregnancies of 3 with an interquartile range (EI) of 2. In general, 31.9% of households earned less than 250 United States of America Dollars (USD) per month, 39.1% earned between 250 and 500 USD, 24.8% earned between 501-1000 USD, 2.9% earned between 1001-1500 USD, and 1.3% earned more than 1500 USD. In Libreville, 18.4% of households earned less than 250 USD per month, 42.8% earned between 250 and 500 USD, 33.4% earned between 501-1000 USD, 3.5% earned between 1001-1500 USD, and 1.9% earned more than 1500 USD. In Lambaréné, 50.4% of households earned less than 250 USD per month, 34.1% earned between 250 and 500 USD, 12.9% earned between 501-1000 USD, 2.2% earned between 1001-1500 USD, and 0.4% earned above 1500 USD.

Generally, the majority of mothers were not working (64%), while 24% were actively working and 12% were students. In Libreville, the percentages were the same: 64% not working, 24% actively working and 12% students. In Lambaréné, 74.1% of mothers were not working, 16.8% were active, and 9.1% were students.

### Breastfeeding duration

The mean duration of exclusive breastfeeding in general was  $0.9 \pm 1.5$  months. The frequency of six-month EBF was 9.2% ( $n = 51$ ; 95% CI = 7.1 - 11.9). In Libreville, the six-month EBF frequency was 4.2% ( $n = 14$ ; 95% CI = 2.6 - 7.2), while in Lambaréné, the frequency was 15.9% ( $n = 37$ ; 95% CI = 11.5% - 21.3). The duration of exclusive breastfeeding differed significantly depending on the mother's level of education, the presence of father's occupation, marital status, and recruitment location (Table 2).

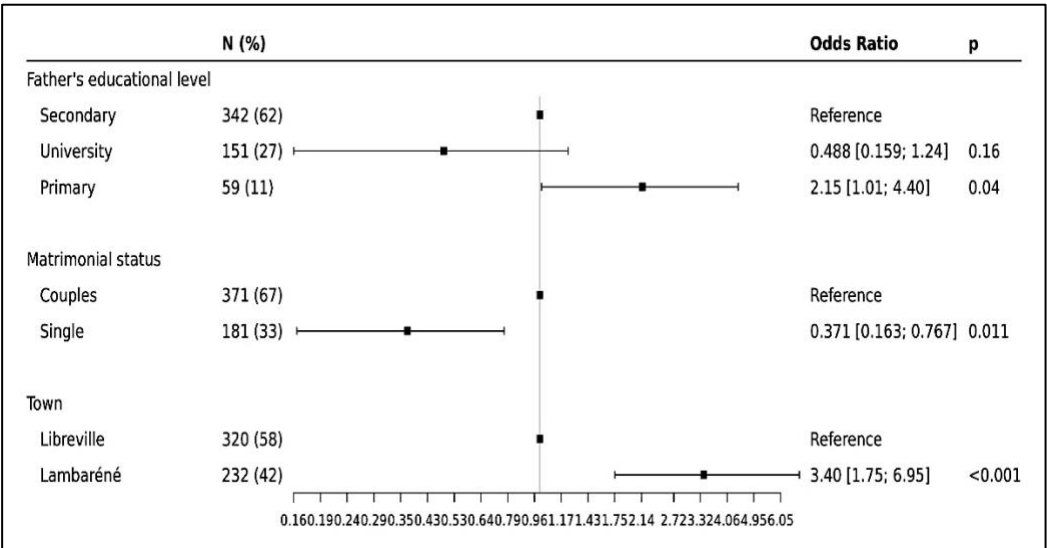
Table 2. Average Duration Exclusive of Breastfeeding

	Mean (SD)	n	p
<b>Mothers' Educational Level<sup>a</sup></b>			0.028
Primary	1.19 (1.90)	125	
Secondary	0.849 (1.48)	331	
University	0.646 (1.40)	96	
<b>Father's Educational Level<sup>a</sup></b>			0.094
Primary	1.24 (2.10)	59	
Secondary	0.909 (1.53)	341	
Upper	0.717 (1.43)	152	
<b>Mother's Occupational Status<sup>a</sup></b>			0.25
Not Working	0.883 (1.58)	351	
Actively Working	1.04 (1.72)	133	
Student	0.647 (1.26)	68	
<b>Father's Occupational Status<sup>b</sup></b>			<0.01
Not Working	0.846 (1.56)	460	
Actively Working	1.47 (1.87)	59	
Student	0.485 (0.755)	33	
<b>Household's Monthly Income<sup>b</sup></b>			0.15
USD 250-500	1.01 (1.71)	216	
< 250 USD	0.898 (1.53)	176	
USD 501 – 1000	0.715 (1.42)	137	
USD 1000 – 1500	1.12 (1.67)	16	
>1500 USD	0 (0)	7	
<b>Sex<sup>c</sup></b>			-0.69
Male	0.917 (1.60)	288	
Female	0.864 (1.56)	264	
<b>Marital status<sup>c</sup></b>			<0.01
Married	1.00 (1.69)	388	
Single	0.628 (1.23)	164	
<b>Town<sup>c</sup></b>			<0.01
Libreville	0.728 (1.49)	320	
Lambaréné	1.12 (1.67)	232	

<sup>a</sup> was analyzed using ANOVA test. <sup>b</sup> was analyzed using Kruskal-Wallis. <sup>c</sup> was analyzed using Welch. SD: standard deviation, n: number of population, p: p-value, USD: United States Dollar



Multivariate analysis showed that the EBF was significantly different according to the father's level of education (all classes combined) ( $p = 0.025$ ); the duration of the EBF was shorter for those in single marital status compared to couple ( $OR = 0.371$ ,  $p = 0.011$ ), and longer when families living in Lambaréné than in Libreville ( $OR = 3.40$ ,  $p < 0.001$ ) (**Figure 1**).



**Figure 1.** Forest plot of the logistic regression for factors impacting breastfeeding in Gabon

Discussion

The characteristics of the families can be superimposed on the results of Gabon's most recent demographic and health survey.<sup>6</sup> The type of study and collection sites ensure that our results represent breastfeeding practices and, more specifically, EBF in these two localities.

The duration of EBF in our study was 0.9 months, almost the same as that published in 2012, which was 1.1 months, but lower than the duration of the 2023 DHS, which was 2.3 months.<sup>5,6</sup> The difference between our result and the DHS is based on the sampling and study design chosen in both cases. The DHS 2023 selected 2/3 of interviewees from other provinces and 1/3 from Libreville and Port-Gentil. In comparison, Libreville and Port-Gentil represent 60% of the Gabonese population.

There is an over-representation of the country's interior, which has, therefore, clearly distorted the national average duration of EBF, as populations in the interior have a longer duration of breastfeeding. We respected this proportion in our survey. In addition, the DHS interviews were sometimes conducted long after the pregnancies,

while our study included mothers who are still actively engaged in childcare and immunization. Therefore, the subjects in our study were less prone to memory bias.

The frequency of EBF in our study was 9.2%. This frequency confirms the data that places Gabon in the category of countries with the lowest rates of EBFs globally, together with Chad at 2.2% but well below Rwanda at 87.7%.<sup>4</sup> The latest data for Gabon reported a frequency of 19% throughout the country.<sup>6</sup> Given the abovementioned biases, we also argue that this frequency is overestimated. This frequency found by the DHS III is close to and included in the confidence interval of the frequency we found in Lambaréné, a semi-rural town with 15.9%.

Our study suggests that the EBF in Gabon is stagnating. This view is reinforced by Bhattacharjee's study, which shows no change in the number of EBF and even breastfeeding in Gabon between 2000 and 2018, despite improvements in other public health markers supported by the WHO and UNICEF.<sup>6</sup>

In our study, women with a higher level of education had a shorter duration of EBF than those with a lower level of education. This finding is similar to Mathew AC et al. in Nepal and Nurokhmah et al. in Indonesia, who found that the median duration of EBF for mothers with a high level of education was lower than that for mothers with a lower level of education.<sup>7,8</sup> Conversely, studies by Pariya et al. in Calcutta, Lopez de Aberasturi et al. in Spain, and Economou et al. in Cyprus noted that a high level of mother's education was associated with a longer EBF.<sup>9-11</sup>

Women living with a partner have a longer breastfeeding duration than women living alone. This situation can be explained by a cultural fact in Gabon: the newly delivered woman ("moussonfi", lactating woman) who is married receives care from the women of her family and her in-laws so that she can devote herself only to the EBF and the care of her newborn. She is provided with everything she needs, including toiletries and meals; she is not allowed to go into a kitchen or do any household chores for two lunar months.<sup>12,13</sup> In its traditional form, this period was longer, but it has been adapted to the residential settings; it tends to be longer in rural than urban areas. The "moussonfi" period is abbreviated in urban areas, as the mother must take her child to a health centre for weighing and vaccinations. This could present challenges such as public transportation, queues, and potential scrutiny from strangers if the mother were to breastfeed in public.<sup>13-15</sup> Longer EBF durations were also found among women with partners at DHS 2023 and populations in Kolkata.<sup>6,9</sup> Family guidance and support were also identified as factors contributing to the success of EBF in the Mpumalanga region of South Africa.<sup>16</sup> This finding is not surprising compared to our survey results because the populations of Mpumalanga consist of Bantu people who share similar cultures to those in Gabon. Some factors regularly analysed in the

literature, such as the number of prenatal consultations and the existence of breastfeeding preparation sessions, could not be analysed in our study due to the absence of uniform pregnancy follow-up logs; some diaries had pregnancy follow-up pages mentioning these sessions, while others did not, or the parents did not have them during the interviews.

The lower father's educational attainment was associated with a longer duration of EBF. This factor may be related to the lack of employment, but it may also be influenced by a more traditionalist aspect of people with low levels of education.<sup>6,13</sup> The factor with the strongest association with a good duration of breastfeeding was living in the country's interior (Lambaréné). Living in the interland brings together all the cultural and societal ingredients that favour a longer practice of EBF as the cultural practice of "moussonfi" is more accessible in this area. Other surveys in India, Indonesia, and South Africa highlighted living in an urban area as a factor favouring EBF.<sup>8,9,16,17</sup> On closer reading of these results, the existence of medical structures encouraging mothers to breastfeed before and after birth also make a difference, as there were more "baby-friendly" structures in these cities.

## Conclusion

The results of our study are a reminder that while mother's milk is free food, EBF has a cost. Women in cities work more outside the home and breastfeed less than women in rural areas. This fact is undoubtedly linked to the professional obligations they must fulfil to not remain on the sidelines of the labour market. Indigenous cultural practices that foster EBF are diluted in the urban world and more robust in the rural world. State structures have compensated for these cultural losses by strengthening labour legislation to protect working women's EBFs. Gabon is, therefore, far from the targets set for 2030. The trend would be reversible if he could rely on his cultural achievements and the increase in the number of mother preparation sessions for the EBF.

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## Conflict of Interest

None declared

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## Original Article

# Bone Mineral Disorders in Children with Predialysis Chronic Kidney Disease Correlates with Short Stature

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Rania G, Puspitasari HA, Hidayati EL, Hafifah CN. Nutritional Status of Children with Chronic Kidney Disease in Stage 3-5 Predialysis: Identifying Influential Factors. *Arch Pediatr Gastr Hepatol Nutr*. 2024;3(1).12-23

**Abstract:**

**Background:** The increasing prevalence of malnutrition and growth impairment among children with CKD could impact the prognosis and the preferred intervention. Therefore, this study aimed to identify the nutritional status of children with CKD and its relating factors.

**Method:** A cross-sectional study was conducted in a tertiary, national-referral teaching hospital in Jakarta. Sociodemographic and laboratory data were obtained from medical records. Body weight and height of participants were measured using digital scale and stadiometer, respectively. Growth and nutritional status indicators such as BMI-for-age, weight-for-height, height for age, and weight for age were quantified and plotted using WHO Anthro and Anthroplus application.

**Result:** A total of 18 participants aged 3-17 years old with CKD stage 3 – 5 were included in this study. BMI measurements showed a z-score average of -1.02, while the mean z-score for height-for-age was of -2.71. Our study demonstrated a significant association between the height-for-age and mineral bone in children with CKD ( $p = 0.005$ ). However, we found no association between mineral bone disorder with other indicators of nutritional status. Furthermore, our study also found no significant relation between nutritional status and other influencing factors including the stage of CKD, duration of CKD, age, gender, primary etiological factor, hypertension, anemia, age, familial economic status, disease duration, and parental education level demonstrates no significant correlation ( $p > 0.05$ ).

**Conclusion:** Children with stage 3 – 5 CKD in the pre-dialysis phase are shown to be underweight and short statured but with normal nutritional status. Mineral bone disorder was revealed to be significantly associated with height-for-age in children with CKD.

**Keywords:** children, chronic kidney disease, nutritional status, pre-dialysis, influential factors

## Introduction

Chronic kidney disease (CKD) poses a major health concern, affecting an estimated 13.4% of the global population and influencing 18.5-58.3 per 1 million children worldwide.<sup>1</sup> Data upon the prevalence of pediatric CKD in Indonesia were still limited; however, an increased trend of the disease incidence was identified from several referral centers across the country.<sup>2</sup> The escalating number of CKD cases had raised concerns, as children with CKD face a higher risk of malnutrition, potentially leading to growth failure. The prevalence of malnutrition in children with CKD is approximately 20-45%. Furthermore, malnutrition not only impacts growth but also elevates the risk of morbidity and mortality in CKD patients.<sup>3</sup>

Nutrition plays a crucial role in preventing infections, hospitalizations, depression, weakness, and cardiovascular diseases in children with CKD.<sup>4</sup> However, the nutritional challenges in pediatric CKD cases extend beyond reduced intake and involve other factors such as decreased appetite, increased catabolism, hormonal imbalances, inflammation, and nutrient loss in dialysate fluid.<sup>3</sup> The accumulation of metabolic by-products due to the kidney's inability to clear it also plays a role in metabolic dysfunction among CKD patients.<sup>3</sup> In addition, the progression of the disease further determines the risk of malnutrition. Study in India had illustrated an increase in the incidence of malnutrition corresponding to the progression of CKD stages.<sup>5</sup> This concludes that nutritional imbalances and metabolic disturbance had happened among CKD patients, even from the pre-dialysis stage.

Management and renal replacement therapy also contribute to growth disturbances in CKD patients. Research in Nigeria indicated a 46.7% prevalence of malnutrition in pre-dialysis CKD patients, higher than those without kidney dysfunction (25.7%).<sup>6</sup> In Iran, a study reported malnutrition in 19 out of 42 pediatric CKD patients, with 4 cases occurred in the pre-dialysis phase, 5 cases were reported in dialysis phase (peritoneal and hemodialysis).<sup>7</sup> Studies have found that one of CKD therapies, hemodialysis, could induce increased CRP, protein muscle breakdown, and oxidative stress that could further affect growth and nutritional status.<sup>3</sup> However, studies have also linked chronic inflammation, characterized by increased TNF-alpha cytokines in CKD patients since the pre-dialysis phase, to bone mineral disorders affecting patients' stature.<sup>8</sup> Therefore, further study must be conducted to find the most ideal intervention to prevent worsening nutritional status.

Given the high prevalence of malnutrition in children with CKD, identifying factors influencing the nutritional status is crucial to prevent malnutrition and growth disturbances. Therefore, this study aims to investigate the nutritional status of pre-dialysis pediatric CKD patients and its relation to multiple factors. In the long run, the result of this study could be used to identify whether early diagnosis and intervention

from pre-dialysis stage can improve the nutritional outcomes among children with CKD.

## Methods

**Data collection** This is a cross-sectional study conducted in October 2022 at a tertiary, national-referral, teaching hospital in Jakarta, Indonesia. The study was approved by the local ethics committee with approval number of KET-915/UN2.F1/ETIK/PPM.00.02/2022. We included all pediatric patients aged 0-18 years in pre-dialysis phase of CKD (stage 3-5) for a minimum of three months prior the study. CKD patients who underwent kidney transplant and whose parents did not provide consent were excluded from the study. This study is a part of a larger study focusing on nutritional status of children with CKD, with the main difference of the participants which are divided into three according to the characteristics; patients who undergo predialysis, hemodialysis (Metasyah, et al)<sup>9</sup>, or continuous ambulatory peritoneal dialysis (Amirah, et al).<sup>10</sup>

**Calculation of growth and nutritional status indicators.** Anthropometric measurements were performed using a combined digital weight scale and stadiometer (SECA 730). Indicators of nutritional status, including weight for height, weight for age, height for age, and body mass index (BMI) for age were calculated in form of z-score using WHO Anthro for participants below 5 years of age and WHO Anthroplus for participants aged 5-18 years old. The result of z-scores were then categorized based on the classification determined based on the Indonesian Minister of Health Regulation No. 2 of 2020. Secondary data comprises of disease stage, duration of disease, primary etiological factors, as well as past medical history of anemia, hypertension, and bone mineral disorder were recorded from medical records. Sociodemographic data, such as parental education levels, family economic status, age, and gender, were obtained through parent/guardian interviews and the completion of the Case Report Form (CRF).

**Statistics.** Data were analyzed using SPSS Statistic ver. 25. Normality test was performed using Saphiro-Wilk test. All numeric data with normal distribution were presented with mean and standard deviation, while data with abnormal distribution were presented in median and interquartile range. Correlation between the indicators of nutritional status with associated factors such as age and duration of disease were quantified using Pearson test for the normally distributed data and Spearman test for data with abnormal distribution. ANOVA test and Kruskal-Wallis test were used to analyze the relationship between the indicators of nutritional status with other factors such as stages of CKD, primary etiological factors, and comorbidities. Meanwhile, independent t-test and Mann-Whitney were used to analyze the indicators of nutritional status with gender of participants.

Results

A total of 18 patients were enrolled in this study. Majority of the participants were male (61.1%) with the median age of 13.5 years. The educational level of both parents was mainly high school or equal (38.9%) and most participants came from family with income below the minimum wage (55.6%). Patients were predominantly diagnosed with stage 5 CKD (38.9%) with median duration of disease of 8.5 months. Other baseline characteristics of participants were illustrated in Table 1.

Table 1. Characteristics of Participants

Variables	Frequency (n (%))
Age (years)*	13.5 (3.0-17.0)
Gender	
Male	11 (61.1)
Female	7 (38.9)
Mother’s educational level	
Primary school or equivalent	2 (11.1)
Middle high school or equivalent	5 (27.8)
Senior high school or equivalent	7 (38.9)
University	4 (22.2)
Father’s Level of Education	
Primary school or equivalent	2 (11.1)
Middle high school or equivalent	5 (27.8)
High school or equivalent	7 (38.9)
University	4 (22.2)
Family Economic Status	
Below the minimum wage	10 (55.6)
Above the minimum wage	8 (44.4)
Duration of Disease*	8.5 (3.0-26.0)
Stage of CKD	
G3a	5 (27.8)
G3b	3 (16.7)
G4	3 (16.7)
G5	7 (38.9)
Primary Etiological Factors	
CAKUT	5 (27.8)
SRNS	6 (33.3)
Chronic glomerulonephritis	2 (11.1)
Renal ciliopathies	1 (5.6)
Thrombotic microangiopathies	1 (5.6)
Others	3 (16.7)
Systolic Blood Pressure (mmHg)**	108.44 ± 20.11
Diastolic Blood Pressure (mmHg)**	65.44 ± 16.30

Variables	Frequency (n (%))
Hemoglobin (g/dL)**	10.48 ± 2.57
Mineral Bone Disorder	
Yes	2 (11.1)
No	16 (88.9)

Saphiro-Wilk test was used to analyze the normality of data distribution. \*Data presented in median (minimum-maximum). \*\*Data presented in mean ± standard deviation. CKD: chronic kidney disease, CAKUT: congenital anomalies of the kidney and urinary tract, SRNS: Steroid-Resistant Nephrotic Syndrome

The relation between the nutritional status indicator with the influencing factors were illustrated in **Table 2, 3 and 4**. Our study evaluated the relation between the indicators of pediatric nutritional status with CKD and various influencing factors. These factors include age, gender, parents’ level of education, family economic status, stage of CKD, duration of disease, primary etiological factors, and comorbidities such as hypertension indicated by systolic and diastolic blood pressure, anemia represented by hemoglobin level, as well as bone mineral disorder. The findings indicated no significant correlation between nutritional status and the influencing factors, except for height for age, which demonstrated a significant correlation with bone mineral disorders (p = 0.005).

**Table 2.** Correlation between nutritional status indicators with age, duration of CKD, systolic blood pressure (SBP), diastolic blood pressure (DBP), and hemoglobin

	Weight-for-Height (n: 3) <sup>a</sup>	Weight-for-Age (n: 8) <sup>b</sup>	Height-for-Age (n: 18) <sup>a</sup>	BMI-for-Age (n: 18) <sup>a</sup>
Age	r: 0.866 p: 0.333	r: -0.217 p: 0.606	r: -0.900 p: 0.722	r: -0.126 p: 0.618
Duration of CKD	r: 0.500 p: 0.667	r: 0.292 p: 0.483	r: -0.196 p: 0.437	r: -0.044 p: 0.863
Systolic Blood Pressure	r: -0.130 p: 0.917	r: -0.335 p: 0.417	r: -0.275 p: 0.270	r: 0.035 p: 0.892
Diastolic Blood Pressure	r: -0.541 p: 0.636	r: -0.395 p: 0.333	r: -0.369 p: 0.132	r: -0.241 p: 0.334
Hemoglobin	r: 0.581 p: 0.605	r: 0.168 p: 0.691	r: -0.155 p: 0.538	r: -0.035 p: 0.890

<sup>a</sup>analyzed using Pearson correlation test. <sup>b</sup>analyzed using Spearman correlation test. CKD: chronic kidney disease, BMI: body mass index.



**Table 3.** Relation between nutritional status indicators with stage of CKD, primary etiological factors, and parents’ educational level.

		Weight-for-Age <sup>a</sup>		Height-for-Age <sup>b</sup>		BMI-for-Age <sup>b</sup>	
		Mean rank	P	Mean square	P	Mean square	P
<b>Stage of CKD</b>			0.362	1.516	0.646	0.538	0.789
	G3a	6.00					
	G3b	3.00					
	G4	8.00					
	G5	3.80					
<b>Primary Factors</b>	<b>Etiological</b>		0.297	1.386	0.789	2.704	0.249
	SRNS	3.67					
	CAKUT	5.75					
	Others	2.00					
<b>Mother’s Level of Education</b>			0.570	0.399	0.937	3.152	0.201
	Primary school or equivalent	-					
	Middle high school or equivalent	4.50					
	High school or equivalent	3.75					
	University	6.00					
<b>Father’s Level of Education</b>			0.240	0.735	0.854	3.254	0.186
	Primary school or equivalent	8.00					
	Middle high school or equivalent	1.00					
	High school or equivalent	4.75					
	University	4.00					

<sup>a</sup> analyzed using ANOVA. <sup>b</sup> analyzed using Kruskal Wallis. CKD: chronic kidney disease, BMI: body mass index, SRNS: Steroid-Resistant Nephrotic Syndrome, CAKUT: congenital anomalies of the kidney and urinary tract

**Table 4.** Relation between nutritional status indicators with gender, bone mineral disorder, and family economic status

		Weight-for-Age <sup>a</sup>		Height-for-Age <sup>b</sup>		BMI-for-Age <sup>b</sup>			
		Mean Rank	p	Mean	Levene's Test	p	Mean	Levene's Test	p
<b>Gender</b>			0.127		0.568	0.420		0.444	0.568
Male		4.00		-2.96			-1.17		
Female		8.00		-2.32			-0.78		
<b>Bone Mineral Disorder</b>			0.827		0.738	0.005		0.957	0.767
Yes		5.00		-5.43			-0.62		
No		4.43		-2.37			-1.07		
<b>Family Economic Status</b>			0.386		0.729	0.657		0.158	0.935
Above the minimum wage		5.25		-2.52			-0.99		
Below the minimum wage		3.75		-2.86			-1.04		

<sup>a</sup> analyzed using Mann-Whitney test. <sup>b</sup> analyzed using independent t-test. BMI: body mass index

Discussion

Based on the BMI-for-age indicator, the average of nutritional status between participants in this study was -1.02, indicating a good nutritional status. This finding is aligned with a study in Brazil which reported that 79.8% of pediatric patient in the pre-dialysis phase of CKD had good nutritional status. Furthermore, the study also indicated that the nutritional status during the initial diagnosis and during follow-up did not exhibit significant changes, with majority maintaining good nutritional status.<sup>11</sup> Another study in Canada also documented a good nutritional status between children in the pre-dialysis phase of CKD, irrespective of gender, with the average z-score between male and female were 0.34±1.20 and 0.40±1.06, respectively.<sup>12</sup> Meanwhile, the nutritional status results among participants, based on height-for-age indicator, indicate short stature, with an average z-score of -2.71. The outcome is also consistent with a study in Brazil, which stated that 54% of children in pre-dialysis phase of CKD were classified as having very short stature.<sup>11</sup>

This study found no direct correlation between CKD stages and body weight, stature, and overall nutritional status. This contrasts with prior studies in the United States and Canada, which demonstrates that a decrease in GFR of 10 ml/min per 1.73m2

was associated with a reduction in height and weight in children, regardless of gender.<sup>12</sup> A study in Brazil also reported a decline in overall nutritional status along with the progression of CKD stages.<sup>11</sup> This was hypothesized to be caused by anorexia resulting from reduced food intake among advanced CKD patients and exacerbated by insulin resistance, leading to protein catabolism and muscle wasting.<sup>3</sup> This discrepancy could be caused by the small number of samples and the concentration of sample in certain CKD stages. Nutritional status was also not correlated with duration of CKD, which is inconsistent with previous study by Sozeri et al. However, the previous study also included patients undergoing dialysis as their participants, which differentiates their population from our study.<sup>7</sup> Another study by Greenbaum et al. reported similar results to our findings.<sup>14</sup> The nutritional status of children with CKD is primarily affected by the hormonal imbalance caused by the decrease of GFR due to kidney dysfunction. However, it is also influenced by the type of therapy received and patients' current condition. Hence, assessing the relation between nutritional status and the duration of disease is better to be conducted on patients with the same stages of diseases.<sup>3</sup>

Our study also found no correlation between nutritional status and several indicators, including age, genders, primary etiological factors, and family economic status, which is consistent with previous studies.<sup>6, 7, 15, 16</sup> The absence of correlation between nutritional status and age could be attributed to other factors influencing the disease. Hogan et al. revealed that nutritional status is not related to the age of patient if the patient has already developed CKD before others, supporting the finding.<sup>17</sup> Additionally, there is no specific phase of growth that could lead to a different nutritional status between male and female children, confirming our result. The difference in growth between boys and girls is relatively minor during the critical growth phase in the first 1000 days of life.<sup>18</sup> Furthermore, there is no significant difference in the nutritional needs of both genders during the prepubertal phase. Despite experiencing growth spurt at different times and having different nutritional needs, both genders eventually undergo growth spurts and changes in body composition, leading to similar nutritional status.<sup>19</sup> Nutritional status is also not associated with the primary etiological factors of CKD. This could be due to the mechanism of malnutrition in CKD being directly related to the consequence of kidney dysfunction, regardless of its primary etiology.<sup>3, 15</sup> The lack of correlation between nutritional status and family economic status is expected, as economic status should not be a barrier for families to seek appropriate healthcare for patients with CKD, with the existence of national health insurance in Indonesia.<sup>20</sup> However, there is currently no government funding that covers the expenses of daily food for children, which is essential for their growth. Particularly for children with CKD, supplementary food is often needed despite its high cost, which is also not covered by the

government.<sup>21</sup> Thus, we do acknowledge that family economic status may indirectly affect the conditions of patients with CKD if the expenses are not prioritized wisely.

This study also reported that parents' level of education is not associated with the nutritional status of children with CKD in the pre-dialysis stage. This is because parents' level of education only partially influences parental upbringing. Parenting styles could be affected by several factors, such as the number of children, antenatal and postnatal care, as well as the family's economic status. Additionally, the quality of education should also be considered. Alderman et al. suggest that parents with a history of education in areas with better educational quality had children with better nutritional status.<sup>22</sup> This concludes that parental education is only one of the various factors influencing the decision-making in child-rearing and is not directly related to nutritional status.

There is no association between nutritional status with anemia and hypertension in this study. This finding is aligned with study by Rodig et al. and Flynn et al. which demonstrated that there is no significant correlation between nutritional status with anemia and hypertension.<sup>13,23</sup> Both anemia and hypertension in CKD occurs as a result of a multifactorial process which does not directly cause the chronic malnutrition in children necessary to impair their growth and overall nutritional status.<sup>23,24</sup>

Conversely, this study revealed a significant correlation between bone mineral disorders and the average of height-for-age during the pre-dialysis phase of children with CKD ( $p = 0.005$ ). Bone mineral disorders is often found in pediatric patient with CKD as CKD induces growth hormone abnormality which may impact the height of children through the increase of TNF alpha cytokine levels induced by chronic inflammation in patients. This elevation is known to inhibit RUNX2, a transcription factor associated with osteoblast differentiation, and stimulate osteoclastogenesis, ultimately leading to disturbances in bone mineralization.<sup>8</sup> Changes in bone mineralization are monitored through numerous laboratory testing, such as serum calcium levels, PTH, phosphate, and alkaline phosphatase activities, since the stage G2 of CKD. This is also done along with routine anthropometric measurement at least every 1 to 3 months. Additionally, certain organizations such as the European ad hoc Committee on Assessment of Growth and Nutritional Status in Peritoneal Dialysis recommending a more intense anthropological monitoring of once monthly measurement for children under the age of 5 and every two months for older children and every 6 – 12 months for bone mineralization marker examinations.<sup>25</sup> The target therapy for bone mineral disorder is a normal level of phosphate and calcium according to age. For patient experiencing growth disturbance, recombinant human growth hormone is recommended as treatment.<sup>26</sup>

As this study is a pilot study in Indonesia, this study possesses several advantages and drawbacks. This study aimed to determine the correlation between CKD stages and malnutrition in children which could provide novel information about the impact of CKD progression in a child's overall nutrition and growth status especially among children with pre-dialysis and dialysis stages of CKD. Despite these advantages, this study also possesses several limitations such as small sample size and small sample variability as the majority of the samples are male children with stage 5 CKD. Lack of samples variability also made it impossible to identify whether certain groups of CKD stages and certain genders possess better nutritional status than others. Therefore, further study with more variations in CKD stages and nutritional status is still needed in the future to help validate the findings of this study.

## Conclusion

In conclusion, children with CKD stage 3 – 5 pre-dialysis have normal nutritional status according to BMI for age and weight for height. However, according to weight for age and height for age, these children are underweight with short stature. Therefore, BMI alone should not be used to evaluate nutritional status. Bone mineral disorders is also found to be correlated with short stature. For this reason, early detection and intervention in addressing growth faltering among children with CKD is paramount to improve their quality of life.

## Conflict of Interest

None declared

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## Case Report

# Water-beads Ingestion with Intestinal Obstruction in Children: A Case Report

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**Abstract:**

**Background:** Foreign body ingestion often occurs in the pediatric population and can cause no symptoms or even cause complications due to swallowing the foreign body. The aim of this case report is to increase clinician knowledge of cases of foreign body ingestion which can cause complications.

**Case:** We report a case of foreign body ingestion water beads with complications of intestinal obstruction in a 1 year 2-month-old girl. The patient came with complaints of vomiting more than ten times containing fluid accompanied by decreased intake. The patient was suspected of swallowing water beads. The patient came to the emergency room with no signs of acute abdomen and the results of plain abdominal radiograph showed no foreign objects. The patient began to show symptoms of obstruction, not being able to defecate and not being able to pass gas. Physical examination revealed abdominal distention accompanied by inaudible bowel sounds. Abdominal CT scan results showed ileal obstruction. An exploratory laparotomy was performed, and two water beads were found intraoperative, which were the cause of the obstruction. The foreign body was removed. After surgery, the patient experienced gradual clinical improvement until he was discharged ten days after the procedure.

**Discussion:** Foreign body ingestion in children is often not witnessed by anyone and may not cause symptoms until complications such as obstruction or peritonitis occur. Some foreign objects are radiolucent in plain radiographs. In emergency cases of foreign body ingestion, it is necessary to carry out emergency endoscopic procedures, even emergency surgical procedures in cases that cause complications.

**Conclusion:** This case report increases clinician knowledge and awareness regarding the clinical approach in evaluating patients with suspected foreign body ingestion in children.

**Keywords:** children, foreign body ingestion, intestinal obstruction, water beads

## Introduction

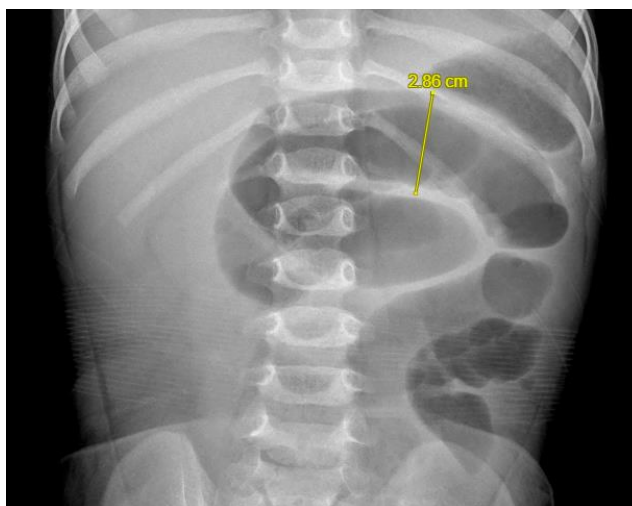
Foreign body ingestion often occurs in the pediatric population as an accidental event. Approximately 75% cases of foreign object swallowing occur in children under 4 years of age,<sup>1</sup> with the highest incidence in the age range of 6 months to 3 years.<sup>2</sup> As many as 50% of cases are asymptomatic.<sup>3</sup> In America, coins are the most frequently swallowed foreign object, whereas in other countries it is fish spines.<sup>4</sup> The death rate due to swallowing foreign objects is relatively low,<sup>5</sup> estimated at around 3%.<sup>6</sup> Swallowed foreign objects can cause complications if they block the digestive tract in parts that experience anatomical narrowing such as the upper and lower esophageal sphincter, pylorus, ileocaecal valve, and anus<sup>7</sup> so that it requires endoscopy and even surgery. The aim of this case report is to increase clinician awareness in establishing a diagnosis of foreign body ingestion, especially in foreign bodies that are not visualized radiologically so that appropriate treatment can be obtained.

## Case

A girl aged 1 year 2 months, weight 8.2 kg, body length 75 cm, came to the emergency department (IGD) brought by her parents with complaints of vomiting more than 10 times containing liquid since one day before entering the hospital. The patient's food and drink intake decreases, accompanied by weakness and flatulence. The patient was suspected by his parents of swallowing water-beads before the symptoms appeared. The patient had his last bowel movement two days before entering the hospital and was still able to pass gas one day before entering the hospital. The patient came to the emergency room in moderate pain, *compos mentis* consciousness, tachycardia 120x/minute, sub-febrile (37.8°C). The initial physical examination in the emergency room did not reveal any signs of an acute abdomen, it looked slightly convex, soft to touch and bowel sounds were still heard. Laboratory examination results are within normal limits. A plain abdominal radiograph was performed, and the results showed dilatation of the small intestine in the left upper abdominal region which was suspected of partial obstruction (**Figure 1**). No foreign objects were seen on the plain abdominal radiograph. It is recommended that an abdominal CT scan be performed for a more detailed evaluation.

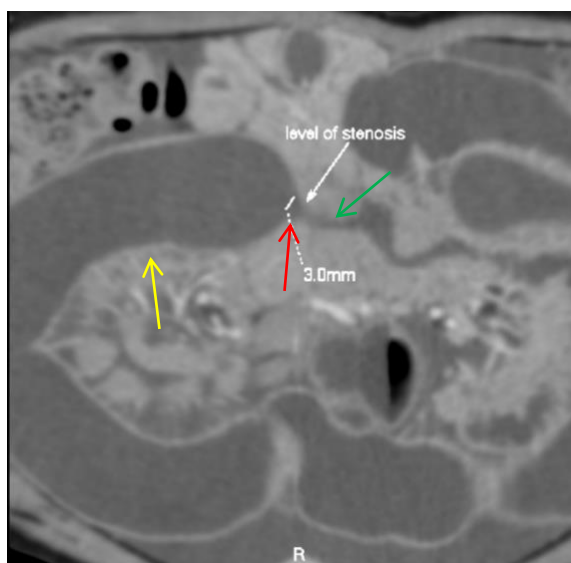
Then the patient was hospitalized and fasted, given medical therapy and an abdominal CT scan. The patient was also consulted to a pediatrician gastroenterologist and pediatric surgeon. In the first 24 hours of treatment, the patient's symptoms increased with the stomach appearing larger than before. Patients also tend to become increasingly weak with decreasing food and drink intake. The frequency of vomiting also increased to fifteen times in the last 24 hours accompanied by inability to pass gas. The patient's consciousness was somnolence-apathetic with a GCS of 14. The patient was given a 2 lpm nasal cannula, a decompression NGT was installed and transferred to the pediatric intensive care unit (PICU). Vital signs were still good,

physical examination of the abdomen revealed distention with an abdominal circumference of 48 cm and bowel sounds were not audible.



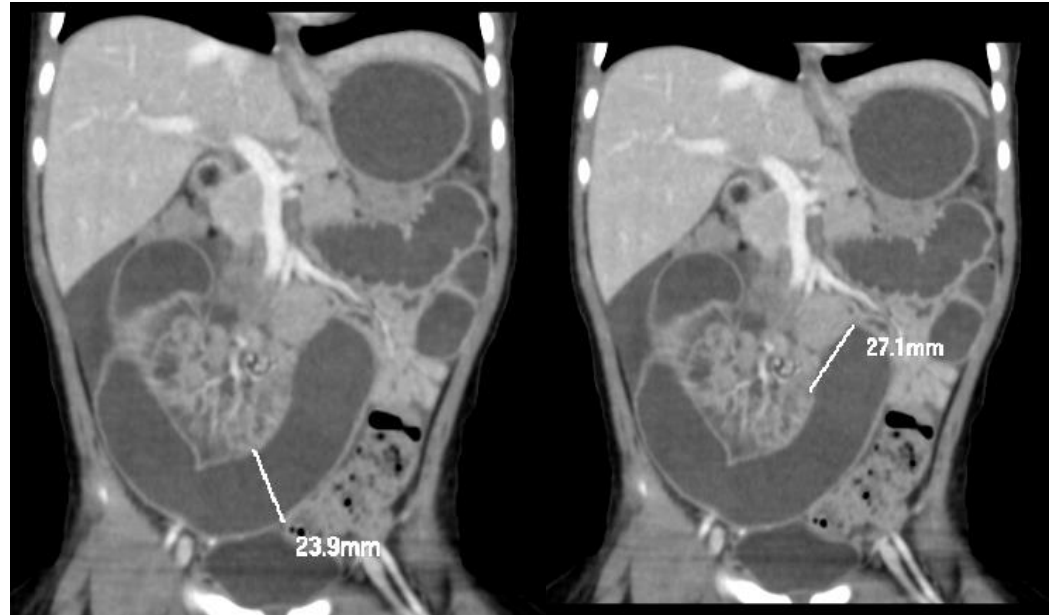
**Figure 1.** Plain abdominal radiograph shows intestinal dilatation (yellow line) with an estimated diameter of 2.86 cm caused by obstruction.

The results of an abdominal CT scan showed dilatation of the distal segment of the jejunum to the proximal segment of the ileum due to significant stenosis accompanied by multiple air fluid levels. This picture is consistent with total obstructive ileus. Other findings from the CT scan were ascites and multiple lymphadenopathy in the mesentery. There were no visible foreign objects on the CT-scan image. (**Figure 2 and Figure 3**)



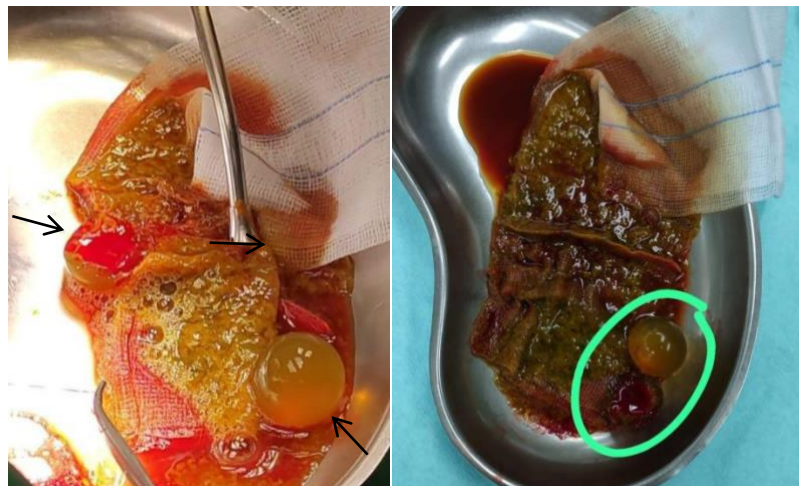
**Figure 2.** CT-scan of the abdomen without coronal contrast shows obstruction of the ileum (red arrow) with dilatation in the proximal segment (yellow arrow) and collapse in the distal segment (green arrow) of the obstruction.





**Figure 3.** Abdominal CT scan without contrast sagittal section shows intestinal dilatation with a diameter of 23.9 - 27.1 mm (white line).

Emergency exploratory laparotomy was performed. Intraoperatively, it was found that the ileum was dilated and appeared to have collapsed, including two round cystic masses (children's ball toys) that could not be pushed distally. It was suspected that the toy was causing obstruction and evacuation of the corpus alienum was carried out. (**Figure 4**) Resection-anastomosis was not performed because the intestinal organs were still vital/viable.



**Figure 4.** Intraoperative findings showed two water-bead foreign bodies (black arrows/circles) colored red and yellow which were the cause of obstruction.

After surgery, the patient continued treatment in the PICU with medication and a gradual diet plan. The patient's condition slowly improved, and the patient was discharged ten days after surgery.

## Discussion

Foreign body ingestion often occurs in children, especially in the age group 6 months to 3 years. Various foreign objects can be found with the most frequency, namely coins, batteries, magnets, fish bones, and even children's toys. Water-beads are spherical gelatin toys with a size of 2-3 mm which can expand when exposed to water, especially at alkaline pH.<sup>8</sup> This children's toy contains superabsorbent polymer material which has increased in popularity recently. Beads expand quickly on varying scales so that marble-sized dry beads can easily expand and cause obstruction in the digestive tract.<sup>9</sup> Studies show that the diameter of water beads can increase from 2 to 9.5 mm and from 7.5 to 40 mm if exposed to water for 12 hours, so swallowed water beads will clog the small intestine which only has a diameter of 25-30 mm.<sup>10,11</sup> Children will be interested in swallowing the toy because it looks like candy with attractive bright colors. So far, there have only been four publications reported and one resulted in death.<sup>12</sup> Our patient was 1 year 2 months old who was in the group susceptible to swallowing foreign objects, namely under 3 years of age. Based on the anamnesis, the foreign object suspected of being swallowed by the patient was a child's water-bead toy. This toy has the potential to cause gastrointestinal obstruction because it expands easily.

The diagnostic approach that can be taken is a thorough history taking regarding the type, quantity, when the foreign object was swallowed by the child, whether the child vomited the foreign object or whether the foreign object was excreted in the feces. Symptoms resulting from foreign body ingestion may vary, ranging from no symptoms to severe symptoms that require immediate action. Symptoms that usually appear are nausea, vomiting, and refusal to eat. Vomiting blood and coughing may also occur. If a foreign object has passed through the stomach or intestines, symptoms of abdominal pain, vomiting and bloody bowel movements may appear.<sup>5</sup> Foreign objects that have passed through the gastro-esophageal junction or have reached the distal gastrointestinal tract can cause symptoms of obstruction or perforation such as abdominal pain, fever, nausea, and peritonitis. Impaction, perforation and obstruction often appear in areas of narrowing such as the area at the level of the cricopharyngeal muscle and ileocecal valve.<sup>7</sup> Other areas that are also of concern because of the risk of blockage are the pylorus and the C-loop form of the duodenum.<sup>6</sup> Assessing the general condition, vital signs, whether there is an emergency, as well as the airway and breathing must be the main focus. The physical examination is continued by assessing whether there are signs of obstruction, acute abdomen, and peritonitis. The initial symptoms found in our patient were vomiting, decreased oral intake and weakness.



There were no signs of acute abdomen at baseline. As time progresses, signs of gastrointestinal obstruction caused by the water beads begin to appear, which may be related to the passage of the water beads through the esophagus, stomach and into the intestines. The patient's consciousness tends to decrease even though the hemodynamics is still stable. The patient's abdomen became increasingly distended, followed by inability to defecate or pass gas and showed signs of an acute abdomen.

The first radiological examination carried out is plain abdominal radiograph. Metallic foreign objects such as coins and batteries will be clearly visible on a pop-up photo, but radiolucent foreign objects may not be visible. Beads are radiolucent and difficult to detect on plain radiographs,<sup>10</sup> so swallowing water beads carries the risk of delaying diagnosis and causing obstruction. Our patient had a plain abdominal radiograph done and no foreign objects were found. This is in accordance with the nature of water beads which are radiolucent, so they are not visible on plain photographs. The initial radiological finding in our patient was abdominal dilatation. Imaging was continued with an abdominal CT scan without contrast. From the results of the CT scan, it was found that there was obstruction, but the cause was not known for certain and there was no visible foreign object.

Around 90% of foreign bodies in the esophagus can pass spontaneously without causing complications, but some cannot pass through the pylorus, duodenum and ileocaecal valve so that around 10% of swallowed foreign bodies still remain in the digestive tract.<sup>13</sup> As many as 10-20% of cases of foreign body ingestion require emergency endoscopic intervention and only 1% require surgical intervention.<sup>6,14</sup> Management of superabsorbent polymer foreign bodies is challenging because they are radiolucent and usually pass through the proximal gastrointestinal tract easily until their size increases causing obstruction. In cases of beads swallowing, emergency endoscopic evacuation is required immediately. If upper gastrointestinal endoscopy fails to find the foreign body, then it is reasonable to suspect that the beads have reached the distal part of the intestine and are at risk of causing obstruction. If this happens, then surgery needs to be considered.<sup>9</sup>

A literature review reported forty-three cases of intestinal obstruction caused by superabsorbent polymer-made product ingestion (beads).<sup>15</sup> The patient's characteristics are in the age range from 6 to 36 months, with the beads always located in the small intestine between the duodenum and the terminal ileum. Endoscopic procedures for removal of the beads were performed in two cases and operative procedures were performed in the other forty-one cases, including enterotomy in thirty six cases and resection in five cases.

A case report reported two cases of water beads ingestion.<sup>16</sup> The first case was a 15-month-old-boy with well-defined anechoic cystic lesion within the dilated proximal jejunum underwent an exploratory laparotomy. Intra operative, a large jelly ball measuring 3x3cm in diameter was found. Post operative, NGT output was quite significant, producing 450mL of greenish fluid. Abdominal USG was repeated on the fourth day after surgery and revealed two round anechoic structures measuring 3x3cm in jejunum. A second laparotomy was performed and two expandable jelly balls in the jejunum removed. The second case was 18-month-old boy with two well defined cystic structures measuring 3x3.7cm in the terminal ileum underwent an exploratory laparotomy and found three gel balls in the jejunum. The patient was discharged on 4th day after surgery.

In another case report, a 6-month-old male infant swallowed one water bead had to be operated because it caused obstruction.<sup>12</sup> The first operation was enterotomy. However, on the 6th postoperative day, the patient developed burst abdomen and was re-operated. At exploration anastomotic leak was found. The anastomosis was revised. The patient developed septicemia and died two days after the second operation.

Our patient was 1 year 2 months old, included in vulnerable age that prone to beads ingestion. From plain abdominal radiograph and abdominal CT scan only found dilatation and obstruction, no foreign object was detected. In our patient, surgery was performed because the patient showed signs of acute abdomen and obstructive ileus. Because it was not known for certain what the cause was, an exploratory laparotomy was carried out and it turned out that two water bead foreign objects were found which were the cause of the obstruction. Our patient did not undergo endoscopy because of suspicion of obstruction in the lower gastrointestinal tract and he already showed signs of obstruction which is a strong indication for operative treatment. An exploratory laparotomy was performed to evacuate the foreign body and no resection-anastomosis was performed because the intestine was still vital and viable. After successful evacuation of the foreign body, our patient experienced gradual clinical improvement until discharge.

## Conclusion

Incidences of foreign objects being swallowed by children are quite common in the vulnerable age group between 6 months and 3 years. Patients who swallow foreign objects may not cause symptoms until they cause complications such as obstruction, peritonitis depending on the type and nature of the foreign object, the duration of the swallowing, and the location of the foreign object. A careful history can direct suspicion towards swallowing a foreign object, especially information from parents or witnesses who saw the patient swallow a foreign object is very important to know.

The physical examination includes monitoring hemodynamics, generalist status, and specifically paying attention to signs of obstruction or peritonitis. The first radiological imaging performed is a plain radiograph but remember that not all foreign objects can be seen. Emergency endoscopic procedures are sometimes needed in emergency cases if the foreign body is still in the upper gastrointestinal tract and operative measures are needed for cases complicated by obstruction or peritonitis.

## Conflict of Interest

None declared

## Funding Statement

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## Case Report

# Recurrent Esophageal Stricture in a Child Post Steven-Johnson Syndrome: A Case Report

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**Abstract:**

**Background:** Esophageal stricture is an abnormal narrowing of the esophageal lumen, resulting in dysphagia. Despite its rarity, this condition could be caused by various etiologies, including Steven-Johnson Syndrome (SJS). In some cases, stricture could recur, which complicates the management. This case report presented a rare case of refractory esophageal stricture in children with Steven-Johnson Syndrome.

**Case:** A 5-years-old boy with a prior history of SJS presented with dysphagia for one month. The patient experienced choking, blood vomiting, stomatitis, swelling on the lips, and difficulty in swallowing solid food. Barium meal and EGD test confirmed the diagnosis of esophageal strictures. The patient then underwent dilation using bougie dilator. However, he continued to experience dysphagia, resulting in a total of 15 serial dilation sessions.

**Discussion:** Esophageal dysphagia is observed in patients who experience difficulty swallowing solid food. SJS can contribute to the development of esophageal dysphagia by causing inflammation of the esophageal mucosa, resulting in lesions and strictures. In patients with esophageal strictures, two types of dilation methods are available: bougie dilator with wire guidance (Savary-Gilliard) and balloon dilator, with the current consensus for dilation procedures supporting the rule of three. For patients with refractory strictures, other modalities such as mitomycin-C injection and stent placement are also available. Esophageal replacement surgery is considered as the last resort for refractory stricture patients who have not responded to previous treatments.

**Conclusion:** Steven-Johnson Syndrome is a rare cause of esophageal strictures. The management of refractory esophageal stricture requires a comprehensive subspecialty care and long-term monitoring.

**Keywords:** dysphagia, esophageal stricture, steven-johnson syndrome, refractory

## Introduction

Esophageal stricture is a rare cause of dysphagia in children. It is defined as an abnormal narrowing of the esophageal lumen due to various etiologies. The most common etiology is the ingestion of corrosive substances. Other etiologies include radiation-related injury, post-anastomosis stricture, and eosinophilic esophagitis.<sup>1</sup> Inflammation of esophageal mucosa in Steven Johnson syndrome is an atypical cause of esophageal stricture.<sup>2</sup> Additionally, management of esophageal stricture remains challenging. Balloon dilation or bouginage is the initial management for esophageal stricture.<sup>3</sup> However, in some cases, evaluation after several dilation sessions revealed a recurrent or refractory esophageal stricture. Administration of steroids or Mytomyacin-C has become one of the treatment choices for those experiencing refractory stricture.<sup>4</sup> Other alternatives, such as stent placement also reported to be successful in some refractory cases. This case report presented a case of refractory esophageal stricture in children with Steven-Johnson Syndrome.

## Case

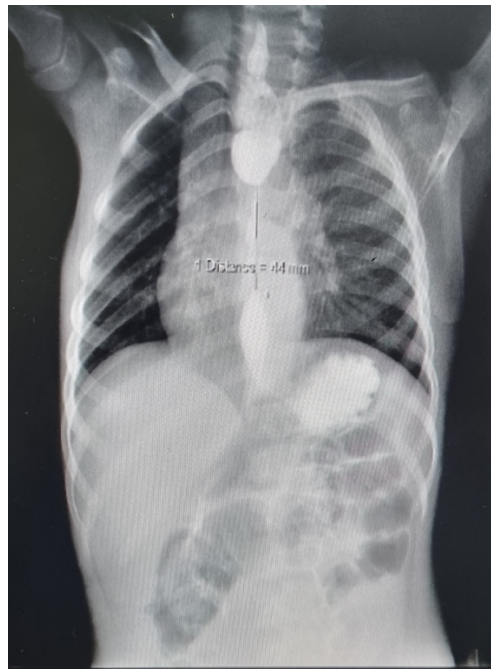
A boy aged five years and eight months old came with a chief complaint of difficulty swallowing for the past month. A month prior, the patient had a choking episode, followed by vomiting reddish vomit mixed with food. He also experienced stomatitis and swelling on the lips (**Figure 1**). The symptoms occurred after the administration of acetylsalicylic acid. He was then hospitalized for three days. Two weeks after the admission, the patient had another episode of vomiting. He could only eat porridge and was unable to eat solid food. Upon eating solid food, the patient felt something stuck in his throat, causing him to vomit. During this period, the patient lost 3 kg of body weight. Physical examination showed no abnormalities in the mouth and tonsils.



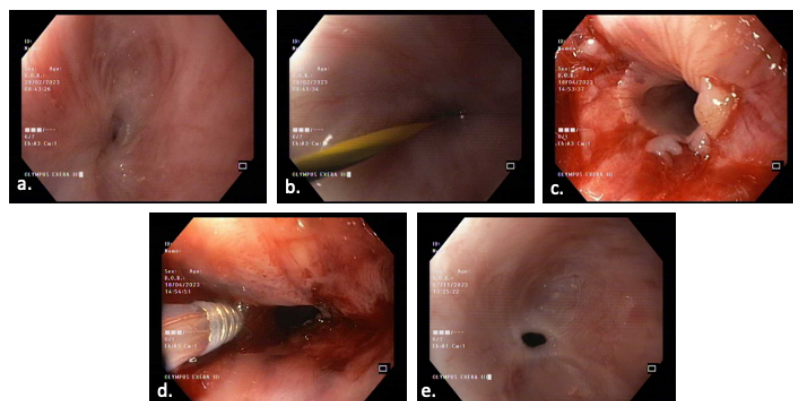
**Figure 1.** Stomatitis dan Swelling presented on the patient



Rhinopharyngolaryngoscopy (RFL) test revealed laryngopharyngeal reflux, adenoid hypertrophy, and mechanical dysphagia. Further examination with flexible endoscopic evaluation of swallowing (FEES) showed good swallowing movement in the pharyngeal phase but also noted reflux from the esophagus. The barium meal test indicated an esophageal stricture from thoracic vertebra 5 to 8, measuring 4.5 centimeters in length (**Figure 2**). The result of esophagogastroduodenoscopy (EGD) also demonstrated a 5-centimeter esophageal stricture, 15 centimeters from the incisors, and a presentation of gastroduodenitis. No furrowing was found in the esophagus (**Figure 3**).



**Figure 2.** Barium Meal Test



**Figure 3.** EGD Results. a. Esophageal stricture pre-dilation; b. First dilation; c. Laceration post-dilation; d. Triamcinolone injection post-dilation; e. The diameter of lumen after multiple dilation sessions



The patient then underwent dilation using Savary-Gilliard bougie number 7 and nasogastric tube insertion with the assistance of a nasal scope. Biopsies from the duodenum, antrum, and distal esophagus revealed no eosinophils was found in any tissues. A second EGD was conducted one month after the first procedure, with the result of an 8-millimeter stricture. Dilation procedures were then repeated using bougie no. 8, 9, and 10.8, until a 9.2 mm scope could enter the stomach and duodenum. The examination showed strictures were still present at 6cm, 14 cm, and 20 cm from the incisor. During the follow-up evaluation three weeks after the procedure, the patient still reported difficulty swallowing solid food. Triamcinolone 20 mg was then injected at several laceration points during the fourth to sixth dilation sessions. Despite the intervention, the patients continued to experience dysphagia 3-4 weeks post-dilation, leading to a total of 15 serial dilation sessions.

## Discussion

Swallowing is a complex process that occurs sequentially. The swallowing process can be categorized into 3 phases: the oral phase, pharyngeal phase, and esophageal phase. Dysphagia can occur in any of these stages, either acutely or chronically. Acute inflammation along the swallowing pathway may cause dysphagia, including retropharyngeal abscess, diphtheria, acute epiglottitis, or Steven-Johnson Syndrome (SJS). Acute dysphagia occurring in the esophageal phase could be caused by swallowing a foreign body, swallowing a caustic substance, or esophageal perforation.<sup>1</sup>

The complaint of difficulty swallowing experienced by the patients one month prior indicated an acute dysphagia. History-taking that should be assessed for this condition includes the presence of ingested foreign objects, pain during swallowing, pain related to eating, fever, skin or mucosal lesion, voice disturbance (dysphonia), acute neurological deficit, and whether swallowing difficulty is limited to solid food or also occurs while consuming liquid food. Dysphagia related to eating might result from gastroesophageal reflux disease (GERD). The presence of fever suggests an infectious etiology. Furthermore, dysphagia occurring only due to solid food indicates esophageal dysphagia, while dysphagia due to both solid and liquid food suggests an esophageal motility disorder such as achalasia.

From the history taking, the patient experienced difficulty swallowing, lesion and swelling on the lips after drug consumption (acetylsalicylic acid). The patient could not swallow solid food but could take liquid food, suggesting esophageal dysphagia. Dysphagia due to solid food is usually caused by esophageal inflammation resulting from GERD, eosinophilic esophagitis (EoE), or esophageal stricture.<sup>5</sup> He then underwent esophageal contrast meal and esophagogastroduodenoscopy with histopathological examination.<sup>6</sup>

The history of lip swelling and bloody vomit after taking acetylsalicylic acid indicated that the cause of dysphagia was esophagitis due to SJS. Steven-Johnson syndrome could cause complications in many organ systems, including gastrointestinal. Among the SJS patients who were suspected of having gastrointestinal complication and underwent endoscopy, 11% of them had abnormalities such as ulceration or stricture, primarily in esophagus.<sup>2</sup>

Barium meal examination confirmed the occurrence of esophageal stricture, which was also visualized using EGD. One of the potential etiologies of esophageal stricture that needs to be considered is EoE. However, the endoscopic findings typically associated with EoE, such as exudate, trachealization, and furrowing, were not observed in this case. Furthermore, the histopathological result indicating EoE, which would be presented as significant eosinophils ( $\geq 15$  cells/LPB) in the esophagus, were also not found in this case.<sup>7</sup>

Two dilation techniques are available to treat esophageal stricture: bougie dilator with wire guidance (Savary-Gilliard) and balloon. Balloon dilation has better safety profile and lower rate of failure compared to the bougie dilation.<sup>3</sup> However, bougie dilation is a safe and effective procedure; thus, it is still commonly practiced in hospital with limited resources. The experience of the hospital and healthcare provider performing one of the dilation techniques also determined the success of therapy.<sup>8</sup> Bougie dilation was used in our case. Currently, there is still no guideline regarding the time and frequency of dilation for children with esophageal stricture. Research comparing the dilation on esophageal atresia per three weeks with on-demand revealed that the on-demand group underwent fewer dilations with the same efficacy and safety.<sup>9</sup> The current general consensus is the rule of three: maximum dilation up to three times the diameter of stenosis, maximum three dilations per session (with an increase of 1 mm per dilation) after resistance, and a minimum interval of three weeks between dilation sessions.<sup>9</sup> In infants under three months of age, optimal dilation could be achieved with 8-10 diameter dilation, while in older children, it can be achieved with 10-15 mm diameter dilation.<sup>10</sup> Long-term management of our patient was challenging, mainly due to the recurrent stricture after dilation sessions. Refractory esophageal stricture occurs due to scarring or fibrosis in the lumen, causing the recurrence of narrowing without inflammation. Refractory stricture is diagnosed if the expected esophageal diameter cannot be achieved after five sessions with maximum four-week intervals, or if the expected diameter could not be maintained for four weeks after the diameter is achieved.<sup>11</sup> In our case, the patient had undergone 15 times of dilation, establishing the diagnosis of refractory stricture. Currently, there is still no standard therapy for refractory stricture. However, several modalities that can be used for refractory stricture.

The application of mitomycin-C at the mucosal lesions post-dilation has been reported to be beneficial in reducing the total dilation sessions. Mitomycin-C is an antifibrotic and cytostatic agent anthracycline group that inhibits fibroblast proliferation and reduces scar formation. For esophageal strictures, mitomycin-C is administered by applying a cotton swab soaked in a 0.1 mg/mL mitomycin-C solution directly to the mucosa following dilation. Mitomycin-C was shown to be safe and effective for refractory strictures.<sup>4</sup>

Management using plastic or nickel-titanium alloy stents could also be considered as they are removable and able to handle angulation in the esophagus. The use esophageal stent in children increased with the presence of self-expandable metal or plastic stents. There is still no guidance regarding the duration of stent insertion, causing varying duration from 1 to 24 weeks. A study in pediatric patients demonstrated an effective use of stents in 52% of the cases without further intervention needed.<sup>12</sup> Additionally, intralesional injection of triamcinolone has been reported to be effective for refractory stricture in adult patients. The main advantage of triamcinolone injection lies in its relatively safe administration technique and its ability to reduce the frequency of dilation sessions. The dosage of triamcinolone administered to patients varied across different centers, typically ranging from 20 mg to 40 mg per lesion. In pediatric patients, the use of intralesional triamcinolone has been reported to yield varying outcomes, with some showing success while others do not.<sup>13</sup>

Esophageal replacement surgery is the last choice of treatment for refractory stricture. Several organs were reported eligible for esophagus substitution, including colon segments, stomach, and small intestine segments, but none can perfectly replace esophageal function. The long-term prognosis for colon interposition following pediatric esophagectomy also did not exhibit promising results with high morbidity and multiple organ disorders, including gastrointestinal symptoms (85%), respiratory symptoms (58%), and eating difficulties (50%). Furthermore, the majority of patients experienced failure to thrive.<sup>14</sup>

## Conclusion

Steven Johnson Syndrome is a rare cause of esophageal strictures. The management of refractory esophageal strictures requires comprehensive subspecialty care and long-term monitoring. In this case, the treatment plan for refractory stricture includes the application of mitomycin-C or stent placement. Consultation with pediatric surgeon for esophagus substitution therapy should also be considered if dilation results are not optimal, despite the high morbidity.

## Conflict of Interest

None declared

## Funding Statement

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## Literature Review

# Neonatal Gastrointestinal Emergencies

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Alatas FS. Neonatal Gastrointestinal Emergencies. *Arch Pediatr Gastr Hepatol Nutr*. 2024; 3(1):41-57.

**Abstract:**

**Background:** Neonatal gastrointestinal emergencies refer to a set of life-threatening conditions affecting the digestive system of a newborn within the first 28 days of life and often associated with high morbidity and mortality. As such, these conditions require immediate and accurate diagnosis as well as proper treatment to optimize the outcomes of these patients. This condition has the potential to obstruct the flow of gastric content leading to vomiting, failure to thrive, and electrolyte imbalances.

**Discussion:** Gastrointestinal obstruction is one of the most common conditions causing emergency condition in neonates. This condition may occur anywhere between the upper part of gastrointestinal tract to the lower gastrointestinal tract. In most cases of neonatal gastrointestinal emergencies, patients almost always present with vomiting that may be bilious or non-bilious. Furthermore, persistent vomiting may also lead to a more severe consequences such as hypovolemic shock and electrolyte imbalances. Therefore, clinicians are expected to address this problem early while also working to find the underlying etiologies of neonatal vomiting. On the other hand, gastrointestinal bleeding is often an alarming sign that indicates a possible emergency condition in neonates. However, some non-emergency condition such as swallowed maternal blood and cow's milk allergy can also result in gastrointestinal bleeding in neonates.

**Conclusion:** Given the critical time window and the vulnerability of the neonate population, the proper identification and prompt treatment of neonatal gastrointestinal emergencies is crucial to minimize morbidity and mortality. Multidisciplinary management with neonatologists, pediatric surgeons, radiologists, and nursing staff working closely together can provide the best possible outcomes.

**Keywords:** emergency, gastrointestinal, neonates

## Introduction

Based on the latest World Health Organization (WHO) data, 11 neonatal deaths per 1000 live births are reported in Indonesia.<sup>1</sup> Among those numbers, neonatal emergencies play a major role in contributing to the neonatal mortality rate especially in developing countries. These emergency conditions may arise from various conditions such as infection, respiratory failure, shock and gastrointestinal problems.<sup>1,2</sup>

Neonatal gastrointestinal emergencies refer to a set of life-threatening conditions affecting the digestive system of a newborn within the first 28 days of life and often associated with high morbidity and mortality. As such, these conditions require immediate and accurate diagnosis as well as proper treatment to optimize the outcomes of these patients. The range of gastrointestinal emergencies includes structural anomalies, such as atresia and fistula, which interfere with food intake and can cause respiratory distress. Another category is necrotizing enterocolitis, characterized by severe inflammation and necrosis of the intestines, leading to systemic illness. All of these conditions have the potential to obstruct the flow of gastric content leading to vomiting, failure to thrive, and electrolyte imbalances.

In some conditions, prenatal imaging is sensitive enough to determine the underlying cause of gastrointestinal emergencies in neonates. However, in most patients, careful history and physical examination as well as imaging modalities are crucial for an accurate diagnosis. Plain radiography, fluoroscopy, ultrasound are the most common modalities being used to diagnose gastrointestinal emergencies.<sup>3</sup> The aim of this review is to discuss the neonatal gastrointestinal emergencies while reviewing the appropriate imaging options as well as focusing on their radiological features and the diagnostic algorithm.

## Approach to Neonatal Intestinal Obstruction

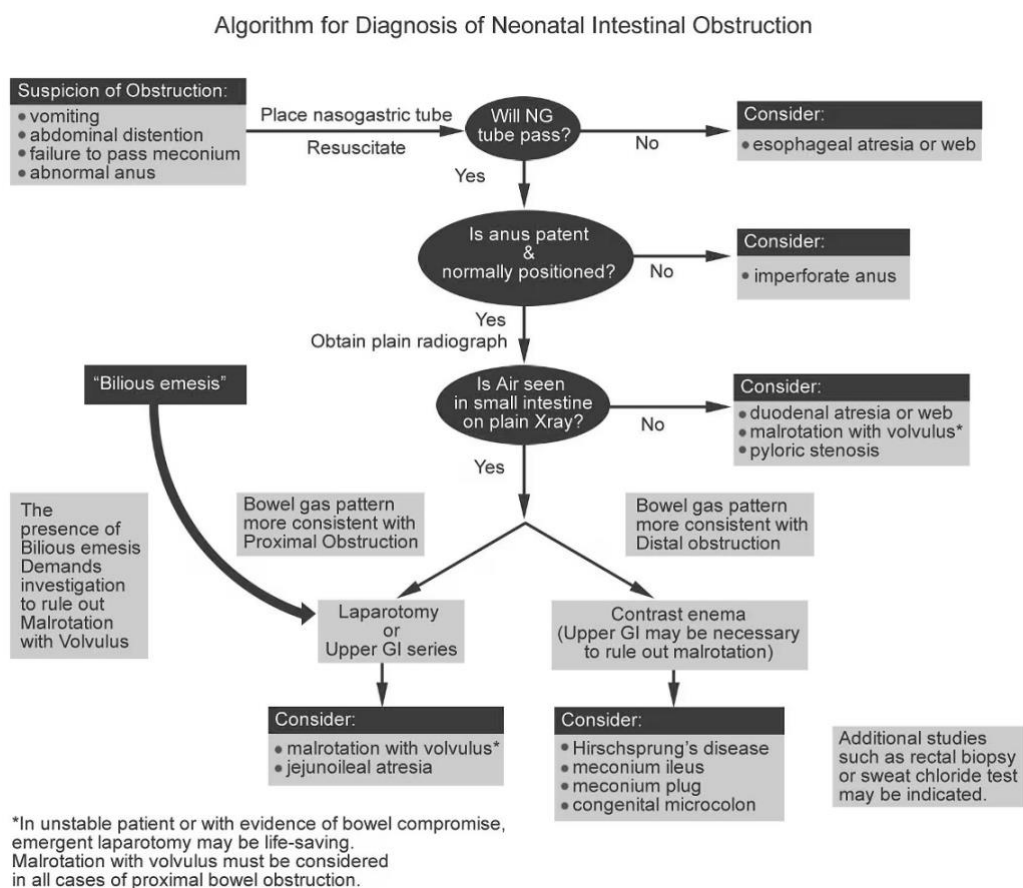
Gastrointestinal obstruction is one of the most common conditions causing emergency condition in neonates. This condition may occur anywhere between the upper part of gastrointestinal tract to the lower gastrointestinal tract. There are several features that are highly suggestive of gastrointestinal obstruction such as polyhydramnios, feeding intolerance, bilious emesis, abdominal distention and delayed passage of meconium.<sup>4</sup>

As neonatal gastrointestinal obstruction may be caused by various etiologies at different site along the gastrointestinal tract, an algorithm for diagnostic approach is essential especially in healthcare facilities with limited imaging modalities. This approach should start with simple history taking, physical examination and plain radiograph before moving on to a more advanced imaging modality for more



sophisticated cases. An algorithm for diagnosis of neonatal intestinal obstruction is presented in **Figure 1**.

Often times, neonates presenting with gastrointestinal obstruction also present with serious life-threatening conditions such as shock, respiratory distress or electrolyte imbalance. Therefore, resuscitation is the main focus for those presenting with emergency conditions before moving on to explore the underlying etiology. Placing nasogastric tube should also be considered in most cases with gastrointestinal obstruction in order to decompress the abdominal pressure swiftly. Inability to pass the nasogastric tube is highly suggestive of esophageal atresia or web. Plain abdominal radiograph is essential in determining the approximate site of obstructions based on the air pattern seen on the image. The complete differential diagnosis algorithm is presented in **Figure 1**.

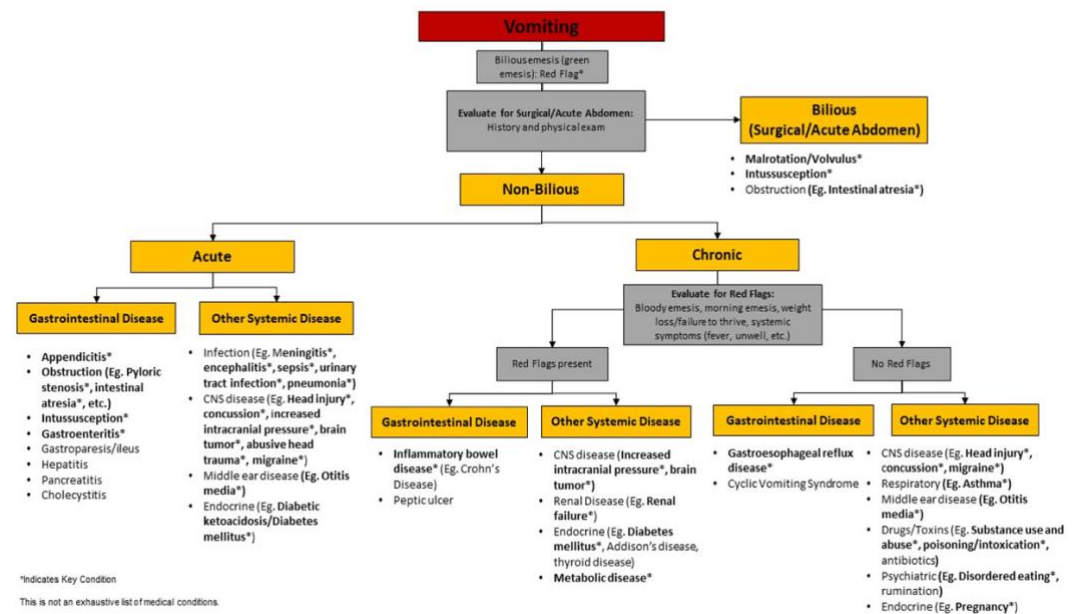


**Figure 1.** Diagnostic approach for neonates presenting with intestinal obstruction.

## Approach to Neonatal Vomiting

In most cases of neonatal gastrointestinal emergencies, patients almost always present with vomiting that may be bilious or non-bilious. Furthermore, persistent vomiting may also lead to a more severe consequences such as hypovolemic shock and

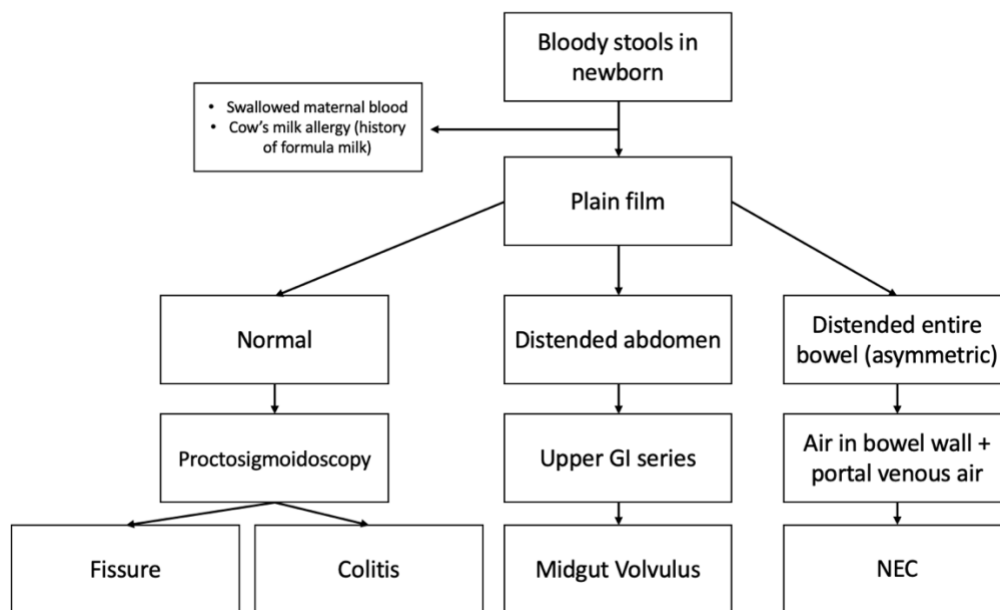
electrolyte imbalances. Therefore, clinicians are expected to address this problem early while also working to find the underlying etiologies of neonatal vomiting.<sup>5</sup> Infants who present with bilious emesis should always be evaluated for possible surgical problems particularly midgut volvulus, intussusception or any other possible cause of intestinal obstruction below the ampulla of Vater. Meanwhile, acute non-bilious emesis can be caused by gastrointestinal diseases such as intestinal obstruction above the ampulla of Vater or systemic diseases such as infection. On the other hand, chronic non-bilious vomiting should be assessed for any red flag to determine the underlying etiologies. The complete algorithm of diagnostic approach for neonatal vomiting can be seen in **Figure 2**.<sup>5</sup>



**Figure 2.** Diagnostic approach for neonates presenting with vomiting.<sup>5</sup>

## Approach to Neonatal Gastrointestinal Bleeding

Gastrointestinal bleeding is often an alarming sign that indicates a possible emergency condition in neonates. However, some non-emergency condition such as swallowed maternal blood and cow's milk allergy can also result in gastrointestinal bleeding in neonates.<sup>6</sup> Patients with gastrointestinal bleeding but with a normal abdominal radiograph should be evaluated for fissure or colitis by using proctosigmoidoscopy.<sup>6</sup> Meanwhile, if patients have distended abdomen, work up for midgut volvulus and necrotizing enterocolitis should be evaluated as depicted in **Figure 3**.



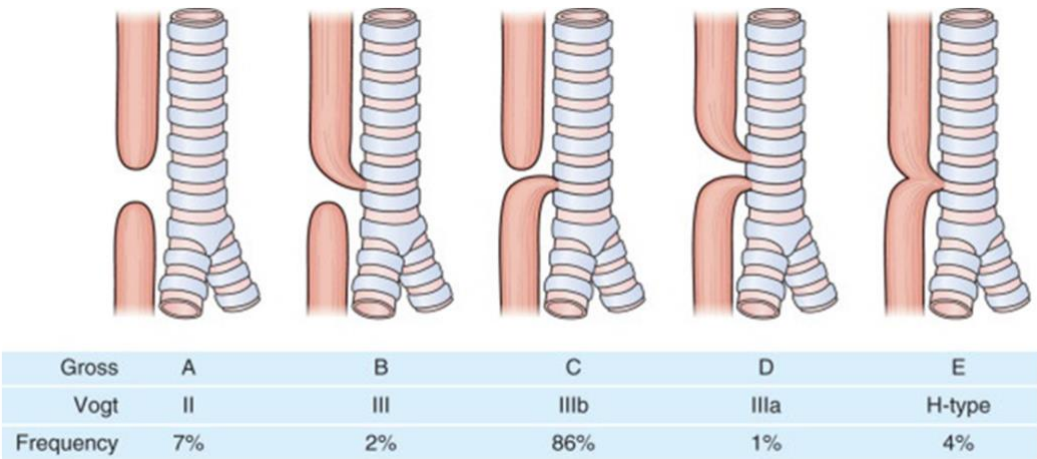
**Figure 3.** Diagnostic approach for neonates presenting with gastrointestinal bleeding.

## Upper Gastrointestinal Emergencies

### Esophageal Atresia

Esophageal atresia is a congenital anomaly of the upper gastrointestinal tract with a prevalence of 1 in 2500-4500 live births.<sup>7</sup> During early fetal development, the common foregut separates to form the trachea and esophagus. Failure of this separation or complete development of foregut tube leads to esophageal atresia with or without tracheoesophageal fistula.<sup>8</sup>

The majority of patients with esophageal atresia often present with associated anomalies, commonly known as VACTERL (vertebral defects, anal atresia, cardiac defects, tracheoesophageal fistula, renal and limb anomalies) or CHARGE (coloboma, heart defects, atresia choanae, growth retardation, genital and ear abnormalities).<sup>7</sup> Based on the anatomical configuration, esophageal atresia can be classified into several types as depicted in **Figure 4**.<sup>9</sup> Type A has a prevalence of 7%, characterized by isolated esophageal atresia without any tracheoesophageal fistula. Type B is characterized by proximal tracheoesophageal fistula and has a rather low prevalence of 2%. Type C is the most common and involves a proximal esophageal atresia with distal tracheoesophageal fistula. Type D is characterized by the presence of both proximal and distal tracheoesophageal fistula with a prevalence of 1%. Lastly, type E which is an isolated tracheoesophageal fistula with a prevalence of 4%.



**Figure 4.** Classification of esophageal atresia.<sup>9</sup>

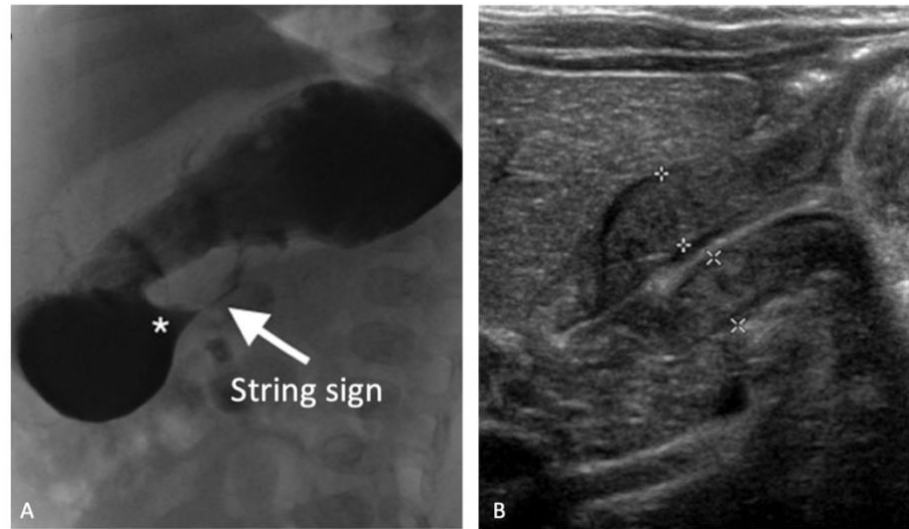
Prenatal ultrasound is often able to diagnose one-third of esophageal atresia cases which usually present with polyhydramnios and absent stomach bubble during ultrasound. However, many cases are not diagnosed before birth especially those in developing countries with limited healthcare facilities. Babies with esophageal atresia are symptomatic shortly after birth with inability to feed and excessive drooling, which may lead to choking and cyanotic episodes during feeding. Typically, esophageal atresia is diagnosed after fail attempt to pass an orogastric tube. On chest radiograph, the tube can be seen coiling above the level of esophageal atresia. Abdominal radiograph may show an absence of bowel gas in isolated esophageal atresia or esophageal atresia with proximal tracheoesophageal fistula (Gross type A and B).<sup>3</sup> However, in cases with distal tracheoesophageal fistula, bowel gas can be seen within 4 hours of life.<sup>3</sup> In addition, other modalities such as bronchoscopy is used as the gold standard to look for tracheoesophageal fistula.<sup>3</sup>

Once diagnosed with esophageal atresia, patients should be intubated to maintain airway patency as well as to prevent choking. Suction catheter should also be placed gently to eliminate excessive secretions. Patients should be given nothing per oral and nutrition should be given through parenteral route. The definitive treatment for esophageal atresia is surgical repair and should be done only after careful evaluation of other associated abnormalities. Neonates with less than 1500 grams birthweight should undergo staged approach with ligation of fistula initially before proceeding for atresia repair once the neonate is larger.<sup>10</sup> Complications are quite common after surgical repair of esophageal atresia. Early complications such as anastomosis leakage occurs in 15-20% of patients.<sup>11</sup> Meanwhile, late complications such as stricture formation occurs in 30-40% of patients.<sup>11</sup> Therefore, patients should be closely monitored to identify any complications should they arise.

### **Pyloric Atresia and Hypertrophic Pyloric Stenosis**

Pyloric atresia is a rare entity with a prevalence of 1 in 100,000 live births and only contribute to 1% of all intestinal atresia.<sup>12</sup> Up to 55% of pyloric atresia is associated with other anomalies such as epidermolysis bullosa and multiple intestinal atresia, which worsen patients' prognosis.<sup>12</sup> Prenatal ultrasound often shows polyhydramnios with dilated stomach bubble without duodenal or other intestinal dilation. Upon birth, patients may present with non-bilious vomiting, feeding intolerance and upper abdominal distention.<sup>3</sup> On plain abdominal radiograph, a single gastric bubble with a complete absence of distal (single bubble appearance) is highly suggestive of pyloric atresia.<sup>3</sup>

Hypertrophic pyloric stenosis occurs due to abnormal thickening and elongation of the pyloric sphincter musculature, obstructing the gastric outlet. The incidence of this abnormality is approximately 2-5 per 1000 live births and is more predominantly in males.<sup>13</sup> Patients with hypertrophic pyloric stenosis usually present with projectile non-bilious emesis. Upon physical examination, a firm, non-tender, hard pylorus measuring 1 to 2 cm in diameter in the right upper quadrant (resembles an 'olive') is palpable. Upper gastrointestinal contrast study was historically used to diagnose hypertrophic pyloric stenosis based on the finding of string sign (narrowed pyloric canal due to compression from the enlarged pyloric sphincter musculature) with no egress of contrast (**Figure 5**).<sup>3</sup> However, with ultrasound device is more readily available nowadays, this modality has started to replace upper gastrointestinal contrast study in diagnosing hypertrophic pyloric stenosis. Moreover, ultrasound also has the advantage that it doesn't produce any ionizing radiation and has a sensitivity that reaches almost 100%.<sup>3</sup> Based on ultrasound finding, diagnostic criteria for pyloric stenosis include single muscular wall thickness greater than or equal to 3 mm, and a pyloric length of greater than or equal to 15 mm.<sup>14</sup> Right later decubitus positioning may also help visualization in case of overdistended stomach.<sup>15</sup>



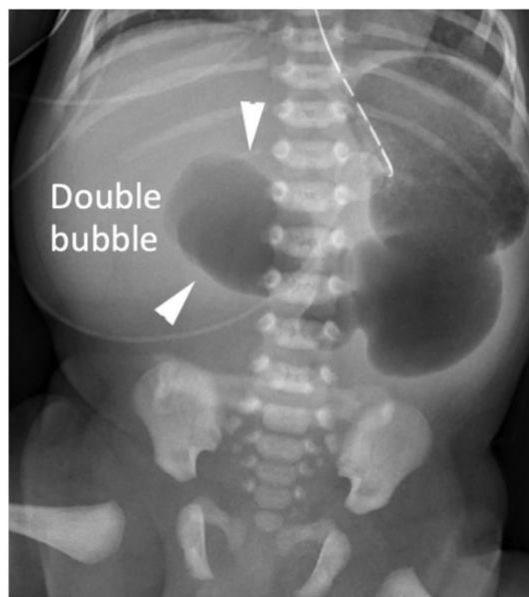
**Figure 5.** (A) Upper gastrointestinal contrast study demonstrates string sign in patient with hypertrophic pyloric stenosis. (B) Ultrasound finding: thickening of the pyloric musculature and elongation of pyloric outlet.<sup>3</sup>

The main focus for treatment of pyloric atresia/ stenosis should be rehydration and correction of any electrolyte imbalances due to excessive vomiting.<sup>16</sup> Nasogastric tube should also be placed for decompression.<sup>16</sup> Lastly, surgical procedure called pyloromyotomy can be considered after rehydration and correction of other metabolic imbalances.<sup>16</sup>

### Duodenal Atresia

Duodenal atresia occurs due to embryologic failure of recanalization, resulting in complete obstruction of the duodenum. There is also an association of this abnormality with trisomy 21 syndrome.<sup>17</sup> Polyhydramnios and double bubble appearance (dilated stomach and duodenum) during antenatal ultrasound are highly suggestive of duodenal atresia.<sup>3</sup> Upon birth and first feeding, infants may develop symptoms such as bilious or non-bilious emesis, depending on the site of obstruction distal or proximal of the ampulla Vater.<sup>3</sup> Plain abdominal radiograph is diagnostic when "double bubble" sign (gas distension of dilated stomach and duodenum and absence of distal air) is seen as shown in **Figure 6**.<sup>3</sup>





**Figure 6.** Abdominal radiograph showing "double bubble" sign in patient with duodenal atresia.<sup>3</sup>

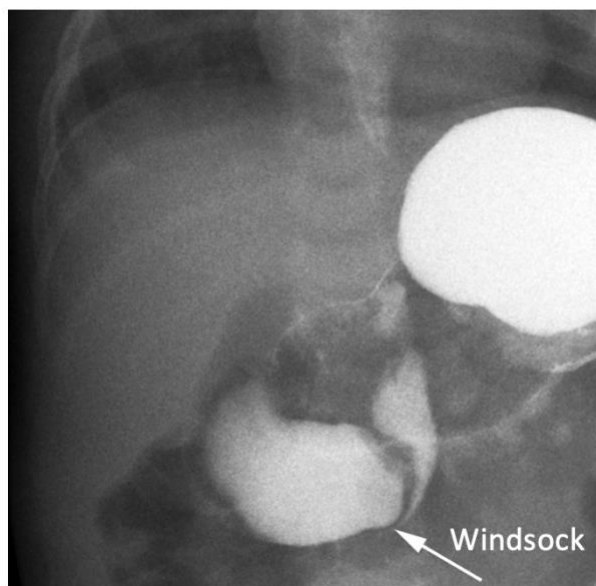
### **Duodenal Stenosis and Duodenal Web**

Duodenal stenosis occurs due to incomplete recanalization of the duodenum, resulting in persistent narrowing of duodenal lumen particularly on the second segment. Meanwhile, duodenal web refers to a condition in which a persistent membrane is partially obstructing the duodenal canal, with a predilection at the second part of the duodenum.<sup>18</sup> Hence, both duodenal stenosis and duodenal web are partial duodenal obstructions.

By the aid of the upper gastrointestinal contrast study, "windsock" deformity is pathognomonic to both duodenal stenosis and duodenal web.<sup>3</sup> "Windsock" deformity refers to a dilation of segment proximal to the stenosis and bulging of web into the non-dilated segment, as shown in **Figure 7**.<sup>3</sup> Furthermore, upper gastrointestinal contrast study is able to differentiate duodenal obstruction and midgut volvulus, which requires emergency surgery.<sup>3</sup>

Nasogastric tube should be gently placed when suspecting duodenal obstruction in order for decompression of the abdominal pressure. Furthermore, clinicians should prioritize in treating emergency conditions that may arise due to this abnormality, such as dehydration and electrolyte imbalances. The definitive treatment of duodenal atresia/stenosis/web is surgical repair (duodenoduodenostomy).





**Figure 7.** Upper gastrointestinal series showing "windsock" sign in patient with duodenal web.<sup>3</sup>

### Malrotation and Midgut Volvulus

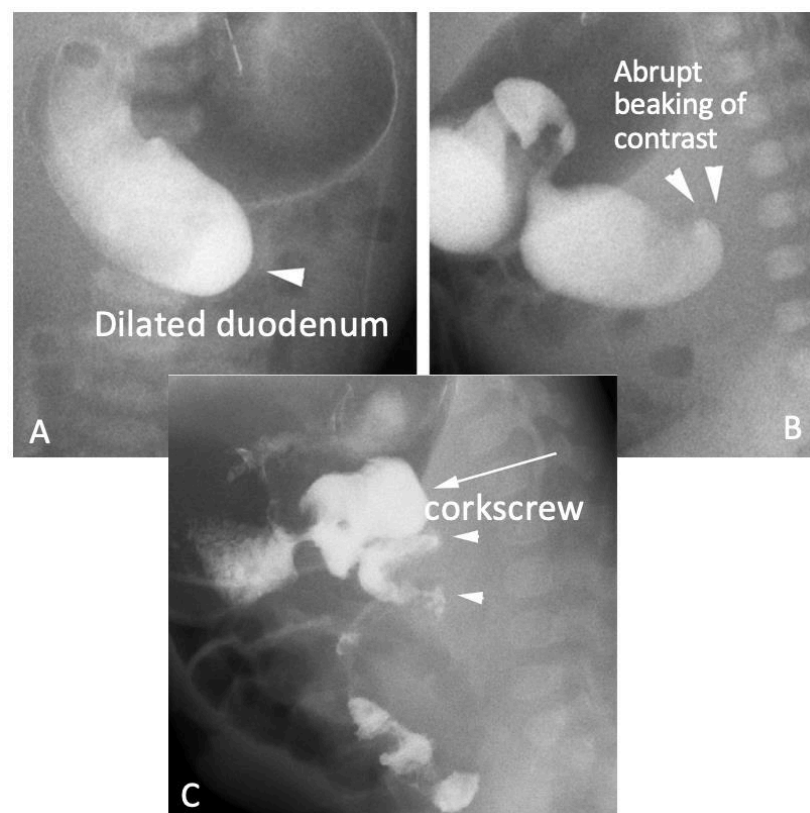
During the fourth to eight weeks of gestation, the small bowel rotates counterclockwise around the superior mesenteric artery axis. In this particular period, the bowel also protrudes through the yolk sac, elongates and rotates 90 degrees counterclockwise before retracting back into the abdominal cavity where another 180 degrees counterclockwise rotation occurs.<sup>19</sup> Malrotation occurs due to an error in this rotational process of the bowel during embryologic development, leading to abnormal fixation of the duodenojejunal junction and/or cecum within the peritoneum.<sup>19</sup> Patients with malrotation have a higher risk to develop midgut volvulus, resulting in bowel ischemia. Furthermore, in these patients, the associated Ladd's bands or peritoneal bands that attempt to fix the cecum might also cause duodenal obstruction.<sup>3</sup>

Classical symptoms of midgut volvulus are bilious emesis and abdominal distention with 75% of cases present as newborns.<sup>20</sup> However, in some cases, non-bilious emesis, hypovolemia and gastrointestinal bleeding may occur.<sup>20</sup> Over time, patients may also develop hemodynamic instability (hypovolemic shock), peritonitis (inflammation associated with volvulus) and hematochezia (bowel ischemia and necrosis).<sup>3</sup>

Plain abdominal radiograph is less useful in diagnosing midgut volvulus as "double bubble sign" produced by abdominal radiograph is not specific only for volvulus.<sup>3</sup> However, this modality may aid in demonstrating pneumoperitoneum in case of bowel perforation.<sup>3</sup> The gold standard modality to diagnose midgut volvulus is upper gastrointestinal series. However, this imaging should only be performed in hemodynamic stable patients. There are several signs that strongly suggest midgut

volvulus during upper gastrointestinal series such as abnormal position of duodenojejunal junction, dilated duodenum and corkscrew appearance of the duodenum (**Figure 8**).<sup>3</sup> On the other hand, even though ultrasound has lower sensitivity and specificity in diagnosing midgut volvulus, this device is more accessible and relatively easier to perform compared to the upper gastrointestinal series. The signs of midgut volvulus based on ultrasound are inversion of the superior mesenteric vein and the superior mesenteric artery and "whirlpool" sign due to twisted mesenteric vessels around the base of mesenteric pedicle.<sup>3</sup>

Nasogastric or orogastric tube should be placed to decompress the abdominal pressure in case of midgut volvulus.<sup>21</sup> Simultaneously, fluid resuscitation and correction of any electrolyte imbalances as well as administration of broad-spectrum antibiotics should be conducted before surgery.<sup>21</sup> Midgut volvulus is an emergency situation and need for immediate surgical intervention to reduce the volvulus.<sup>21</sup> Moreover, appendectomy is also typically performed during the procedure as the malposition of the appendix may make the diagnosis of appendicitis atypical and challenging to diagnose in the future.<sup>22</sup>



**Figure 8.** Upper gastrointestinal series of patient with midgut volvulus. (A) Dilated duodenum and abrupt cutoff of contrast from frontal view. (B) Abrupt beaking of contrast from lateral view. (C) "Corkscrew" appearance of distal duodenum.<sup>3</sup>

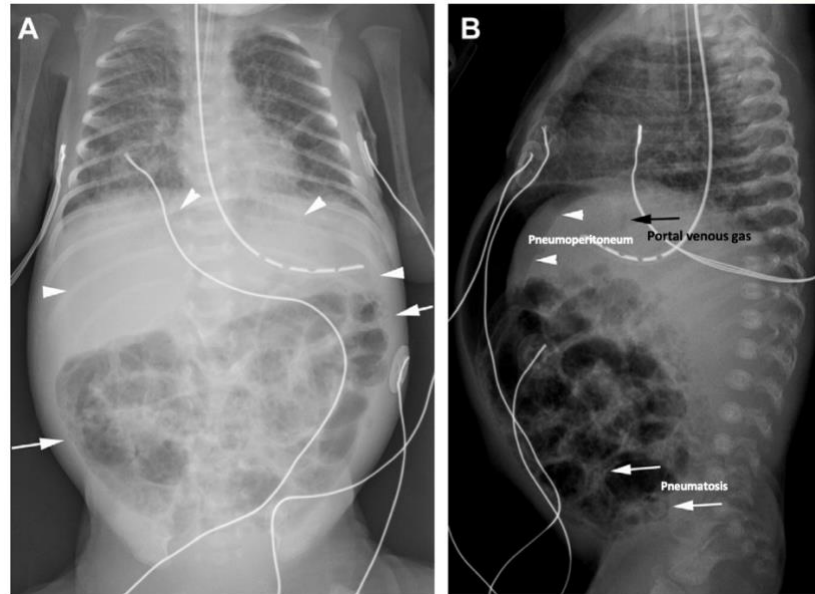
## Lower Gastrointestinal Emergencies

### Necrotizing Enterocolitis

Necrotizing enterocolitis (NEC) is the most common etiology of neonatal gastrointestinal emergencies with high mortality rate.<sup>23</sup> Primarily, this disease affects premature or low birthweight infants.<sup>23</sup> Various etiologies are known to be associated with NEC such as immature bowel function and disruption of the gut microbiota.<sup>23</sup> The pathophysiology of NEC is bacterial invasion due to inflammation of the intestinal wall that leads to necrosis of the colon and intestine and ultimately will also result in perforation and peritonitis.<sup>23</sup>

Symptoms of NEC often times are vague, variable and subtle, making clinical diagnosis become more challenging.<sup>24</sup> Patients can present with various complaints such as lethargy, mottling, bradycardia, abdominal distension, feeding intolerance and bloody stools.<sup>24</sup> In more severe cases, respiratory and circulatory failure may occur.<sup>24</sup> Therefore, imaging modalities play a crucial role in helping clinicians not only diagnosing but also detecting any complications. On abdominal radiograph, several signs can be found in case of NEC such as abnormal gas pattern, pneumatosis and portal venous gas (**Figure 9**).<sup>3</sup> In early stage, diffuse nonspecific gaseous pattern can be seen while in the later stage, fixed dilation or "persistent loop sign" can be observed, indicating an imminent bowel perforation.<sup>3</sup> Pneumatosis, which occurs as a bubbly appearance as gas enters the submucosa or subserosa layer, is a pathognomonic sign of NEC. Meanwhile, the presence portal venous gas is an indication of more severe disease that may require surgical intervention.<sup>3</sup> Lastly, pneumoperitoneum is a sign of full thickness bowel necrosis with perforation and is an absolute indication for surgery.<sup>3</sup>

The treatment modalities of NEC include stabilization of the patient's airway, breathing by intubating, fluid resuscitation if hypotension occurs, as well as nasogastric tube placement with nothing per oral.<sup>25</sup> Intravenous antibiotics should be given and should cover gram negative and anaerob bacteria. Surgical intervention or laparotomy is indicated when patients have worsening conditions despite medical treatments or patients have bowel perforation.<sup>25</sup> Recent systematic review and meta-analysis found an overall beneficial effect of probiotics in the prevention of NEC.<sup>26</sup> However, implementation in clinical practice has been difficult because of concerns about the efficacy and safety of probiotics due to variability of probiotic strains across the studies.<sup>26</sup>

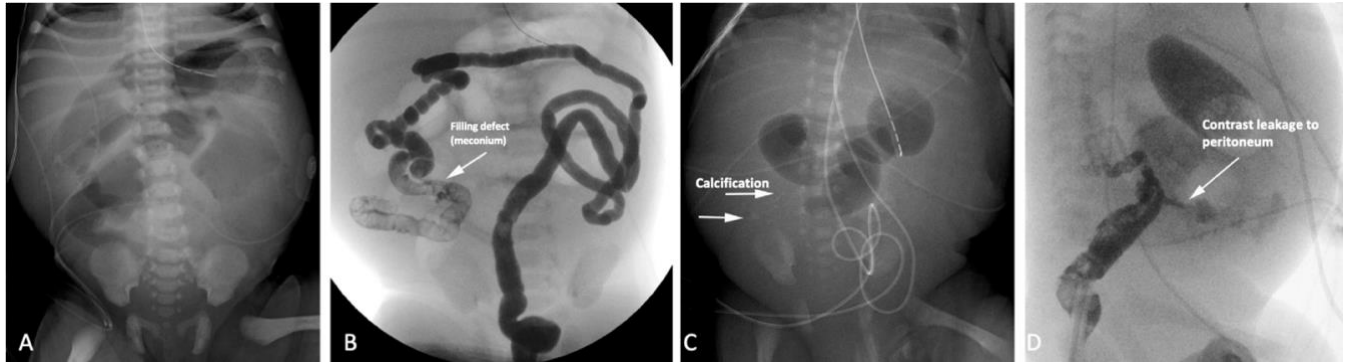


**Figure 9.** Abdominal radiograph of patient with NEC in (A) supine (B) lateral view showing multiple dilated bowel loops with pneumatosis (white arrow), pneumoperitoneum (white arrowhead) and portal venous gas (black arrow).<sup>3</sup>

### Meconium-related Disorder

Meconium ileus occurs due to lumen obstruction by thick adhesive meconium and may be an early manifestation of cystic fibrosis in neonates.<sup>27</sup> In simple meconium ileus, thick meconium obstructs the terminal ileum causing the small intestine proximal to the obstruction site to dilate and fill with meconium, gas and fluid.<sup>27</sup> On the other hand, complex meconium occurs when the meconium-distended segment develops into volvulus, necrosis, ischemia or even perforation and spillage of meconium to the peritoneum, leading to meconium peritonitis.<sup>27</sup>

Infants usually present with intestinal obstruction with bilious emesis and abdominal distension within hours of birth and first feeding.<sup>27</sup> If meconium peritonitis has occurred, patients may present with abdominal tenderness, fever and even shock.<sup>27</sup> On plain abdominal radiograph, multiple dilated bowel loops with "soap bubble" appearance (meconium mixed with swallowed air) can be observed (Figure 10).<sup>3</sup> Typical obstruction sign such as air-fluid level sign is not usually seen in case of meconium ileus due to the thick consistency of meconium. Water-soluble contrast enema with hyperosmolar agents may be diagnostic as well as therapeutic and may show multiple filling defects consistent with meconium (**Figure 10**).<sup>3</sup> Meanwhile in meconium peritonitis, abdominal radiograph will show diffuse calcification throughout the peritoneal cavity and pseudocyst formation of the persistent spillage of meconium, which are highly predictive of the need for surgical intervention.<sup>3</sup>



**Figure 10.** (A) Abdominal radiograph of patient with meconium ileus showing multiple dilated bowel loops and "soap bubble" appearance. (B) Water-soluble contrast radiograph demonstrating multiple filling defects, consistent with meconium. (C) Abdominal radiograph of patient with meconium peritonitis showing multiple scattered calcifications on the abdomen. (D) Water-soluble contrast radiograph demonstrating contrast leakage to the peritoneum, suggesting a perforation.<sup>3</sup>

### Hirschsprung's Disease

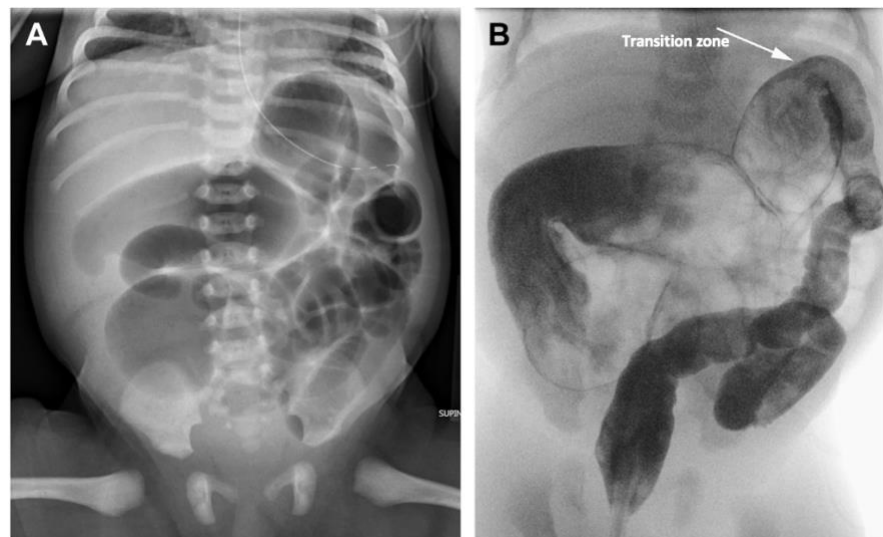
Hirschsprung's Disease is once considered deadly with a prevalence of 1 in every 5000 live births and predominantly occurs in male infants.<sup>28</sup> This disease is characterized by the absence of ganglion cells at both the Meissner and Auerbach plexus in the terminal rectum and may extend proximally to a variable distance.<sup>29</sup> Hirschsprung's disease occurs due to a sudden arrest of migration and differentiation process of the neural crest cells at the enteric nervous system, leading to the absence of parasympathetic plexuses and overactivity of the intestine with persistent acetylcholine release.<sup>29</sup> As the consequence, continuous contraction causes narrowing of the colon while dilation occurs on the healthy proximal colon.

Hirschsprung's disease can be classified into 4 different types based on the length of the affected colon.<sup>3</sup> First is the ultrashort segment which only involves up to 4 cm of the distal rectum and can be easily missed by rectal suction biopsy if placed too deep.<sup>3</sup> Second is the short-segment Hirschsprung's disease which is the most common form with the aganglionic section extending to the mid-sigmoid colon.<sup>3</sup> On the other hand, long-segment Hirschsprung's disease usually affects colon segment proximal to the mid-sigmoid without involving the whole colon. Last is the total colonic aganglionosis if the aganglionic segment affects the whole part of the colon.<sup>3</sup>

The diagnosis of Hirschsprung's disease involves the combination of clinical signs and symptoms as well as radiological findings and histopathological evaluation of the biopsied sample. Infants with this disease typically present with polyhydramnios, bilious vomiting, failure to pass meconium in the first 48 hours and abdominal distention. Plain abdominal radiographs in case of Hirschsprung's disease shows multiple dilated bowel loops which is consistent with the description of distal bowel



obstructions. By using water-soluble contrast enema, a transition zone and altered rectosigmoid ratio can be observed as cone-shaped area of transition between aganglionic narrow segment and distended bowel segment proximally (**Figure 11**).<sup>3</sup> Furthermore, irregular contraction and mucosal irregularity is pathognomonic signs of Hirschsprung's disease but only present in 20% of cases.<sup>3</sup> Gold standard for diagnosing Hirschsprung's disease is rectal suction biopsy which shows the absence of ganglion cells in myenteric and submucosal plexus as well as hypertrophy of nerve fibers in the aganglionic colon segment.<sup>3</sup> The International Gastroenterology Committee recommends at least 2 biopsies including submucosal and mucosa layer with a minimum diameter of 3 mm.<sup>30</sup> Biopsies should also be performed at least 2 cm above dentine line and should be well oriented.<sup>30</sup> Furthermore, acetylcholinesterase staining can also demonstrate the increased parasympathetic activity of the nerve fibers.<sup>30</sup>



**Figure 11.** (A) Plain abdominal radiograph showing multiple dilated bowel loop without gas in the rectum. (B) Water-soluble contrast enema demonstrating transition zone and multiple filling defects.

Hirschsprung's disease usually requires staged reconstruction with an initial placement of temporary decompressive colostomy. Definitive pull-through surgery is typically performed four to six months after the colostomy placement. Rectal irrigation before the surgery is highly recommended to reduce the dilated colon size as well as to prevent the devastating complication of enterocolitis.<sup>31</sup> In case of Hirschsprung associated enterocolitis, administration of broad-spectrum antibiotics, fluid and electrolytes as well as rectal irrigation should be performed.<sup>32</sup>



## Conclusion

Given the critical time window and the vulnerability of the neonate population, the proper identification and prompt treatment of neonatal gastrointestinal emergencies is crucial to minimize morbidity and mortality. Multidisciplinary management with neonatologists, pediatric surgeons, radiologists, and nursing staff working closely together can provide the best possible outcomes.

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## Conflict of Interest

None declared.

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