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The position of the anal dimple in newborns and infants with anorectal malformations and its correlation with the normal anal position☆,☆☆

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ABSTRACT

Background/purpose: The anal position index (API) defines the normal anal position as the ratio of fourchette–anal distance to fourchette–coccyx distance for females and the scrotum–anal distance to scrotum-coccyx distance for males. In this study, measurement of the API in newborns and infants with anorectal malformations (ARM), using the center of the midline anal dimple (AD) to represent the center of the proposed neoanus, was performed to assess whether or not the AD was located in a significantly abnormal position as correlated with the normal anal position.

Methods: The cases comprised 65 consecutive newborn and infants with ARM, divided into 2 age-based groups (Group A: 1st day to 1 month; Group B: 1–12 months), without sacral or significant perineal abnormalities. Controls included an equal number of age and gender matched patients admitted for other conditions. The characteristics of the AD ('well developed' or 'moderately developed') as well as those of the midline perineal raphé in males and the perineal groove in females were also recorded.

Results: Combining both age groups, the mean API ± SD was 0.41 ± 0.012 in male cases and 0.53 ± 0.07 in male controls (p value 0.003). The corresponding values for female cases and controls were 0.31 ± 0.09 and 0.36 ± 0.07 respectively ($p = 0.040$). In male newborns, the API in cases was significantly lower than the API in controls ($p < 0.001$). Combining both males and female newborns, the differences between the API in cases and controls was also statistically significant ($p < 0.001$). In older boys as well as in girls from both age groups, apart from cases of persistent cloaca, the API was lower in cases than in controls, especially in boys, although the difference was not statistically significant. On comparing the API in individual male ARM with that in male controls, cases of rectobulbar urethral fistula (RBUF) and rectovesical fistula had a statistically significant lower API ($p < 0.001$). In the 4 cases of persistent cloaca, the mean API (0.40 ± 0.1) was higher than the mean API in female controls and the AD was well developed with a well-delineated, narrow perineal groove. The majority (53/65; 81.53%) of patients had a 'well developed' AD. Twelve patients (18.47%) had a 'moderately developed' AD including 10 females and 2 males. There were 15 patients with a depressed AD (anal fossette); of these 12 (80%) were females. All boys had a well delineated perineal raphé in contrast to 10 girls (29.4% of total female ARM) who had a poorly delineated midline perineal groove.

Conclusions: During definitive surgery for ARM, if the AD is taken as the site of the proposed neoanus, the neoanal position is likely to be anterior to the normal anal position in both males and females and especially so in males. Overall, girls with ARM appear to be more likely to have a relatively poorly developed and/ or depressed AD and a poorly delineated perineal groove.

Type of study: Clinical Research.

Level of evidence: Level III.

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After reports of a correlation between slight anterior displacement of the anus and chronic constipation [1,2], Reisner et al. [3] described the anal position index (API) as a quantitative measurement to define the normal anal position by measuring the ratio of fourchette-anal distance to fourchette–coccyx distance for females and of scrotum–anal distance to scrotum-coccyx distance for males. In 200 newborns, the mean API was 0.44 and 0.53 in girls and boys respectively (SD = 0.05), and in 30 infants, 4–18 month old, the mean API was 0.40 (SD = 0.06) in

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girls and 0.56 (SD = 0.04) in boys [3]. Bar-Moar and Eitan [4] described a similar study, terming the result as the anogenital index. The mean anogenital index in normal girls was 0.39 (SD +/− 0.09) while in boys, it was 0.56 (SD +/− 0.2). Subsequently, there have been several reports on measurements of the API in newborns and infants [5–11].

In children with anorectal malformations (ARM), the perineum shows an 'anal dimple' (AD) which is an area of usually raised, hyperpigmented skin in the midline of the perineum [12]. This is usually a cutaneous areola with a surrounding halo of pigmentation and puckering [12] but although the AD is frequently everted, its epicenter may sometimes be depressed (anal fossette) [12]. The AD is usually well-delineated even in patients with 'low' ARM who have a perineal fistula frequently located anterior to the dimple [12]. According to Peña and Levitt [13], the vertical fibers of the striated muscle complex (SMC) are located within the limits of the AD and extend from the skin to the levator ani muscle. The AD represents the center of the external sphincter muscles i.e. the SMC [14–16] and during definitive surgical repair, it's anterior and posterior ends should define the anterior and posterior limits of the new anus [13].

To date, no study describing an objective measurement of the API in children with ARM, using the center of the AD to represent the center of the proposed neoanal position, has been reported in the literature. The present study was undertaken to assess whether or not the AD was located in a significantly abnormal position along the midline of the perineum as correlated with the API in normal newborns and infants. The characteristics of the AD and the midline perineal raphé or groove in the various types of ARM were also studied.

1. Material and methods

The material for this prospective, observational study comprised 65 consecutive patients with ARM, between the ages of 1st day of life to 1 year, managed from December 2014 to August 2016 at our institution. The patients were categorized into two age-based groups: Group A, patients of age 1st day of life to 1 month, and Group B, patients between the ages of 1 month to 1 year. An equal number of male and female patients in each age-group, admitted for other conditions and without any significant congenital malformation, were taken as controls.

For the purposes of this study, the midline (sagittal) AD was defined as the area of hyperpigmented skin along the line of the midline in the perineum [12,13]. Patients with partial or complete sacral agenesis, scrotal and/ or severe perineal malformations, and those in whom the AD was poorly developed or poorly delineated were excluded from the study.

Institutional Ethics Committee approval was obtained. A written, informed parental consent was obtained before enrolling a case or control for the study. In newborns and infants, controls and cases, the sex and the age at the time of participation in the study was recorded. Patients included as controls underwent clinical evaluation and a study of the patient's record to rule out any congenital malformation involving major organ systems. In each case of ARM, the relevant clinical findings, results of radiologic investigations including plain X-rays of the lumbosacral spine to rule out sacral abnormalities, and the final diagnosis were also recorded.

1.1. Measurement of the anal position index (API) in normal controls [1,3,5]

The child was held in a genopectoral position and a transparent adhesive scotch tape was placed on the longitudinal axis of the midperineum. The inferior border of the coccyx, the middle of the anus, and the posterior margin of the fourchette or first fold of the scrotum (the line from the beginning of the transverse rugae; scrotal-perineal junction) was marked on the tape (Fig. 1). The distances were measured with the tape placed on a table, using a caliper and graduated scale. The measurements were double checked every time.

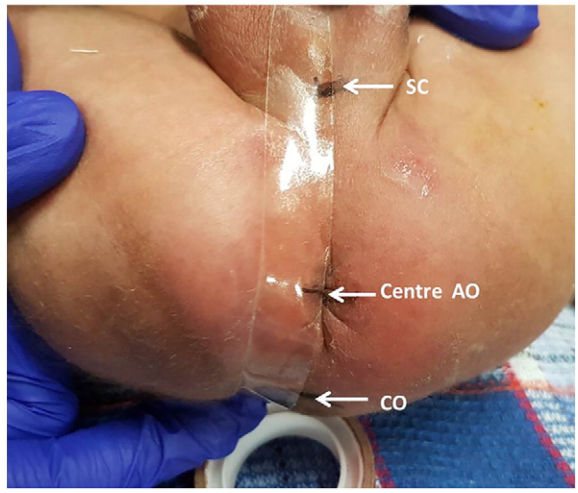

Fig. 1. Male control, 6 months old, markings for measurement of API. SC = Scrotum; AO = Anal Opening; CO = Coccyx.

The API was calculated as follows:

$$\text{API} = \text{Posterior fourchette (scroto)—anal distance (in cm)} / \text{Posterior fourchette (scroto)—coccygeal distance (in cm)}.$$

1.2. Measurement of the API in newborns and infants with ARM

On a subjective basis, the AD was categorized as 'well developed' or 'moderately developed'. The characteristics of the midline raphé of the perineum in male cases and of the sagittally placed midline groove – the perineal groove – [12] in female cases were also recorded. Measurement of the API was performed in a similar manner to that described for controls, using the center of the AD to represent the center of the proposed neoanus. The anterior and posterior limits of the AD were marked and the midpoint of this distance was taken to represent its center (Figs. 2 and 3).

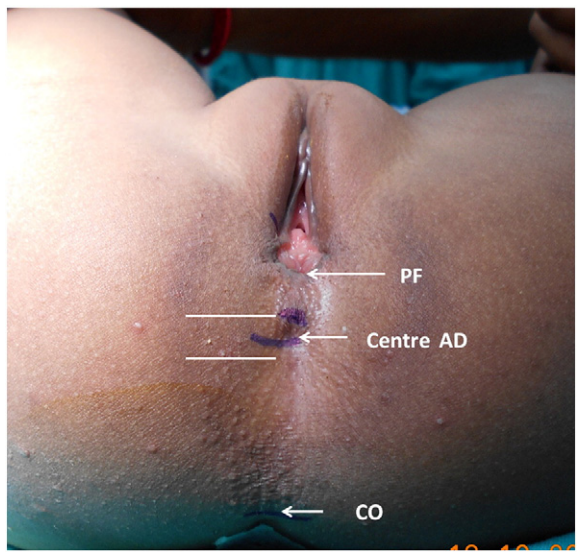

Fig. 2. Girl with large VF displacing fourchette margin posteriorly. The AD is moderately developed and depressed with a poorly delineated, wide perineal groove; API 0.18. VF = Vestibular fistula; PF = Posterior Fourchette. Horizontal lines mark limits of AD.

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