Original Article

Evaluation of Anal Position Indexes from Anal Dimple and Neoanus at Sphincter Muscle Complex in Children with High Anorectal Malformations in Kano, Nigeria

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Abstract

Background: Anorectal malformations (ARMs) comprise a spectrum of congenital anomalies that continue to present a challenge to patients, caregivers and paediatric surgeons. The main objective of this study was to determine the anal position index (API) from anal dimple and neoanus at the sphincter muscle complex in children with high ARM. **Methods:** Using a cross-sectional study design, the API was assessed from anal dimple and neoanus at the sphincter muscle complex in children with high ARM. All eligible children with high ARM who presented to AKTH during the study period were recruited into the study. **Results:** Forty eligible patients with an age range of 8 months to 108 months were recruited. Twenty-five were males (62.5%) with a male: female ratio of 1.7:1. The mean pre-operative API using anal dimple was 0.47 ± 0.05, while that of the post-operative API using neoanus was 0.53 ± 0.04. There was a statistically significant difference between the two indices ($t = -11.8$, $P < 0.01$). There was also a statistically significant difference amongst male and female indices $P < 0.05$, but there were no significant differences between different age groups. **Conclusion:** This study has shown that there was a statistically significant difference between the pre-operative API and post-operative API; thus, the position of the anal dimple does not significantly predict the central position of the sphincter muscle complex.

Keywords: Anal dimple, anal position index, high anorectal malformations, neoanus

Introduction

Anorectal malformations (ARMs) comprise a wide spectrum of diseases, which can affect both boys and girls and involve the distal anus and rectum as well as the urinary and genital tracts. Defects range from the very minor and easily treated with an excellent functional prognosis, to those that are complex, difficult to manage, are often associated with other anomalies and have a poor functional prognosis.[1]

ARMs occur commonly throughout the African continent. Even under the best circumstances, children undergoing operative treatment for ARMs may have life-long faecal continence issues amenable to bowel management.[2] ARMs comprise a spectrum of congenital anomalies that continue to present a challenge for paediatric surgeons. Worldwide, it is estimated that ARMs occur at the rate of 1:4000 live births; the anomalies ranging in severity from mild anterior displacement of the anus to very complex malformations of the hindgut and urogenital tract.[3]

Advances in modern surgical techniques and neonatal care have greatly improved survival amongst ARM patients over the last decades, and early mortality is now unusual in the

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absence of associated cardiac or chromosomal anomalies. Posterior sagittal anorectoplasty (PSARP), first introduced in 1982 by De Vries and Pena and followed later by its limited modified anterior sagittal anorectoplasty, represents the basis of the modern surgical approach to ARMs with termination of the anal canal outside the voluntary sphincter complex.[3] Other significant developments in the management of ARMs include the centralisation of care to tertiary centres, increasing understanding of their pathologic anatomy and improved treatment of major functional complications.[3]

PSARP has become the preferred technique for surgical management of ARMs. The procedure involves an incision from the coccyx to the perineal body, to widely expose the external sphincter, the levator ani muscles, the rectum and the distal fistula to facilitate repair.[4] In ARMs, it is a fact that the anal opening is either absent or present but in an abnormal position. The position of the anal dimple can be identified by the fact that this area will pucker in response to pinprick in those patients who have an obvious dimple,[5] and that the anal dimple is also hyperpigmented. Anal position index (API), which is an index used to determine the normal position of the anus is measured as the ratio of scroto-anal distance divided by sacrococcygeal distance in males and fourchette-anal distance divided by fourchette-coccygeal distance in females. It is a valuable tool in the determination and detection of normal anal position.[6] The normal API values have been documented to be 0.56 ± 0.04 for boys and 0.40 ± 0.06 for girls.[7]

In the treatment of patients born with ARMs, the ultimate goal has moved from mere survival to alleviating symptoms and improving quality of life after surgical reconstruction.[8] Despite the technical advances in the surgical repair of ARMs that have occurred over recent years, complications that require a secondary procedure are still common.[9]

The patient's best chance for a good functional result is achieved when the proper operation is performed during the first definitive procedure and complications are avoided. This is especially true in those patients born with a defect that has a good prognosis.[9] The introduction of PSARP has significantly reduced the number of patients requiring reoperation for complications such as faecal incontinence over the years. This procedure provides superior exposure of the sphincter muscle complex and prevents the complete mislocation of the rectum that was seen with other techniques. However, in the tropics, despite the availability of skilled workforce to manage such patients, the non-availability or near absence of such facilities as electrical muscle stimulators and sophisticated ultrasound scans, which aid in the exact localisation of the sphincter muscle complex and identification of the centre, leaves many paediatric surgeons with careful tactile stimulation and visual localisation to give a rough guide to the site of the neoanus.[10]

This study was designed to determine whether the position of the anal dimple will exactly predict the centre of the sphincter muscle complex by measuring the API preoperatively using the anal dimple, determining the centre of the muscle complex by intraoperative muscle stimulation through which neoanus was constructed and measuring post-operative API using the neoanus.

Methods

The study was conducted at Aminu Kano Teaching Hospital, which is a tertiary health institution situated in the ancient city of Kano, North-Western Nigeria. The sample size was determined using the sample size formula for two related groups or measurements,[11,12] which was found to be 36, and adding 10% attrition rate in case of default and loss to follow-up was approximated to 40. All eligible patients of age range between 8 months and 108 months with a diagnosis of high ARMs who presented to paediatric surgical outpatient clinic during the period of this study and who met the inclusion criteria were recruited until the desired sample size was attained.

For each patient, a detailed history and clinical examination were performed, to rule out other associated anomalies. The perineal examination was done looking for evidence of the anus or its dimple, and in females, additional, emphasis on a number of openings in the perineum and their locations. Other systemic examinations of the cardiovascular, musculoskeletal and central nervous systems were conducted to rule out associated congenital anomalies.

Following confirmation of diagnosis and level of the anomaly and after 3–6 months of colostomy, definitive treatment was offered depending on the level of the malformation.

Pre-operative preparation of the patients

Preoperatively, each patient had investigations done to ensure fitness for surgery (complete blood count, electrolyte, urea and creatinine), blood was grouped and cross-matched for the surgery. A distal loopogram was done to show the position of rectal stump before the surgery and also to outline fistula to the urogenital tract if present.

Pre-operative determination of anal position index

Preoperatively, the anal dimple was identified. An anal dimple was considered as the centre from which the measurement was taken. Patients were placed in a prone position with the buttocks elevated. The three reference points for measurement: the tip of the coccyx, the middle point of the anal dimple and the base of the scrotum for males, and the apex of the posterior fourchette in females were determined and marked. A transparent adhesive tape was applied to the perineum over these markings. Measurements were taken from the base of the scrotum to the marked centre of the dimple as scroto-anal dimple distance and from the base of the scrotum to the coccyx as sacrococcygeal distance in males as well as from the posterior fourchette to the marked centre of the dimple as fourchette-anal dimple distance and from posterior fourchette to coccyx as fourchette-coccygeal distance in females using a tape measure. The ratios of scroto-anal to sacrococcygeal distance in males and the ratios of fourchette-anal to fourchette-coccygeal distance in females were determined as the pre-operative API for both males and females, respectively.

Intraoperative determination of the central position of the sphincter muscle complex

Intraoperatively, the choice of anaesthesia was general anaesthesia with endotracheal intubation with no muscle relaxation. Following successful induction of anaesthesia, patients were placed in a prone jackknife position and the buttocks were slightly elevated. An electrode nerve stimulator (stimuplex® HNS 12) was improvised to achieve muscle stimulation. A skin incision was made in the perineum along the midline from the coccyx posteriorly, extending anteriorly to the perineal body. This was done to ensure a wide exposure. The incision was deepened down with meticulous haemostasis to the muscle layers. Using the stimulator, the muscles were stimulated and their point of maximum contraction was identified and tagged. The posterior and the anterior borders were also held in stay sutures. The neoanus was centred on the tagged central point.

Post-operative determination of anal position index

Postoperatively, on the 14th day after the construction of the neoanus following the resolution of tissue oedema, the measurements were repeated using the neoanus as the centre. The patient was placed in a prone position with the buttocks elevated; the API was determined again for each patient as performed preoperatively.

Ethical approval

Ethical approval was obtained from the Aminu Kano Teaching Hospital Health Research Ethics Committee. A consent form was administered to the respondents after the purpose of the research was explained to them. A written informed consent was sought from all participating caregivers. The Helsinki Declaration for research with human subjects was respected throughout the study. The respondents reserved the right to withdraw from the study for whatever reasons without any penalty. All information gathered from the respondents was kept confidential.

Data collection and management

During data collection, biodata was obtained: age, sex, tribe, address, informant and other aspects of history as specified in the pro forma. All API measurements were obtained using tape rule measure and marker. Data obtained were collected into a pro forma prepared for the study. Office editing was done, whereby data were checked manually for errors, and coding of pro forma was done before data entry. Data analysis was conducted using IBM-compatible computer software IBM Corp. (IBM SPSS Statistics for Windows, Version 21.0. Armonk, New York, USA). Quantitative data were summarised using mean, standard deviation, range and median as appropriate. The paired *t*-test was used to compare means. In all tests of associations, statistical significance was set at $P < 0.05$ for statistical analysis. Results were presented in tables.

Results

A total of 40 children with high ARMs were recruited for the study over the period from August 2020 to July 2021.

The median age of the children was 48 months (range: 8–108 months) and 62.5% (n = 25) were males. The male-to-female ratio was 1.7:1. Majority of patients were from Kano, 18 (45.0%) out of 40. The rest were from the neighbouring states of Katsina, Jigawa, Bauchi and Yobe.

Table 1: Socio-demographic characteristics of the respondents

Socio-demographic feature	Frequency (%)
Sex distribution	
Male	25 (62.5)
Female	15 (37.5)
Total	40 (100.0)
Geographic distribution	
Kano	18 (45.0)
Jigawa	6 (15.0)
Katsina	8 (20.0)
Bauchi	6 (15.0)
Yobe	2 (5.0)
Total	40 (100.0)
Distribution of parents by religion	
Islam	38 (95.0)
Christianity	2 (5.0)
Total	40 (100.0)
Ethnic distribution of mothers	
Hausa	27 (67.5)
Fulani	11 (27.5)
Yoruba	2 (5.0)
Total	40 (100.0)
Type of marriage of the parents	
Consanguineous	16 (40.0)
Non-consanguineous	24 (60.0)
Total	40 (100.0)
Distribution of mothers by occupation	
Housewives	18 (45.0)
Farmers	10 (25.0)
Civil servants	4 (10.0)
Traders	8 (20.0)
Total	40 (100.0)
Associated anomalies in children	
Present*	6 (15.0)
Absent	34 (85.0)
Total	40 (100.0)
Clinical features at presentation	
Anaemia	26 (65.5)
Normal	10 (25.5)
Underweight	4 (10.0)
Total	40 (100.0)

*The associated anomalies were ventricular septal defects (2), polydactyly (2), syndactyly (1) and radial hemimelia (1)

Table 2: Anal position index parameters of the patients

Parameter	Mean±SD	Range
Pre-operative API	0.47±0.05	0.36–0.58
Post-operative API	0.53±0.04	0.45–0.59

API: Anal position index, SD: Standard deviation

Table 3: Gender- and age-specific anal position index parameters

	Parameter	Mean±SD	Range	*t*	*P*
Gender					
Male	Pre-operative API	0.48±0.04	0.41–0.54	−10.6	<0.01
	Post-operative API	0.54±0.03	0.49–0.59		
Female	Pre-operative API	0.45±0.07	0.36–0.58	−6.1	<0.01
	Post-operative API	0.52±0.05	0.45–0.59		
Male versus females	Pre-operative API–Post-operative API			*F*=2.53	0.023
Age (months)					
8–36	Pre-operative API	0.47±0.07	0.36–0.58	−8.4	<0.01
	Post-operative API	0.53±0.05	0.45–0.59		
37–72	Pre-operative API	0.46±0.03	0.41–0.50	−7.1	<0.03
	Post-operative API	0.53±0.03	0.49–0.57		
73–108	Pre-operative API	0.49±0.01	0.49–0.51	−4.5	<0.02
	Post-operative API	0.56±0.03	0.53–0.58		
Between groups				*F*=0.96	0.48

API: Anal position index, SD: Standard deviation

Majority of the mothers were Muslims 38 (95.0%) and the predominant tribe was Hausa 27 (67.5%). The type of marriage amongst the parents was found to be consanguineous in up to 16 (40.0%) of the parents. Most of the mothers were full-time housewives 18 (45.0%); others were farmers, traders and a few civil servants. Only 6 (15.0%) of the patients were found to have other associated anomalies, these were musculoskeletal (four) and cardiovascular (two) anomalies. Anaemia was the most common pre-operative derangement amongst the patients, 26 (65.0%), as shown in Table 1.

Anal position index

The mean pre-operative API amongst the patients was 0.47 ± 0.05 (range 0.36–0.58) [Table 2]. The mean post-operative API using the neoanus was 0.53 ± 0.04 (range 0.45–0.59) [Table 2]. The overall mean difference between the indices was − 0.067 ± 0.036 and $t = -11.8$, $P < 0.01$.

The gender-specific values for pre-operative and post-operative APIs for males were 0.48 ± 0.04 (range 0.41–0.54) preoperatively and 0.54 ± 0.03 (range 0.49–0.59) postoperatively, $P < 0.01$ and for females were 0.45 ± 0.07 (range 0.36–0.58) preoperatively and 0.52 ± 0.05 (range 0.45–0.59) postoperatively, $P < 0.01$. The API values were also computed by age groups and compared, and the findings were summarised [Table 3].

Discussion

API is an index used in determining the exact position of the anus. The API defines the normal position of the anus.[6,8] Determination of the normal position of the anus has gained increasing clinical importance since the reports of an association between abnormal anal position and constipation were demonstrated. API was first determined by Reisner *et al*.[13] as 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, to define the normal position of the anus in the newborn. They found the mean API to be 0.40 ± 0.06 for girls and 0.56 ± 0.04 for boys, and they suggested that API of <0.46 in boys and <0.34 in girls was indicative of the anterior displacement of the anus.[14]

The median age of the children demonstrated in this study was 48 months (range 8–108), this was not comparable to that mostly reported in the literature. The difference may be explained by the fact that the age range in this study was 8 months to 108 months, while in most of the other literature, it was amongst neonates and infants. The sex distribution of males 62.5% and females 37.5% was similar to that reported by Lukong *et al*.[15] in Zaria. This similarity may be due to the similarity of the problem and the common geographical locations of these patients. However, Archibong and Idika[16] in Calabar, Southern Nigeria reported a contrasting male-to-female ratio with slight female preponderance.

The current study found that the mean pre-operative API using anal dimple was 0.47 ± 0.05, and the post-operative value using neoanus was 0.53 ± 0.04. The pre-operative value differs from what Patel *et al*.[17] reported in the determination of API from anal dimple in children with ARM and compared to control; they reported mean API values of 0.41 ± 0.012 and 0.31 ± 0.09 for male and female cases, respectively. This difference may be explained by the difference in methodology, while in this study, we used tape rule and adhesive tape for measurements, they used direct measurement with rulers. Another possibility may be due to the geographical and racial differences. The post-operative values demonstrated in this study, however, were almost similar to what was reported by Patel *et al*.[17] in their controls with the value of 0.53 ± 0.07. An important finding in this study was that of the statistically significant difference between the pre-operative API using anal dimple and the post-operative API using neoanus after the determination of the centre of the sphincter muscle complex using muscle stimulation intraoperatively. The demonstrated mean post-operative value of API was almost exactly to what Patel *et al*.[17] found in the controls of their study (0.53 ± 0.04 vs. 0.53 ± 0.07) portray the significant difference between the indices shown in this study and buttress their conclusion that

siting neoanus using an anal dimple position will result in the anterior anus.

The gender-specific API values of both males and females showed statistically significant difference between the pre-operative and post-operative measurements with statistically significant difference between the genders. These values were not similar to what was reported by Davari and Hosseinpour[18] however, they also showed statistically significant difference between genders. The findings were also supported by that of [15] who found statistically significant difference between genders, although he used normal children. The observed difference was also corroborated by Reisner *et al*.,[13] Ertürk and Kandemir *et al*.[14] and Núñez-Ramos *et al*.[19] who all reported statistically significant difference between the sexes.

Another important finding of this study was that there was a statistically significant difference between pre-operative and post-operative anal position indexes within the age groups but no statistically significant difference in these values between the groups. These findings were supported by the findings of Davari and Hosseinpour.[18] who also reported no statistically significant difference amongst age groups. With respect to the research hypothesis of whether the position of the anal dimple will predict the centre of the sphincter muscle complex, the findings of this study showed a significant difference between pre-operative API using anal dimple and post-operative API using neoanus implying that the position of anal dimple did not correspond to the centre of the muscle complex. This finding may be supported by the embryology of development of these structures from different origins as well as that in patients with perineal fistula or poorly developed perineum, there may be an absent anal dimple or, if present, poorly defined as reported in the literature.[3,8,17] In accordance with the present results, previous studies have also demonstrated that during definitive surgery, if the anal dimple is taken as the site of the proposed neoanus, the neonatal position is likely to be anterior to the normal in both sexes[16] also that API may have utility in preventing complications associated with incorrect anal placement (incontinence, constipation) post-anorectoplasty.[14,19]

The generalisability of the findings in this study may be limited by the nature of the study as a hospital-based study with 40 patients; this is an important issue for future research consideration.

Conclusion

This study demonstrated that there was a statistically significant difference between the anal position indexes measured preoperatively using anal dimple and postoperatively using neoanus in children with high ARMs. Thus, concluding that the position of the anal dimple does not significantly determine the central position of the sphincter muscle complex.

Recommendations

With the above findings of this study, the following recommendations could be made: the anal dimple should not be used as a guide for siting neoanus in children with ARM without determining the centre of the sphincter muscle complex as this will result in mislocated neoanus leading to complications. Surgeons should endeavour to always determine the centre of muscle complex intraoperatively before siting neoanus during anorectoplasties.

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Conflicts of interest

There are no conflicts of interest.

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