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Quality of Life of Children After Completion of Surgical Treatment for Anorectal Malformation: A Single-Centre Cross-Sectional Study in South-Western Uganda

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Abstract

Background

Anorectal malformations (ARMs) range from minor abnormalities such as imperforate anus to complex forms such as cloaca, and are often associated with other anomalies and residual functional stooling problems. The present study aimed to evaluate the quality of life (QoL) of children after surgical treatment for ARMs at Mbarara Regional Referral Hospital (MRRH) in South-Western Uganda.

Methods

In this cross-sectional study, we enrolled children who had completed surgical treatment for ARMs between 2014 to 2021 at MRRH. A 23-item PedsQL 4.0 inventory was used to evaluate health-related QoL in children using a multidimensional parent proxy reporting for children aged 3 months to 7 years and child self-reporting for children aged 8 to 17 years. Regression analysis was used to determine the association between sociodemographic and clinical variables and QoL. Crude and adjusted coefficients and their corresponding 95% confidence intervals (CI) were calculated. The significance level was set to a p-value < 0.05.

Results

A total of 88 participants (F:M ratio = 1.15:1) aged 3 months to 18 years with a median age of 4.5 (IQR 3–7) years were enrolled. The median age at diagnosis of ARM and preliminary diverting colostomy was 3 days (range: 2 to 30 days) and 3 days (range: 2 to 60 days) respectively. Rectovestibular fistulas, 47 (53.4%) were the most prevalent subtype of ARMs, and 5.7% of children had associated anomalies. Over one-third of the participants (38.6%) had definitive surgery after 3 years of age, and PSARP was the most common procedure. The average PedsQL score was 94 out of a maximum score of 100. The mean physical functioning score was 96.7 in males and 98.9 in females. The overall mean emotional functioning score was 91 ± 2. The mean social functioning score was 92 ± 3.0. The mean school functioning score was 95.7 in males and 98.5 in females. Associated anomalies and reoperation significantly predicted poor QoL. Increasing years after completion of surgery was significantly associated with good QoL scores.

Conclusion

The overall QoL of the participants was good. Associated anomalies and reoperation were associated with poor QoL, while years after completion of definitive surgery was associated with good QoL. There is need for increased awareness and utilization of QoL assessments as an outcome measure after definitive surgery for ARM.

Introduction

Anorectal malformations (ARMs) involve the distal anorectum and the urogenital tract, and affect approximately one in every 5000 live births, inclusive of males and females (1). They may occur as part of genetic or chromosomal syndromes, in association with other congenital anomalies, or as isolated anomalies (2). The etiology of ARMs is unknown, but is thought to be multifactorial, involving both genetic and environmental influences. In Africa, ARMs are one of the most common congenital anomalies, and a leading congenital cause of intestinal obstruction in infants (3, 4). ARMs account for 24.7% of all neonatal congenital malformations (5), and nearly 63% of these are diagnosed after the first 48 hours of life (2). More than two-thirds of cases are high ARMs, and most receive delayed surgical repair, at a median age of 11 months (4–6). This delay is attributable to limited pediatric surgical expertise, poor community awareness (7), and lack of systematic follow-up after preliminary colostomy (8). Some patients even undergo definitive surgical correction as adolescents (7, 9).

The concept of quality of life (QoL) concerns an individual's wellness and satisfaction in all aspects of life and is a holistic concept that constitutes a multidimensional assessment of a person's current life across several domains, including physical, emotional, social, and cognitive functioning (10, 11). According to some studies in developed countries, the QoL of children after definitive surgery for ARMs does not significantly differ from that of healthy children (12). Some patients however experience lowered social, physical, emotional, and cognitive functioning (10, 13, 14), hence the need for long-term follow-up (15). Even after definitive surgery, approximately 47.7% of children develop complications (4). This is more common with high and complex types of ARMs that lead to poor functional outcomes with regards to fecal continence and constipation, with subsequent poor QoL (6, 14). In the long-term, QoL outcomes can be improved following improvement in continence and appropriate surgical interventions, which are possible through involvement in a structured bowel management program (15). Despite the high burden of ARMs, limited research has been conducted about QoL in Africa. Moreover, structured bowel management programs are also not well established at tertiary hospitals in Uganda.

Studies have shown that the overall QoL and its dimensions are considerably influenced by an interplay of socio-demographic factors such as sex and age after surgery, and medical factors such as the type of ARM, associated anomalies, and time after definitive surgery (10, 14, 16). The primary aim of this study was to obtain baseline data on QoL and its modifiable determinants. This will be used as an advocacy tool to highlight the need to build capacity and establish partnerships for a sustainable formal bowel management program in order to address disease-specific concerns.

Methodology

Study design, population, and setting

This monocentric cross-sectional study involved children who had completed surgical treatment for ARMs and their caregivers at a tertiary teaching hospital in South-Western Uganda. The hospital has a

catchment area of 3 to 8 million people and serves the districts of Bushenyi, Ibanda, Isingiro, Kiruhura, Lyantonde, Mbarara, Ntungamo, Rakai, and beyond. The study was conducted in the pediatric surgery clinic that became functional in 2014. The clinic receives referred cases from Hoima, Mubende, Masaka, and Kabale districts, and neighboring countries like the Republic of Rwanda, Burundi, and the Democratic Republic of Congo.

Sample size estimation and sampling method

The sample size of 88 participants was calculated using the Kish Leslie (1965) formula, basing on the prevalence of good QoL of 93.9% among children after definitive surgery for ARM (14); considering a 95% confidence interval and a precision level of 0.05. Participants were consecutively enrolled into the study from April to August 2022.

Data assessment and collection

The pediatric surgery register at Mbarara Regional Referral Hospital (MRRH) was accessed for data on all patients who underwent definitive surgical repair for any type of ARM between 2014 and 2021. Baseline sociodemographic, clinical, and procedure-related characteristics were reviewed and extracted from the register. Contacts of caregivers of identified children were obtained from the register, and the caregivers were informed about the purpose of the study and invited via a phone call to come with their children to the surgical outpatient department of MRRH.

Written informed consent was obtained from the parents or legal guardians of children (aged < 18 years) before the process of completing the interviewer-administered questionnaire. For participants who had crossed to adulthood at the time of data collection, consent was specifically sought from them. Separate consent forms were provided to them as well as the caregivers. A translated consent form in the Runyankole language was provided to non-English-speaking participants. Assent was sought from children who were aged ≥ 8 years. Children were examined and were referred to pediatric surgeons for care in case any complications were identified. Confidentiality of the information was ensured by using unique identifiers (codes).

Quality of life assessment

QoL was assessed using the Pediatric Quality of Life Inventory (PedsQL 4.0) in four domains, namely: physical (8 items), emotional (5 items), social (5 items), and cognitive functioning (5 items). PedsQL 4.0 is a generic, multidimensional measure of Health-Related Quality of Life (HRQoL) in both children and adolescents (10, 17). The PedsQL is a reliable and validated tool that has been widely used to collect data regarding QoL of children aged up to 18 years (11, 17). This 23-item questionnaire is comprised of the core dimensions of health delineated by the World Health Organization (physical, social, and emotional functioning), as well as role/cognitive (school) functioning. Each item had a minimum score of 0 and a maximum score of 4 (0 = never a problem; 1 = almost never a problem; 2 = sometimes a problem; 3 = often a problem; 4 = almost always a problem), and the lowest scores represented a lower QoL. The questions are designed to evaluate the frequency of problems experienced over the previous month.

Children aged 5 to 18 years completed the tool by self; whereas their caregivers were evaluated for the perceived QoL of their children aged 2 to 4 years (18). Children and caregivers completed their questionnaires separately, but children aged 5–7 years were assisted by a trained research assistant. All questionnaires were checked for completeness by the principal investigator and timely corrections instituted.

A total summary score and a score for each subdomain was generated by adding up individual item scores within a distinct subdomain. Items were reverse-scored and linearly transformed to a 0–100 scale (0 = 100, 1 = 75, 2 = 50, 3 = 25, 4 = 0) so that higher scores indicate better QOL. Poor QoL was defined by a total score of ≤ 69.7 or < 65.4 based on the child's self-reported or parent-reported PedsQL 4.0 score respectively (18). The internal consistency for each scale was assessed using Cronbach's alpha coefficient.

Data management and statistical analysis

Data were entered using Epi-Data software version 7.2.1, and then exported to Stata version 17.0 for analysis. Univariate analysis using summary statistics were used to describe the baseline characteristics of the study population. This included frequencies and percentages/proportions for categorical variables such as gender. Categorical variables were compared using chi square test. Mean with standard deviation (SD), or median with interquartile range (IQR) were computed for continuous variables such as age. QoL scores were computed and expressed as mean (± SD). Bivariate and multivariate linear regression analysis was performed to determine the relationship between various sociodemographic and clinical variables and QoL domains. Crude and adjusted coefficients and their corresponding 95% confidence intervals (CI) were calculated. Variables with a p-value < 0.05 were considered statistically significant.

Results

Baseline sociodemographic characteristics

A total of 88 children (aged 3 months to 18 years) who had completed surgical treatment for ARMs between 2014 and 2021 and their caregivers were studied. The median age of children at the time of recruitment to the study was 4.5 years (range: 3 to 7 years). The median age at the time of ARM diagnosis and preliminary colostomy were 3 days (range: 2 to 30 days) and 3 days (range: 2 to 60 days) respectively. The mean age of participants at the time of definitive surgery was 3.2 ± 2.8 years, while the mean period after completion of surgery for ARM was 2.3 ± 1.6 years. In this study, there was a slight female predominance, with a female to male ratio of 1.15:1. Other sociodemographic characteristics of children and their caregivers are summarized in Table 1.

Table 1
Socio-demographic characteristics of study participants

Variable	Mean/median	Frequency (%)
Gender	-	
Male		**41 (46.6)**
Female		**47 (53.4)**
Age of participant (years), Median (IQR)	4.5 (3–7)	-
Age at completion (years), Mean ± SD	3.3 ± 3.3	-
Years after completion of surgery, Mean ± SD	2.3 ± 1.6	-
Primary caretaker	-	
Mother		68 (77.3)
Father		18 (20.4)
Other		2 (2.3)
Caretakers education level	-	
No education		10 (11.4)
Primary		51 (57.9)
Secondary		17 (19.3)
Tertiary		10 (11.4)
Caretakers occupation	-	
Unemployed		5 (5.7)
Peasant		61 (69.3)
Business		15 (17.1)
Civil employment		7 (7.9)
Monthly income (UGX), Median (IQR)	200,000 (60,000-300,000)	-
Distance from hospital (Km), Median (IQR)	195 (73–367)	-

Clinical and procedure-related characteristics

In this study, all females, 47 (100%) had rectovestibular fistula. More than three-quarters of males, 32 (78.1%) had rectourethral fistula, while only 9 (21.9%) had perineal fistula. Majority of participants, 87 (98.9%) underwent PSARP, while only 1 (1.1%) underwent anoplasty. Of the 87 participants who

underwent PSARP, 47 (54.0%) had rectovestibular fistula, 8 (9.2%) perineal fistula, while 32 (36.8%) had rectourethral fistula. This is summarized in Fig. 1.

More than one-third of the study participants, 34 (38.6%) had definitive surgery at a later stage in life (> 3 years). Five participants (5.7%) had associated anomalies, out of which 2 (2.3%) had cardiac anomalies, 2 with extra digits (polydactyly), and 1 with Down's syndrome. Other clinical and procedure-related characteristics of children with ARMs are summarized in Table 2.

Table 2
Clinical and procedure-related characteristics of children with ARMs

Variable	Median (IQR)	Frequency (%)
Type of corrective surgery		
PSARP		**87 (98.9)**
Anoplasty		**1 (1.1)**
Age at corrective surgery		
< 28 days		**2 (2.3)**
1 month – 1year		**22 (25.0)**
> 1–3 years		**30 (34.1)**
> 3 years		**34 (38.6)**
Anomalies associated		
Cardiac		2 (2.3)
Other		3 (3.4)
Symptoms		
Acute intestinal obstruction		44 (50.0)
Chronic constipation		1 (1.1)
Abnormal opening		43 (48.9)
Duration of symptoms (days)	4 (2–60)	

Quality of life scores

The mean total QoL score among children who had undergone surgery for ARMs was 91.3 for rectourethral fistulas, 95.7 for rectovestibular fistulas, and 94.1 for perineal fistulas. No difference was observed between overall PedsQL scores among the three types of ARMs. The distribution of individual

total PedsQL 4.0 score is presented in Fig. 2. Overall, the total score was high; and only two patients had a score < 70, consistent with poor QoL. The total PedsQL 4.0 scores in 48 (53.8%) children with rectovestibular fistulas, and 30 (34.1%) children with rectourethral fistulas were < 95, and the differences in the scores were not statistically significant ($p = 0.820$). As shown in Table 3, Cronbach's alpha coefficient for the overall parent proxy-report form was 0.90, indicating internal consistency.

The mean physical functioning score was 96.7 in males and 98.9 in females. Functional outcome scores declined with increasing level of fistula (95.6 in rectourethral and 96.3 in rectovestibular fistulas [$p = 0.001$]). There was no correlation between current age of the child and physical functioning (Pearson $r^2 = 0.001$). Male participants who underwent PSARP showed lower physical functioning outcomes compared to females with a mean score difference of 3.1. The overall mean emotional functioning score was 91 ± 2. The mean scores were distributed by gender as follows; 89.9 and 92 in males and females respectively. Emotional functioning scores were worse with increasing age at definitive surgery (correlation coefficient = 0.204, $p = 0.05$). The mean social functioning score was 92 ± 3.0. There were better social functioning outcomes among female participants than the males though this correlation was not statistically significant (Pearson r2 = 0.007). The mean school functioning score was 95.7 in males and 98.5 in females. Children with ARMs associated anomalies showed poor school/cognitive functioning scores (F = 8.7; $p = 0.001$) than those participants without associated anomalies.

Table 3
Scale descriptives for PedsQL™ 4.0 Generic Core Scales: parent proxy-report

Scale (items)	N	Cronbach's α	Mean ± SD	Range
Total score (23)	-	.900	-	-
Physical functioning (8)	84	.951	97.9 ± 9	37.5–100
Emotional functioning (5)	88	.738	91.0 ± 13	40–100
Social functioning (5)	86	.790	92.2 ± 14	25–100
School functioning (5)	55	.572	97.1 ± 1.4	70–100

Factors associated with poor QoL

Correlation analysis showed that PedsQL scores were not correlated to age of participant at time of interview, duration of symptoms, and age at time of definitive surgery. No correlations were found between the caregiver characteristics and the total PedsQL score. We used the total PedsQL 4.0 score and the scores of each dimension for dependent variables in the linear regression analysis as summarized in Table 4 and Table 5. Associated anomalies and reoperation were associated with poor QoL. Children with more years after definitive surgery were more likely to have good QoL. The power of this study was 0.85.

Table 4
Bivariate analysis for factors associated with poor QoL after definitive surgery of ARMs

Variable	Unadjusted coefficient	95% CI	P value
Age of participant (years)	-0.2875	-0.8717 to 0.2966	0.330
Gender	3.1997	-1.1630 to 7.5625	0.148
Male			
Female			
Age at corrective surgery (years)	-0.2095	-0.8822 to 0.4631	0.537
Years after definitive surgery	-0.6786	-2.0628 to 0.7055	0.032
Duration of symptoms (months)	-0.1765	-0.7213 to 0.1932	0.435
Associated anomalies	-4.6262	-7.4087 to 1.8431	0.001
Reoperation	-8.1983	-14.551 to -1.8451	0.012
ARMs subtypes	-1.0377	-2.1745 to 0.0991	0.073
Rectovestibular fistula			
Rectourethral fistula			
Perineal fistula			
Symptoms	Ref	.01115 to 4.3424	0.049
Chronic constipation	2.1767		
Abnormal opening			
Intestinal obstruction			

Table 5
Multivariate analysis for factors associated with poor QoL after definitive surgery of ARMs

Variable	Adjusted coefficient	Standard error	95% CI	P-value
Associated anomalies	4.1619	1.47	1.23 to 7.08	0.006
Reoperation	5.1071	3.40	1.66 to 11.87	0.048
Years after definitive surgery	-0.8850	-0.65	-2.18 to -0.41	0.050
Overall R-squared = 0.1613, $p = 0.0027$				

Discussion

We evaluated the QoL of children after definitive surgery for ARMs; which is the ultimate endpoint of research in health outcomes (15). Generally, children with ARMs experience lower physical, emotional, social, and school functioning compared to healthy peers (10). The overall QoL of participants in this study was good, with an average PedsQoL of 94, which is comparable to 93.9 as reported by Raman et al. (14) and the findings of other researchers (15).

In this study, the physical functioning outcome was good, with a mean score of 97.9. This is consistent with the findings of Bhojwani et al. (15) who studied 13 patients in India, and Shahbal et al. (19) who studied 25 patients at Kenyatta National Hospital. All participants in these studies experienced significant improvement in their physical functioning outcomes following definitive surgery. The study by Grano et al. (10) observed no significant differences in overall physical functioning among patients with ARMs and healthy counterparts. In the current study, male participants who underwent PSARP showed lower physical functioning outcomes compared to females. This is not congruent with the results of Grano et al. (10) who found worse QoL in the physical functioning domain among adolescent females compared to males. According to earlier studies in sub-Saharan Africa, males are more likely to have rectourethral fistulas that are classified as high ARMs (2). The higher the level of fistula, the more complex the surgical technique and likelihood of repeat surgery and complications that impact on the physical functioning outcomes (14, 15). We did not establish a relationship between age of participant and level of physical functioning outcome.

Emotional functioning was good, with a mean score of 91; and worsened with increasing age at definitive surgery. This high QoL score is possible because adolescents with ARMs develop stronger psychosocial strategies and coping capabilities, probably because they learn to live with chronic functional problems (15). Nonetheless, evidence shows that children with ARMs are more sad, angry, scared, or worried when compared to their normal peers (10).

Earlier studies that assessed social relationships found that unlike healthy children, those with ARMs are less capable to be welcomed by peers and teachers (10, 20). Our results showed that children who were reoperated or had associated anomalies had a significantly low social functioning ($p = 0.002$ and $p = 0.008$ respectively). Parental support with respect to strategies that minimize incontinence reduces as the child ages, and yet, fecal continence scores may worsen with increasing age (20). Although bowel function improves with increasing age, QoL outcomes remain unsatisfactory by adulthood, usually as a result of the continued influence of earlier negative self-perceptions of illness during childhood (21).

The Cronbach's alpha coefficient of the cognitive domain in our study was very low (0.572). This therefore compromised the results of school functioning because of repeated school absence and dropout due to incontinence; while other participants had not yet joined school. However, those that responded to this subscale had good QoL, similar to a study that compared children with ARMs and healthy peers. The study revealed no differences in any of the QoL domains between patients and healthy peers (14). Researchers have offered explanations for this finding; suggesting that once children with

ARMs join school, they try to fit in and tend to hide their difficulties from their peers, hence presenting better QoL scores.

Regarding the second aim of this study, we described the predictors of poor QoL among children who had undergone definitive surgical repair for ARMs. All female participants in our study presented with rectovestibular fistula, whereas majority of the males had rectourethral fistula. There was no correlation between the type of ARMs and QoL, and no significant difference was observed in PedsQL score among the three types of ARMs. The mean total scores of children with rectourethral, rectovestibular, and perineal fistulas were 91.3, 95.7, and 94.1 respectively. A study published by Kayima and colleagues (2) found similar results, based on the patient population at Mbarara Regional referral hospital. Whereas high ARMs account for more than two-thirds of ARM cases in Uganda (2, 5), some studies in sub-Saharan Africa and India have not found a significant difference between low and high ARMs in relation to the functional outcomes and QoL (6, 14). On the other hand, a two-centre study of 48 children reported a significant decrease in the stooling scores with increasing complexity of the ARM (22).

In the current study, other congenital anomalies were present in 5.7% of participants. This prevalence is lower than 20% reported by Kayima et al. in Uganda (2), 16.5% by Mfinanga et al. in Tanzania (4), and 20.5% by Ogundoyin et al (23) in Nigeria. Variations in the prevalence may be related to challenges in the diagnosis of genetic syndromes in resource-constrained settings, given that diagnostic investigations are not routinely performed (24), and the fact that associated anomalies are significantly linked to increased mortality (2, 4). Moreover, some of these patients may not even present to the hospital. Earlier studies established that other congenital anomalies, commonly categorized under the VACTERL association are very common among patients with ARM (14). The associated anomalies documented in our study were Down's syndrome, congenital heart disease, and polydactyly. In the current study, the presence of associated congenital anomalies was associated with poor QoL ($p = 0.005$). This is similar to the findings of other studies (14, 22).

Our results revealed a significant association between reoperation and poor QoL compared with primary successful anastomosis. This is comparable to other studies (19). Findings by Shahbal and colleagues (19) show that 70% of patients who undergo reoperation experience unsatisfactory bowel function. Another potential explanation for poor QoL scores is that reoperation is associated with depression relating to body image as well as anxiety and chronic pain syndrome, which in turn affect the QoL. Several studies have documented theories explaining the likely cause of repeat surgery. These span from poor quality colostomies to inadequate nursing care (8). Appropriate pre-operative preparation and management of constipation are essential for ensuring good outcomes. Even when children develop treatable complications, they often return late (8). Families also experience fatigue from going through multiple stages of surgeries, and they as well lack adequate information about the importance of continuous follow up, and experience challenges in accessing healthcare (8, 24). This further compromises the QoL.

In our study, the QoL scores improved with increase in the years after definitive surgery. The assumed explanation for this finding is that a longer period after surgery results in better healing; and that these children increase the ability to do things on their own. Few studies have attempted to compare the differences in QoL and years after definitive surgery. Harumatsu et al. (16) reported higher post-operative bowel function scores among 52 males in Japan. By 9 years of age, satisfactory bowel movement score had been achieved; and a slow but continuous improvement was observed in relation to the constipation, incontinence and soiling scores until the age of 11 years. Conversely, Hashish et al. (22) found that functional stooling scores significantly worsened with increasing age among patients with high imperforate anus at two centers in the United States and Egypt. These differences may be explained by variations in the age of the study population and research methodology. Nevertheless, it is likely that a high prevalence of functional symptoms persists among patients who have undergone definitive surgery, affecting the QoL even in adulthood (25).

This study was not without limitations. There was a possibility of social desirability and recall bias from study participants. However, this was minimized by the use of a standardized tool. In addition, participants were informed that all information obtained would be handled with utmost confidentiality, and they were given ample time to respond to the questions.

Conclusions

The overall QoL of the study participants was good. Associated anomalies and reoperation were directly associated with poor QoL of children after completion of surgical treatment for ARMs. Increasing number of years after completion of definitive surgery was associated with good QoL. Children with ARMs should be thoroughly screened for associated anomalies and interventions for QoL included in the treatment package. Train all staff and caregivers about domains of QoL of children with ARMs and the bowel management program.

Abbreviations

ARMs, Anorectal malformations; MRRH, Mbarara Regional Referral Hospital; PedsQL, Pediatric Quality of Life Inventory; PSARP, Posterior Sagittal Anorectoplasty; QoL, Quality of Life.

Declarations

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Contributions. DM, FO, WIE, and MMM conceived and designed the study. DM collected the data. DM, WIE, and MA analyzed the data and interpreted the results. DM, WIE, and MA performed literature review and wrote the first manuscript. FO, MMM, KDK revised and edited the manuscript. DM and WIE wrote the final manuscript. All authors read and approved the final manuscript.

Ethics approval and consent to participate: Ethical approval was obtained from the Mbarara University of Science and Technology Research Ethics Committee (No. MUST-2022-374). Administrative clearance was subsequently sought from the office of the Executive Director of MRRH. All study procedures were performed in accordance with the Declaration of Helsinki. Written informed consent was obtained from all study participants.

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Figures

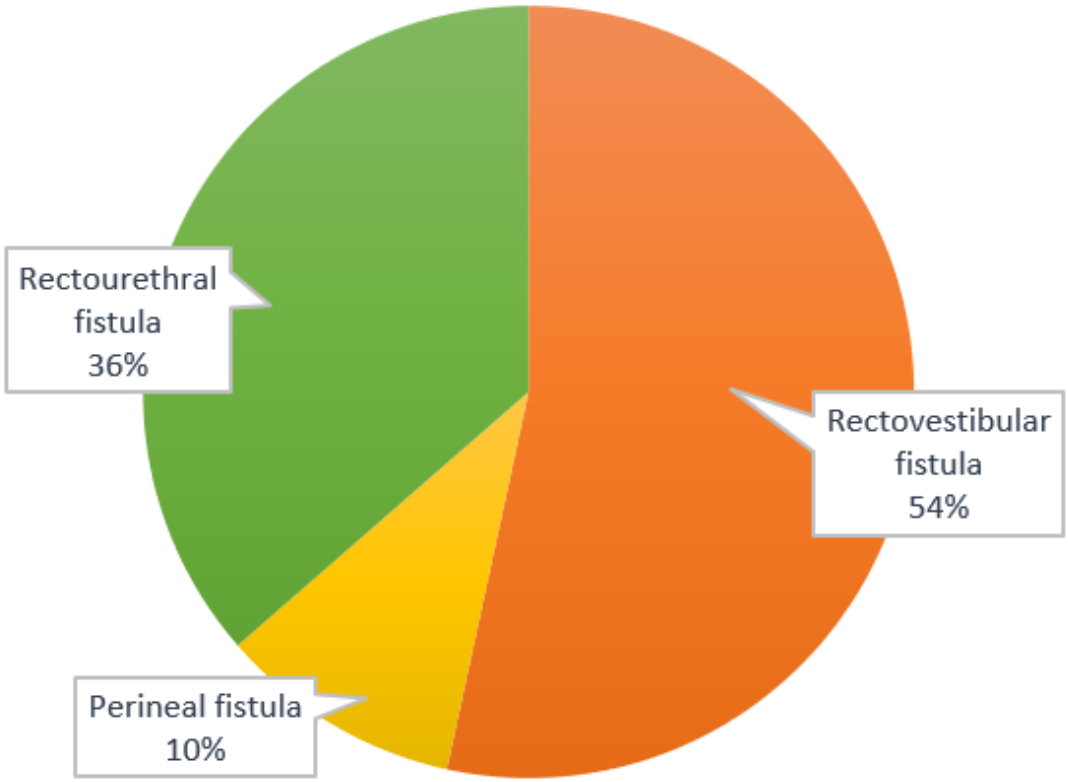

Figure 1

Distribution of the subtypes of ARMs

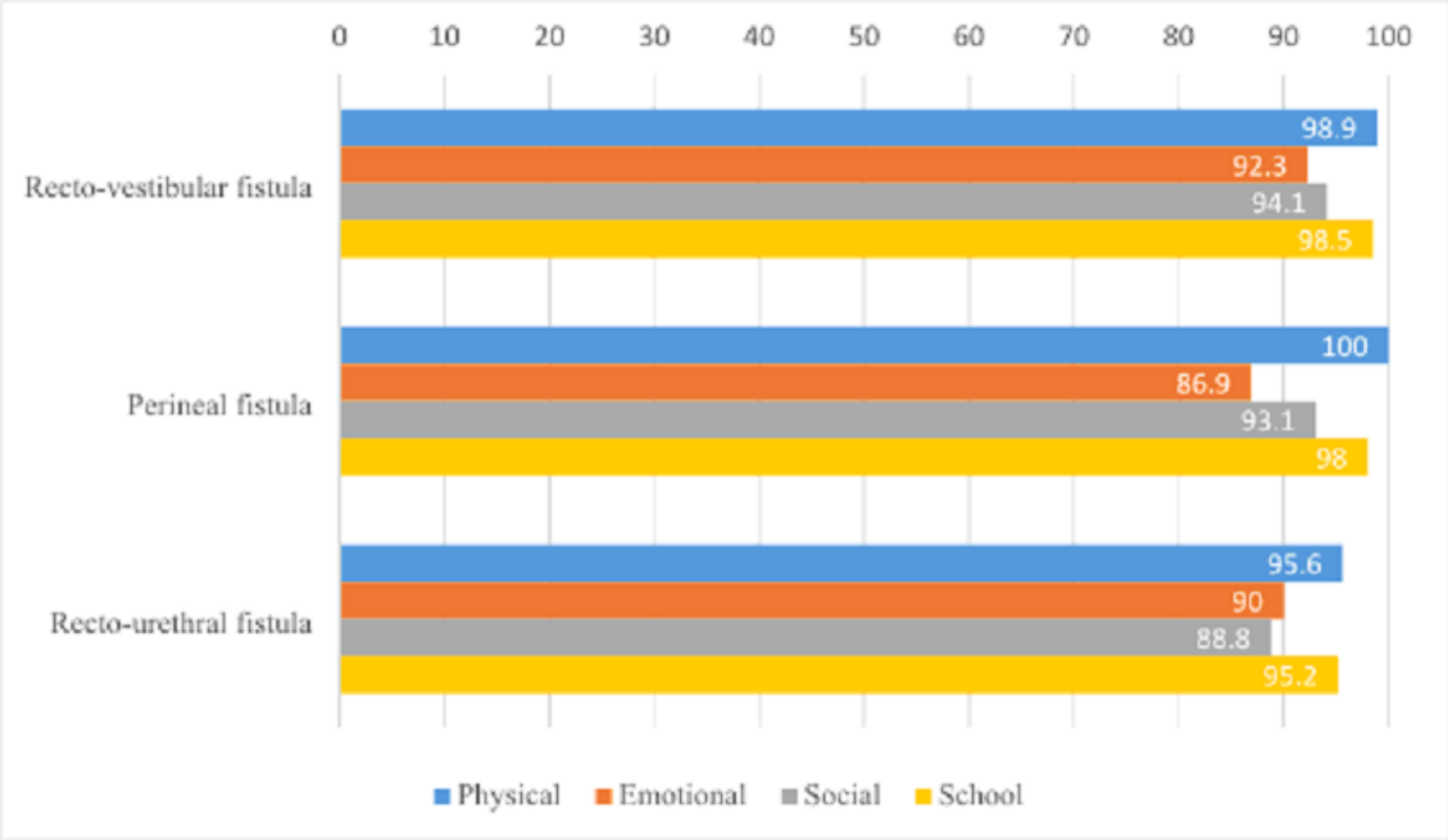

Figure 2

Distribution of PedsQL scores by subtype of ARMs