Original Article

Malformations: A 5-Year Review of the Presentation and Management in a Teaching Hospital in Ghana

Abiboye C. Yifieyeh, Babatunde M. Duduyemi[1], Anthony Enimil[2], Michael Amoah, Boateng Nimako

Department of Surgery, Komfo Anokye Teaching Hospital, Departments of [1]Pathology and [2]Child Health, Kwame Nkrumah University of Science and Technology, Kumasi, Ghana

Abstract

Background: Anorectal malformations (ARMs) are congenital defects affecting the distal gastrointestinal tract and anus with frequent fistulous connections to the genitourinary system. The spectrum of the disease is considerably wide, and thus, an individualised approach to its management is required. There are few recent publications about the burden of this malformation in Africa and the outcome of surgical intervention. We present our experiences with the management of ARM, peculiar challenges and the outcome at a tertiary hospital catering for the Northern and Central parts of Ghana. **Materials and Methods:** This was a retrospective folder review of children with ARM at the Paediatric Surgical Unit of Komfo Anokye Teaching Hospital, Kumasi, Ghana, from 2011 to 2015. The data extracted included diagnosis, associated features, initial and definitive surgical treatment and post-operative complications. **Results:** Of the 53 records that were conclusively retrieved, the sex ratio was 1:1. The median age of presentation was 4.5 days (neonates), 4.5 months (infants) and 1.6 years (older children). There were 12 (44%) rectoperineal and 18 (69%) rectovestibular fistulas, representing the most common types of ARM in boys and girls, respectively. Posterior sagittal anorectoplasty and abdominoperineal pull-through were the corrective procedures performed. The overall complication rate was <10%. **Conclusion:** Our patients generally presented later than their Western counterparts, making a single-stage correction rare. The types of ARM documented in our study suggest the existence of a geographical variation.

Keywords: Abdominoperineal pull-through, anorectal malformation, congenital, posterior sagittal anorectoplasty

Introduction

Anorectal malformations (ARMs) are congenital defects of the rectum and anus frequently having fistulous connections with the genitourinary system.[1] Most patients are born without a patent anus and pass meconium from fistulas in the perineum, urethra and vagina. The spectrum of the disease is considerably wide with various classification methods in use.[1,2] ARM is referred to as either high or low with respect to the rectal tube blindly ending either above or below the levator ani complex.[2] Syndromic presentation of the condition is known.[1] Alberto Pena's algorithm for the management of ARM is a useful tool in individualising treatment for these patients.[1] The algorithm ends with the patient (presumably a newborn) either having a colostomy or a definite corrective procedure before he/she is 48 h old. Posterior sagittal anorectoplasty (PSARP) is the corrective procedure of choice performed by many surgeons.[3] Prophylactic anal dilatation lasting for 6 months is routine. We present our experience with the management of patients with ARM who presented at our centre (Komfo Anokye Teaching Hospital, Kumasi, Ghana) over a 5-year period.

Access this article online

Quick Response Code:

Website: www.afrjpaedsurg.org

DOI: 10.4103/ajps.AJPS_10_18

Materials and Methods

The folders of patients in whom a diagnosis of ARM was made between January 2011 and December 2015 were retrieved from the records department of Komfo Anokye Teaching Hospital, Kumasi. The biodata, specific anatomic diagnosis, accompanying anomalies, preliminary and definitive surgical procedures and documented complications were extracted. SPSS version 23.0 (SPSS Inc Chicago, IL, USA) was used for data analysis and the findings are presented in graphs.

Results

Fifty-three patients' records were conclusively retrieved for the study period. The median age of presentation was 4.5 days,

Address for correspondence: Dr. Babatunde M. Duduyemi, Department of Pathology, Kwame Nkrumah University of Science and Technology, Kumasi, Ghana.
E-mail: tundeduduyemi@gmail.com

How to cite this article: Yifieyeh AC, Duduyemi BM, Enimil A, Amoah M, Nimako B. Malformations: A 5-year review of the presentation and management in a Teaching Hospital in Ghana. Afr J Paediatr Surg 2018;15:118-20.

4.5 months and 1.6 years for neonates, infants and older children, respectively [Figure 1]. Although the male-to-female ratio was 1:1, 18 (69.23%) of those that presented during infancy were male and 18 (66.7%) of those presenting later were female. There were 12 (44%) rectoperineal and 18 (69%) rectovestibular fistulas, representing the most common types of ARM in boys and girls, respectively [Figure 2]. Thirty-eight patients (72%) had low malformations and females (24) were in the majority [Figures 3 and 4]. ARM without a fistula was found in five (9.43%) patients, and the majority were male. Seven (13.20%) patients had associated uterine, limb and penile anomalies. Polydactyly was documented in five of our patients. Divided sigmoid colostomies were performed as first-stage procedures for patients with high malformations and those with rectovestibular fistulas. PSARP and abdominoperineal pull-through (APT) were the corrective procedures performed. The latter was for those with rectobladder neck fistulas. Post-operative complications occurred in eight (15%) patients. Anal stenosis, urethral injury and surgical site infection were the documented post-operative complications [Figure 5].

Discussion

ARMs have been shown to be common even in Africa.[4,5] Most reports have documented near equal incidence in sex, a finding similar to ours.[4,5] A few series from Nigeria have shown a male preponderance.[6,7] Late presentation, particularly among females, is also well known in developing countries.[8,9] The most common reasons identified for lateness in presentation include poor neonatal services at birth, poverty and poor social support. Two-thirds of all those presenting to us after infancy were female. Most of these girls had rectovestibular fistulas, which are low lesions, known to be wide enough for adequate bowel emptying at birth.

Low ARMs predominated in our patients. The low: high malformation ratio of 2.5:1 was generally higher than reported in literature.[4,5] However, Archibong *et al*. and Adejuyigbe *et al*. reporting from different regions in Nigeria both found that high malformations were more common.[6,10] This may suggest geographical variations in the disease. Most of the high malformations were found in boys (86.7%) and were rectourethral fistulas. Our finding of two cases of rectovaginal fistulas in females confirms the existence of this condition in our environment.[1] Of the five boys who presented without fistulas, three had phenotypic characteristics of Down's syndrome. No chromosomal tests had, however, been carried out to confirm this diagnosis at the time of writing. ARM without fistula is more common in children with Down's syndrome.[11]

Associated anomalies in other systems occur in 26%–50% of reported cases.[6,12] Genitourinary anomalies are among the

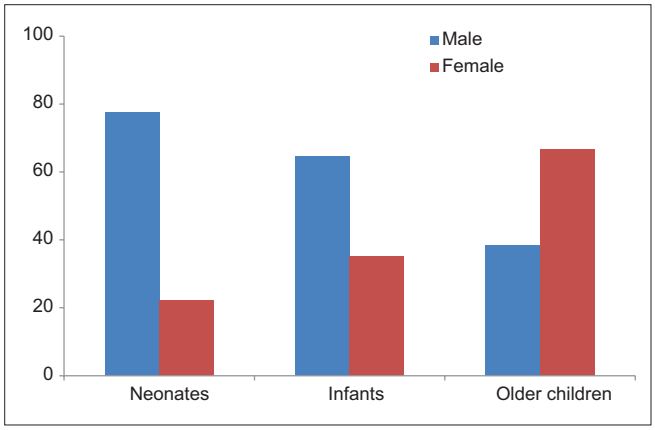

Figure 1: Distribution of our patients according to age group and gender

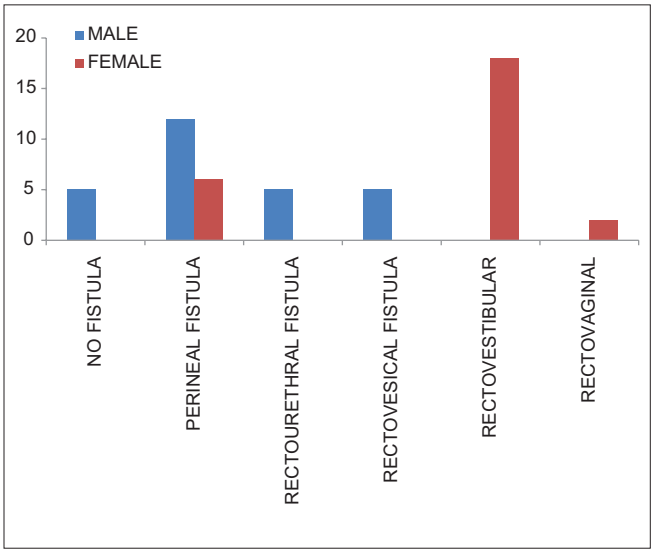

Figure 2: Gender distribution of the types of anorectal malformations

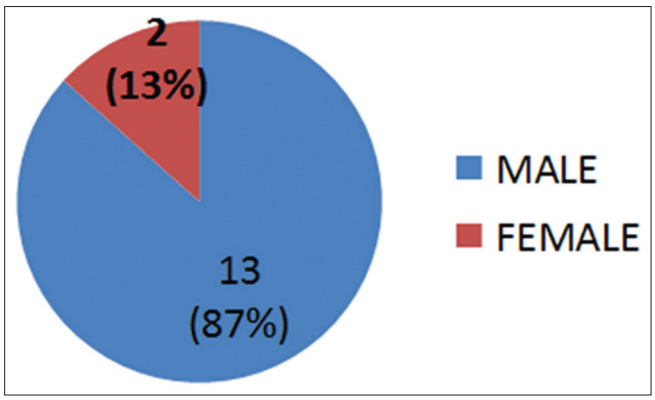

Figure 3: Gender distribution of our patients with high lesions

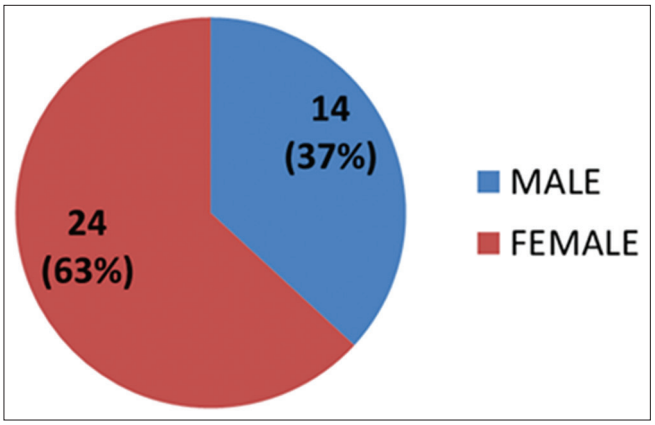

Figure 4: Gender distribution of our patients with low lesions

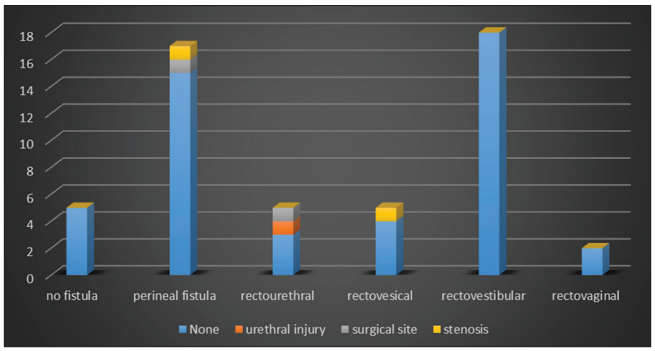

Figure 5: Frequency and types of complications encountered in our patients

most frequently encountered.[13] The VACTERL (Vertebral, Anorectal, Cardiac, Tracheoesophageal, Renal and Limb) group of anomalies is a well-documented association. Chromosomal abnormalities have also been described. Only 13% of our patients had associated congenital defects, polydactyl being the most common anomaly. Others were uterine septum, hypospadias and renal agenesis. We, however, did not carry out chromosomal studies to thoroughly exclude chromosomal defects as this is not routine in our centre.

All patients with high malformations, rectovestibular fistulas and those who presented beyond 72 h of age had preliminary sigmoid colostomies, therefore, confining them to a three-stage procedure: Colostomy, PSARP or APT and closure of the colostomy. Amanollahi *et al*. advocated a single-stage procedure for rectovestibular fistula even though their comparative study revealed that this approach resulted in more wound complications.[14] They however surmised that these complications were more tolerable than the overall loss of time, cost of treatment and emotional adverse effects on the child and parents. High-pressure distal colostograms were performed for all the patients with high malformations before the definitive corrective surgery as a standard practice.[15]

Complication rates following surgery for ARM range from 10% to 30%.[16-18] Two of our patients with rectourethral fistula presented with urethral strictures following PSARP and had to undergo corrective urethroplasty. Documented genitourinary complications following PSARP include urethral stenosis, urethral diverticulum and neurogenic bladder.[19-21] Sudakar Jahdav *et al*. discovered in their series that non-closure of the urethral fistula reduced such complications.[19] Other complications we recorded in our study were anal stenosis and surgical site infections. Average follow-up of our patients was 6 months. Long-term follow-up is instrumental to ascertaining the quality of life these patients have as adolescence and/or adults.[22]

Conclusion

The types of ARM we documented in our study suggest the existence of geographical variation. Our patients generally presented later than their counterparts in developed countries, making a three-stage treatment protocol more common. The complications after PSARP were similar to those recorded in literature. Better documentation and public health education are advocated for earlier detection and improved management of ARM in Ghana.

Financial support and sponsorship

Nil.

Conflicts of interest

There are no conflicts of interest.

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