

Original article

Anterior displacement of anus: a common association with constipation in infancy

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Background: Anterior displacement of the anus (ADA), a common congenital abnormality of anorectal region, has been recognized as a common cause of constipation. However, the more recent studies have failed to demonstrate an association between the anterior displacement and constipation self.

Objective: Investigate the association between ADA and constipation.

Methods: Four hundred and three neonates have been examined and their anal position index (API) has been determined. API less than 0.46 and 0.34 in males and females, respectively, is defined as ADA. A 6-month prospective cohort study has been conducted to evaluate the association between ADA and constipation.

Results: Overall incidence of ADA is 24.8%, while the incidence is significantly higher in females than males (32.0% and 17.7%, respectively; $p < 0.01$). Compared to the first child, later birth order, especially the second child, trends to have an increased risk of ADA (Odd ratio=1.75; 95%CI=1.06-2.90). Average maternal age of ADA is higher than control (28.2 and 26.6 years, respectively; $p=0.02$). Overall incidence of constipation is 10.1% without any difference between ADA group and control (12.0 and 9.5%, respectively). Nevertheless, at six months incidence of constipation is higher in ADA than in control (6.7% and 0.8%, respectively; $p < 0.01$). The incidences of ADA in infants with constipated events at 2, 4 and 6 months of age are 10.0%, 33.3%, and 71.4%, respectively.

Conclusion: ADA is a common finding with reported incidence of one-quarter of neonates. Female genre, later birth order and higher maternal age are risk factors of ADA. Children with ADA have an increased tendency toward constipation corresponding with increasing age. API measurement during pediatric examination is recommended to achieve a complete evaluation of infancy constipation.

Keywords: Anal position, anterior displacement, anus, constipation, infant

Anterior displacement of the anus (ADA) is recognized as a common congenital developmental abnormality of anorectal region [1]. This abnormality is easily diagnosed by simple physical examination and measurement at perianal and anorectal region. In order to define the normal position of anus, Reisner et al. [1] proposes a quantitative measurement using the anal position index (API), the ratio of anal-fourchette distance to coccyx-fourchette distance for females and the ratio of anal-scrotum distance to coccyx-scrotum distance for males. API less than 0.46 in boys

and less than 0.34 in girls is indicative of anterior displacement of anus. ADA may be a common cause of constipation [2-4]. Clinical presentation of ADA is usually consistent with early onset of constipation since birth or at the time of weaning from breast-feeding [3]. According to the study of Leape and Romenofsky [3], ADA is accounted in more than one third of patients examined in consultation for chronic constipation. However, the more recent cross-sectional studies fail to demonstrate a direct association between ADA and constipation [5, 6]. To date, one well-designed prospective cohort study to clarify the relationship between constipation and anal position has not been reported yet. The aim of the present study is to investigate the association of anterior displacement of the anus with constipation during infancy.

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Materials and methods

Four hundred and three normal neonates born at Srinakharinwirot University Hospital between November 2003 and September 2004 were enrolled into the study. We excluded from the study all congenital malformations or syndromes easy to notify, such as Down syndrome, anencephaly or congenital heart diseases. The presence of abnormalities in genitourinary area and lower extremities, such as ambiguous genitalia, imperforate anus, ectopic anus, and achondroplasia, was also included in the exclusion criterion. However, occult abnormalities that looked normal on physical examination and needed further investigations for diagnosis like Hirschsprung's disease or congenital spine deformity were not considered as exclusion criteria.

At birth, demographic data, clinical and laboratory data including parental ages, birth order, gestational age, birth weight, length, head circumference, anus fourchette (scrotum) distance, coccyx-fourchette (scrotum) distance and thyroid function screening tests were collected. To measure all anatomic distances we used the same method previously reported [7].

Briefly, the newborn was held in the lying position with hips and knees flexed. An adhesive tape was positioned along the long axis of coccyx-fourchette or coccyx-scrotum. The positions of coccyx, anus, and posterior fourchette or scrotum were marked and the distances between them were measured on a plain surface as shown in **Fig. 1**. Anal position index (API) was calculated by the ratio of anus-scrotum distance to coccyx-scrotum distance and anus-posterior fourchette distance to coccyx-posterior fourchette distance for boys and girls, respectively. API less than 0.46 in boys and less than 0.34 in girls were defined as anterior displacement of the anus (ADA) [1]. All neonates were prospectively followed at child health clinic at the ages of two, four, and six months for bowel habit patterns and feeding practice. During these follow up visits, information about bowel habit patterns including the frequency of bowel movements, stool consistency, and feeding practice were collected by parental interviews. Infants who had history of bowel movements less than three times per week or defecated of any large hard stools were considered as constipated children [8, 9].

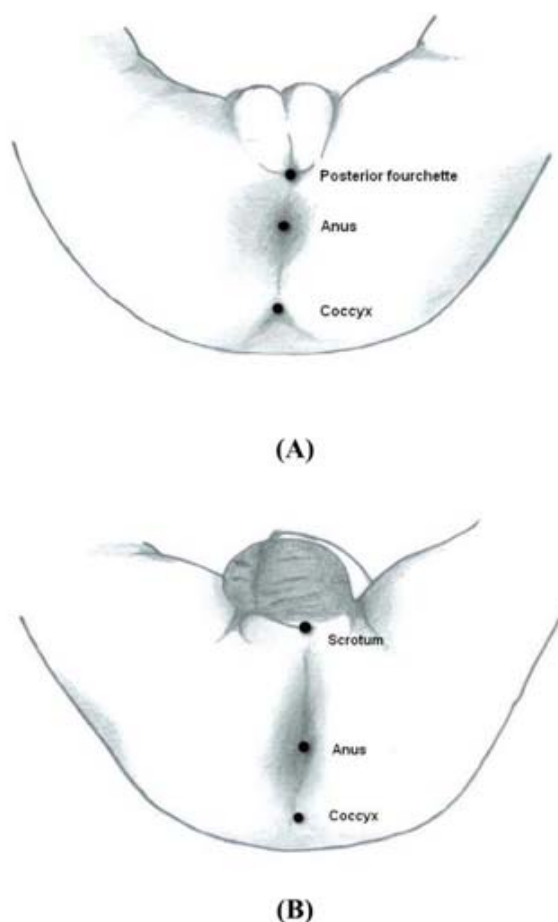


Fig. 1 Measurement of anal position index. (A) the ratio of anal-fourchette distance to coccyx-fourchette distance for females and (B) the ratio of anal-scrotum distance to coccyx-scrotum distance for males.

To evaluate the association between ADA and constipation, infants who were missing during the follow-up period, consumed solid food before four months of age, or had biochemical evidence of hypothyroidism resulting from national screening program were excluded from further analysis.

The study was approved by the Ethics Committee of Faculty of Medicine, Srinakharinwirot University and informed consent was obtained from the parents of each infant.

Statistical analysis

Demographic data, anal-fourchette (scrotum) distance, coccyx-fourchette (scrotum) distance, and anal position index were expressed as mean and standard deviation (SD). Data were analyzed using the Chi-square or Fisher exact test. Continuous

variables were compared using a Student's t-test. Statistical analyses were performed using a SPSS 11.5 software package. Differences were considered as significant if p-value <0.05.

Results

Four hundred three neonates were included into this study, 203 (50.4%) were males and 200 (49.6%) females. The mean anal position index (API) in male neonates was 0.51 (SD=0.07) whereas in female neonates was 0.38 (SD=0.08) (p <0.001). Based on API measurements, anterior displacement of the anus (ADA) was diagnosed in 100 neonates of 403 (24.8%). Demographic characteristics and anthropometric data of neonates with ADA or normal API were detailed in **Table 1**.

Table 1. Demographic characteristics and anthropometric data of 403 neonates.

	All (n= 403)	ADA* (n= 100)	Normal API [†] (n= 303)	P-value
Gender, n (%)				0.001
Males	203	36 (17.7)	167 (82.3)	
Female	200	64 (32.0)	136 (68.0)	
Gestational age week (SD)	38.3 (1.8)	38.2 (1.9)	38.3 (1.8)	0.55
Birth weight, g (SD)	2926.8 (458.5)	2966.0 (421.6)	2913.8 (470.0)	0.32
Length, cm (SD)	51.4 (2.9)	51.4 (2.3)	51.4 (3.1)	0.84
Head circumference cm (SD)	32.9 (1.6)	32.9 (1.6)	32.8 (1.6)	0.60
Anus-fourchette (scrotum) distance cm (SD)	1.8 (0.7)	1.3 (0.5)	2.0 (0.7)	<0.001
Coccyx-fourchette (scrotum) distance cm (SD)	3.9 (1.0)	3.7 (0.9)	4.0 (1.0)	<0.001
Anal position index (SD)	0.44 (0.1)	0.34 (0.06)	0.48 (0.08)	<0.001
Males	0.51 (0.08)	0.40 (0.05)	0.53 (0.05)	<0.001
Females	0.39 (0.08)	0.31 (0.03)	0.42 (0.06)	<0.001
Time at the first meconium passing hour (SD)	5.1 (6.0)	5.1 (6.0)	5.1 (6.0)	0.99
Paternal age year (SD)	30.8 (7.3)	31.6 (7.5)	30.6 (7.2)	0.22
Maternal age year (SD)	27.0 (6.1)	28.2 (6.0)	26.6 (6.2)	0.02
Birth order, n (%)				Odd ratio (95%CI)
1	200	39 (19.5)	161 (80.5)	1
2	141	42 (29.8)	99 (70.2)	1.75 (1.06 to 2.90)
3	36	11 (30.6)	25 (69.4)	1.82 (0.82 to 4.01)
4	18	7 (38.9)	11 (61.1)	2.63 (0.96 to 7.21)

*ADA= anterior displacement of the anus, [†]API= anal position index, CI=confidence interval.

The incidence of ADA was significantly higher in females than males, 32.0% and 17.7% ($p < 0.01$), respectively. There were no significant differences in gestational age, birth weight, length, head circumference, paternal age, and time of meconium passage between neonates with ADA or normal API. Considering the birth order, second child had a significantly higher risk of ADA than the first one (odd ratio (OR) = 1.75; 95% confidence interval 1.06 to 2.90). The third and fourth child trended to have more risk of ADA than the first, but the difference didn't reach a statistic significance (OR of the third child = 1.82; 95% CI 0.82 to 4.01 and OR of the fourth child = 2.63; 95% CI 0.96 to 7.21, respectively). Neonates with ADA had significantly higher maternal age than normal anal position infants (28.2 years and 26.6 years, respectively: $p = 0.02$).

During the six-months prospective cohort study, 87 (21.6%) infants were excluded from further analysis because of loss to follow-up. None infant consumed solid food before four months of age or any biochemical evidence of hypothyroidism was observed at the time of study. Among 316 infants that completed a six-month cohort study, 75 (23.7%) infants had anterior displacement of the anus and 156 (49%) were males. Thirty-two (10.1%) infants reported constipated events during the 6-months cohort study. Female infants had a significantly higher incidence of constipation than male infants (14.4% and 6.0%, respectively; $p = 0.01$). The incidence of constipation was 12.0% and 9.5% in ADA group and normal anal position group, respectively. Then, we analyzed the presence of constipation events at 2, 4, and 6 months of age, and summarized the results in **Table 2**. The estimated incidences of ADA in infants showing constipation were 10.0% (2 of 20), 33.3% (3 of 9), and 71.4% (5 of 7) at 2, 4 and 6 months of age, respectively.

Discussion

Abnormal delays difficulty in defecation less than three times per week, or hard painful defecation are clear symptoms of constipation [8, 10]. This problem, encountered by both primary and specialty medical personals, has a prevalence ranging from 0.3% to 28% [11, 12]. In particular, constipation prevalence is influenced by age and gender. The overall incidence rate of male to female is 1:1. During infancy and childhood, constipation is more frequent in boys than girls with the ratio of 3:1. After puberty, the incidence

is inverted to 1:3 [13]. Few data about constipation in young infants are available. For instance, Rudolph et al. [13] reported a prevalence of 16% in children 22 months old. The present results demonstrated that the incidence of constipation young infants is 10%. Female gender has a greater risk of constipation compared to males, with the ratio of 2.4:1.

The etiology of constipation in childhood may be multifactorial [14]. In most children, constipation is functional, without objective evidence of pathological condition. Otherwise, the congenital abnormality of anus position is considered a possible cause of constipation, especially in infancy and early childhood [1-3, 15, 16]. In previous studies, the diagnosis of ADA is usually relied on inspection; therefore, the incidences widely vary, according to physician experience [2, 3]. Reisner et al. [1] firstly has reported a simple and reliable method to determine the normal position of the anus; this parameter has been called anogenital (anal position) index by Bar-Moar and Eiton [15].

From previous studies, the overall incidence of ADA in children aged one month to 10 years old and admitted in the hospital with unrelated disorders is 33% and the incidence in girls is nearly double of the incidence in boys (43.4% and 24.6%, respectively) [5]. Moreover, Leape and Ramenosky [3] reported that ADA accounted for more than one third of pediatric patients seen in the consultation for chronic constipation. We also showed that in neonates, ADA is common with an incidence of approximately 25%. Unlike other anorectal malformations and Hirschsprung's disease which have higher incidence rate in males, ADA occurs more often in females than in males with a ratio of approximately 2:1. Moreover, the presence of co-existing abnormalities has not been observed in ADA patients, as in anorectal malformations and Hirschsprung disease [17, 18]. The results of the present study show a significant increase in ADA incidence in second birth order compared to the first order child. Furthermore, incidences of ADA trend to increase in the third and the fourth birth order child, without reaching statistical significance. This might be explained by the small sample size in these groups. Based on the present study, the risk factors of ADA presentation are female gender, higher maternal age and later birth order.

The association between anterior displacement of the anal opening and constipation is still controversial [2-6, 15]. In a large case series of 134 patients with

Table 2. Details of 32 constipated infants.

Anal position	Sex	At 2 months	At 4 months	At 6 months	Gestational of age age(wk)	Time at the first meconium passing(hr)	Birth weight (g)	Birth length (cm)	Head circumference (cm)	anus forchette distance (cm)	coccyx forchette distance (cm)	Anal position index	Paternal age (yr)	Maternal age (yr)	Birth order
ADA*	1 female	normal	normal	constipation	37.40	6.00	2700	47.00	30.00	1.10	3.30	.33	24	20	1
	2 female	constipation	normal	normal	37.00	1.00	2500	49.00	31.00	1.00	3.90	.26	18	17	1
	3 female	normal	normal	constipation	38.00	7.50	2700	51.00	32.50	1.10	3.60	.30	27	27	1
	4 male	normal	constipation	normal	38.60	2.00	2600	47.00	32.50	2.50	5.70	.44	24	24	1
	5 male	constipation	constipation	normal	38.00	29.00	3250	53.00	35.00	2.00	5.20	.38	31	24	1
	6 female	normal	normal	constipation	39.60	11.00	3500	55.00	36.00	1.10	3.50	.31	38	35	2
	7 female	normal	normal	constipation	39.00	7.00	2800	50.00	30.00	.80	2.40	.33	29	25	2
	8 female	normal	normal	constipation	40.00	7.50	3650	51.00	35.00	.90	3.60	.25	32	35	4
	9 male	normal	constipation	normal	38.00	14.00	3200	54.00	34.50	1.80	4.50	.40	40	35	2
Normal API†	1 female	constipation	normal	normal	38.60	6.00	2550	49.00	31.00	1.60	3.80	.42	23	23	1
	2 female	constipation	constipation	normal	40.00	24.00	3100	56.00	34.00	1.00	2.50	.40	33	29	2
	3 female	normal	normal	constipation	38.00	3.00	2450	51.00	31.00	1.50	3.20	.47	26	36	1
	4 female	constipation	normal	normal	37.00	1.00	2700	54.00	32.00	1.70	3.50	.49	39	19	1
	5 female	constipation	normal	normal	38.10	24.00	2950	52.00	32.00	1.20	3.10	.39	27	25	1
	6 female	constipation	normal	normal	37.20	12.00	2750	50.00	34.00	1.20	3.10	.39	38	31	1
	7 female	normal	constipation	normal	38.40	2.00	2750	52.00	33.00	1.20	2.90	.41	20	18	2
	8 female	constipation	normal	normal	39.00	2.00	3800	49.00	31.00	1.50	3.70	.41	25	26	1
	9 female	normal	constipation	normal	38.60	6.00	2800	48.00	31.00	1.20	2.20	.55	36	36	1
	10 female	constipation	normal	normal	37.90	1.00	2950	53.00	34.00	1.30	3.10	.42	20	17	1
	11 female	constipation	normal	normal	38.00	6.00	2550	56.00	33.00	1.00	2.90	.34	27	26	1
	12 female	constipation	normal	normal	37.00	13.00	2850	50.00	31.50	1.30	3.30	.39	26	27	2
	13 female	constipation	normal	normal	38.00	11.00	3600	51.00	36.00	1.20	3.20	.38	25	22	1
	14 female	constipation	constipation	normal	37.00	12.00	2400	52.00	32.50	.90	2.10	.43	24	22	1
	15 female	constipation	normal	normal	35.00	2.00	1450	48.00	28.00	1.10	3.10	.35	25	20	1
	16 male	constipation	normal	normal	40.90	8.00	3050	54.00	33.00	2.30	4.90	.47	28	22	2
	17 male	constipation	normal	normal	39.00	6.00o	2850	51.00	34.00	2.80	4.60	.61	23	33	1
	18 male	constipation	normal	normal	37.00	5.00	3600	53.00	33.50	2.30	4.80	.48	38	38	5
	19 female	constipation	normal	normal	41.10	10.00	3200	50.00	33.50	1.50	3.10	.48	23	20	1
	20 male	constipation	normal	normal	40.00	6.00	3000	52.00	34.00	2.30	4.70	.49	29	19	2
	21 female	normal	constipation	normal	38.00	1.00	3050	53.00	34.00	1.50	3.20	.47	42	41	2
	22 male	constipation	normal	normal	40.40	1.00	3780	54.00	34.50	2.40	4.40	.55	21	17	1
	23 male	normal	constipation	constipation	39.00	6.00	3400	65.00	33.00	2.80	4.70	.60	40	37	3

*ADA= anterior displacement of the anus; †API= anal position index

ADA, nearly all patients have constipation from birth or from the time of weaning from breastfeeding and require surgical correction [2]. This association has been further supported by Leape and Romenofsky and Upadhyaya [3, 4]. However, studies that are more recent do not describe any association between anal position and constipation [5, 6, 15]. Using cross-sectional study, Bar-Moaz and Eitan do not show significant difference in API of healthy children and children with idiopathic constipation [15]. Two different reports from healthy children [6] and from children admitted to the hospital with unrelated disorders [5], show that the incidence of constipation is not different between children with normal anal position and those with ADA. However, both studies have employed a cross-sectional methodology. Presently, there are no studies that prospectively follow-up infants with or without ADA over long periods, in particular since the birth, in order to clarify the natural history of constipation. The six-months prospectively follow-up presented in this study shows that the overall incidence of constipation in healthy infants and in infants with ADA is not significantly different (9.5% and 12.0%, respectively). This is in accordance with previous study [5]. However, the incidence of constipation in normal anal position group is usually higher in the first two months of age and declines thereafter. At six months of age, almost all infants with normal anal position are free from constipation. In contrast, the incidence of constipation in infants with ADA increases and is significantly different in comparison to the normal group at 6 months of age. Physiologic change and the learning process of defecation may be postulated as the cause of this difference. Beyond the neonatal period, the most common cause of constipation is functional constipation, without objective evidence of a pathological condition [8]. After birth, the normal physiologic changes of intestines and colon are in process and lead to normal frequency of bowel movement pattern at different age. During the first week of life, neonates have a mean of four stools per day [8]. This frequency gradually declines to a mean average of 2.0-2.9 stools per day at three months of age and 1.8 stools per day at one year of age [8]. The learning process of defecation may also contribute to decrease constipation incidence in normal infants. The classic history of straining and crying prior to defecation in infants aged between 1-10 weeks is mostly explained by the incapability to increase intra-abdominal pressure and relax the pelvic floor muscles

in a coordinated fashion as is required to allow the easy passage of stools [13]. This condition will be resolved in the first few months of life by learning process. No treatment or manipulations are required. However, this problem may confuse parents with true constipation. [13].

Apart from functional constipation, it is important to determine whether the constipation is a consequence of organic causes. Even if only a minority of children has organic or anatomic causes for constipation, most of them are present in neonates [19]. This study showed that constipated events in infants with ADA usually arise at four to six months, thus corresponds to the time of weaning onto solid food, as reported previously [3]. Considering the immature digestive process in infants, undigested solid foods may predispose to constipation. Indeed, the incidence of ADA in constipated infants increase during this prospective cohort study from 10% at two months of age to approximately 30% and 70% at four and six months of age, respectively. It might be reasonable to suggest a thorough physical examination including external examination of the perineum and perianal area in order to calculate anal position index and exhaustively evaluate constipation in infants who present symptoms at six months of age.

Symptomatic therapy with laxative administration is usually adequate in mild cases [3]. On the other hand, posterior anoplasty with or without sphincterotomy to remove the deranged lower loop of the external sphincter is recommended as a procedure of choice in severe cases [2-4]. However, the surgical operation is generally not indicated in the majority of infancy constipation associated with ADA. A prominent posterior rectal shelf and an anal dimple posterior to the anus are the most common characteristics reported in patients requiring surgical treatment [2-4, 16, 20, 21].

Conclusion

Anterior displacement of the anus is a common congenital abnormality of the anorectal region in neonates with the incidence of one-quarter of the healthy neonates. Female gender, later birth order, and higher maternal age are the risk factors of ADA presentation. Although the overall incidences of constipation are comparable among children with or without ADA, children with ADA tend toward constipation with increasing age. Based on the present results, anal position index (API) is a predictive parameter for evaluation of constipation in infants.

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