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ORIGINAL ARTICLE

Survey on the management of anorectal malformations (ARM) in European pediatric surgical centers of excellence

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Abstract
Purpose Various management strategies for anorectal malformations (ARM) have been proposed. The aim of this study was to assess the current management in centers of excellence in Europe.
Methods An online survey on the pre- and post-operative concepts, surgical techniques, and the management of complications was sent to the representative experts of 28 selected European centers of pediatric surgery with special expertise in the treatment of ARM.
Results The survey was completed by 25 experts from 14 countries. To assess the level of the rectal pouch in newborns 60 % of participants perform a prone cross-table X-ray and 52 % ultrasound. If an ostomy is required, 84 % create divided Peña stomas. Primary repair in the newborn period is performed in females with rectoperineal (92 %), rectovestibular (60 %), and no fistula (32 %), and in males with rectoperineal (92 %), rectourinary (17 %) and no fistula (38 %). For 68 % of surgeons, the PSARP is the preferred surgical approach for "low" malformations. Laparoscopically assisted pull-throughs are routinely performed by 48 % of experts for ARM with bladderneck and 28 % for rectoprostatic fistula. 88 % perform postoperative dilations.
Conclusions The management of ARM in Europe is very heterogeneous. High-quality clinical studies are needed to provide scientific evidence for the optimal treatment strategies.

Keywords Anorectal malformation · Survey · Management · European

Introduction

Anorectal malformations (ARM) represent a wide spectrum of anomalies. The classification and operative management of ARM have been dominated by the extensive work of Alberto Peña. Since the first description of posterior sagittal anorectoplasty (PSARP) in 1982 [1, 2] this procedure has evolved as the standard technique for numerous surgeons worldwide. However, there are still controversial topics in the management of ARM, including various diagnostic and peri-operative protocols.

Several surveys on the management of other colorectal diseases, such as Hirschsprung disease (HD), have been published [3, 4]. A survey among pediatric surgeons from the UK showed that the primary pull-through during the first 3 months of life, using an open Duhamel or laparoscopic-assisted Soave–Boley technique, had become the operative strategy of choice in rectosigmoid HD in the UK. Marked variation in practice remained for right-sided HD [3]. A survey conducted among US American surgeons revealed that a 1-stage transanal operation with or without laparoscopy had become the most common strategy for the surgical management of the typical baby with HD. Opinions varied about the amount of colonic resection, length of the rectal cuff, and site of initiation of the anorectal dissection [4].

To our knowledge, a survey on the current management of ARM in different institutions has not been carried out.

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Therefore, we assessed the current management strategies of ARM in European centers of excellence.

Materials and methods

An internet-based survey consisting of 38 questions on the management of ARM was launched in April 2014 using an online provider for web-based surveys (SurveyMonkey™). The heads or subheads of departments (representatives) of 28 centers from 16 different European countries with a special expertise in pediatric colorectal surgery were invited to participate.

Centers with an international academic profile indicated by recent publications on ARM were included. Moreover, these centers should also represent a large and prominent institution of their country. The invitation was sent out by E-mail with an embedded link to connect to the survey. Three reminder messages were sent during a 3-week period using the same mailing list.

The first section of the survey comprised questions on the professional background of the respondent, including general information on his/her institution, the average number of ARM cases treated per year, and the number of pediatric surgeons individually operating on ARM. In a second part surgeons were asked to complete sections regarding their management of ARM: preoperative diagnostic workup, preoperative management of anorectal repair, surgical techniques, postoperative management after anorectal repair, management of complications, and other aspects.

Statistical methods

Answers were anonymously collected, converted into a database with Microsoft Office Excel (version 2007), and analyzed using descriptive statistics (frequency and percentage of pre-defined subgroups according to the options for each question). Fisher exact test was performed to compare categorical variable frequencies. p value <0.05 was considered as the level of significance.

Results

General information

The online questionnaire was completed by representatives from 25 of 28 (89 %) centers from 14 different European countries. The characterization of participating institutions is summarized in Table 1.

Table 1 Characterization of participating centers

Basic institutional data	*N* (%)
Primary workplace	
University hospital	20 (80 %)
Hospital with academic affiliation	5 (20 %)
Others	0 (0 %)
Number of ARM cases treated on average per year	
<5	0 (0 %)
5–9	11 (44 %)
10–19	11 (44 %)
20–30	3 (12 %)
>30	0 (0 %)
Fully trained pediatric surgeons independently operating on ARM	
1	2 (8 %)
2	7 (28 %)
3	9 (36 %)
4 and more	7 (28 %)

Preoperative diagnostic workup and management

The data on preoperative workup are summarized in Table 2. Preoperatively a Foley catheter is routinely placed in the operating room by 22 (88 %) of the participants in both male and female patients, while 2 (8 %) place it occasionally and 1 (4 %) never. A central venous line prior to surgery is placed routinely by 2 (8 %), occasionally by 19 (76 %), and never by 4 (16 %) of the respondents. In presence of an external orifice, a preoperative full bowel preparation before surgery is performed regularly by 9 (36 %), occasionally by 6 (24 %), and never by 10 (40 %) of the participants. An endoscopy (cystoscopy and vaginoscopy) is routinely performed by 6 (24 %), while 15 (60 %) perform it occasionally, and 4 (16 %) never.

Surgical management

Ostomies

When a diverting enterostomy is required, a divided descending colostomy with mucous fistula ("Peña stoma") is chosen by 21 (84 %) of the participants, while 4 (16 %) prefer a transverse or descending loop colostomy.

Anorectal repair in the newborn period

In female patients a primary anorectal repair without a diverting enterostomy is routinely performed for perineal fistula by 23 participants (92 %), vestibular fistula by 15 (60 %), and in patients with no fistula with rectal gas below the level of the coccyx by 8 (32 %).

Table 2 Preoperative diagnostic workup

Questions, possible answers	*N* (%)
Investigations routinely performed in the neonatal period	
Plain and/or lateral X-ray of the entire spinal column	11 (44 %)
Plain and/or lateral X-ray of lumbar and sacral spine	11 (44 %)
Echocardiography	23 (92 %)
Abdominal ultrasound, including the urinary tract	24 (96 %)
Spinal ultrasound	22 (88 %)
Determination of the level of the rectal pouch if the patient has no external orifice	
Upside-down inversion X-ray	2 (8 %)
Prone cross-table lateral X-ray	15 (60 %)
Ultrasound	13 (52 %)
Only	4 (16 %)
Combined with prone cross-table lateral X-ray	7 (28 %)
Combined with upside-down X-ray	2 (8 %)
Other	4 (16 %)
Not perform any investigation and directly open a colostomy if no bulging of meconium is evident	4 (16 %)
Performance of a voiding cystourethrography (VCUG) in a boy with suspected rectourinary fistula	
Routinely	10 (40 %)
Occasionally	9 (36 %)
Never	6 (24 %)
Calculation of the sacral ratio as predictor of future continence	
Routinely	8 (32 %)
Occasionally	9 (36 %)
Never	8 (32 %)

In male patients a primary repair without enterostomy is attempted in perineal fistula by 22 (92 %) of the respondents, and in patients with rectal gas below the coccyx in both rectourinary fistula by 4 (17 %) and no fistula ($n = 9$, 38 %).

For "low" ARM (e.g., with rectoperineal fistula) the posterior sagittal anorectoplasty (PSARP) is performed by 17 (68 %), while 4 (16 %) prefer an anterior sagittal approach (like either the Mollard approach [5] or its modifications [6–8]) and 4 (16 %) use other techniques (e.g., cut-back, V–Y plasty).

Regarding the percentage of children in whom a primary anorectal repair without a diverting enterostomy is performed in the newborn period, there was no significant difference between centers with a "low" versus a "high" patient volume (Table 3).

The role of laparoscopy

The data on laparoscopic-assisted anorectal pull-throughs (LAARP) are shown in Table 4. Briefly summarized, LAARP is routinely performed by 48 % of experts for ARM with bladderneck and 28 % for rectoprostatic fistula. Regarding the judgement of functional outcome of LAARP, 17 (71 %) of participants stated that LAARP has superior results.

Postoperative management after anorectal repair

Feeding after anorectal repair in patients without a diverting enterostomy is usually restarted after the first bowel movement by 12 (48 %) of respondents, 3 (12 %) restart on the day of surgery, 3 (12 %) within the second/third postoperative day, 6 (24 %) after 1 week, and 1 (4 %) after 2 weeks. Most of the surgeons remove the Foley catheter within the first postoperative week (67 % in female and 63 % in male patients; range 2–6 days). Various clinical practices were found for the postoperative antibiotic administration, as 3 (12 %) of the participants give antibiotics only for 24 h, 4 (16 %) for 2 days, 10 (40 %) for 3–6 days, 6 (24 %) for 1 week and 2 (8 %) only in selected patients for variable duration.

The vast majority of the participants routinely perform postoperative dilations ($n = 22$, 88 %) and 1 participant (4 %) never dilates his/her patients postoperatively. Patients are usually scheduled for the first dilation 2 weeks after surgery ($n = 17$, 74 %). In most of the centers the frequency of the initial dilations is twice daily ($n = 12$,

Table 3 Surgical management: primary repair without a diverting enterostomy in the neonatal period

	Centers performing 0–10 new cases/year ($N = 11$) "low volume" *N*	Centers performing 11–30 new cases/year ($N = 14$) "high volume" *N*	*p* value ("low" versus "high" volume)
Female patients			
Perineal fistula	11 (100 %)	12 (86 %)	ns
Vestibular fistula	6 (55 %)	9 (64 %)	ns
No fistula + rectal gas below the level of the coccyx	4 (36 %)	4 (28 %)	ns
Cloaca	0 (0 %)	0 (0 %)	ns
Male patients			
Perineal fistula	10 (91 %)	12 (92 %)	ns
No fistula + rectal gas below the level of the coccyx	5 (45 %)	4 (28 %)	ns
Rectourinary fistula + rectal gas below the level of the coccyx	2 (18 %)	2 (14 %)	ns

Table 4 The role of laparoscopy

Questions, possible answers	*N* (%)
Performance of laparoscopically assisted anorectal pull-through (LAARP)	
Never	7 (28 %)
Occasionally, for rectourethral fistula (bulbar)	1 (4 %)
Occasionally, for rectourethral fistula (prostatic)	5 (20 %)
Occasionally, for bladderneck fistula	6 (24 %)
Routinely, for rectourethral fistula (bulbar)	1 (4 %)
Routinely, for rectourethral fistula (prostatic)	7 (28 %)
Routinely, for bladderneck fistula	12 (48 %)
For other types of ARM	3 (12 %)
Judgement of functional results of laparoscopically assisted anorectal pull-through (LAARP) compared to the traditional approach	
LAARP has equal results	17 (71 %)
LAARP has superior results	3 (12 %)
LAARP has worse results	4 (17 %)
Surgical technique for closing the fistula (only centers which perform LAARP)	
Endoloop	2 (11 %)
Suture ligation	11 (61 %)
Clip	2 (11 %)
We do not close it and leave it open	2 (11 %)
Other (please specify)	1 (6 %)
Prostatic fistulas can be left open, bladder neck fistulas suture ligated	

52 %), while in 7 (31 %) dilations are performed only once daily, 1 (4 %) every second day, 2 (9 %) every third day, and 1 (4 %) center usually chooses the frequency according to the pain of the patient. Concerning the dilation protocol and the Hegar dilator size that has to be reached, 17 (68 %) of the respondents follow Peña's protocol [9], while 4 (16 %) use a regimen with smaller sizes, 1 (4 %) bigger sizes, and 3 (12 %) do not follow any specific protocol. Dilations are usually continued for 2 weeks to 3 months by 10 (43 %), for 3–6 months by 11 (48 %), and for more than 6 months by 2 (9 %) participants. The majority of the surgeons ($n = 22$, 88 %) believe that routine dilations are useful in reducing the incidence of anal stenosis.

Management of complications

In case of a superficial dehiscence of the perineal body during postoperative dilations almost all participants ($n = 22$, 88 %) choose a conservative management. Nine

(37 %) would stop the dilations while 5 (21 %) would continue, and 10 (42 %) did not express any preference regarding dilations. In case of a therapy-resistant rectal stenosis (e.g., short ring-like stricture, 1-year old child) 13 (52 %) would perform a surgical plasty (e.g., Heineke-Mikulicz), 10 (40 %) would perform a redo-anorectoplasty via a posterior sagittal approach, 1 (4 %) would perform corticoid injections, and 1 (4 %) would choose a different technique depending on the degree of the stenosis. Eight (32 %) of the respondents had experienced a symptomatic postoperative urethral diverticulum and different surgical solutions for this complication were chosen by the participants (33 % laparoscopic resection, 5 % laparotomic resection, 10 % combined abdomino-perineal resection, 19 % resection via posterior sagittal approach, and 33 % other techniques including a transvesical approach).

Other aspects

Almost all participants ($n = 22$, 88 %) include their patients with ARM in scientific studies, and 7 (28 %) routinely perform genetic investigations. A contact with parents' organizations is regularly promoted by 19 (76 %) of the centers, while 3 (12 %) of the respondents stated that a parents' organization does not exist in their country.

Discussion

To the best of our knowledge, we present the first multinational survey on the management of anorectal malformations (ARM) in selected European centers of Pediatric Surgery.

Centralization of the surgical and medical treatment of rare conditions in children such as orofacial clefts [10], brain tumors [11], cardiac malformations [12] and biliary atresia [13–15] has been recommended by numerous authors. However, this survey showed that 88 % of the centers treat less than 20 new ARM cases per year while having 3 or more surgeons who are independently performing anorectal repairs. This indicates that with a low case load per surgeon the training opportunities for new surgeons may be limited. A similar conclusion was drawn for the treatment of Hirschsprung disease in the survey conducted in the UK by Bradnock et al. [3].

The current survey showed that the role of echocardiography to detect cardiac defects, ultrasound to screen for urinary tract anomalies, and spinal ultrasound to rule out tethered cord, is generally accepted as these investigations are routinely carried out by 92, 96, and 88 % of participants, respectively.

In 1995, the determination of the "sacral ratio" was proposed by Peña to predict functional outcome of patients with ARM [16]. Torre et al. [17] demonstrated that, although there is considerable variation in abnormalities in sacral development, a sacral ratio of less than 0.52 could be considered pathological. In the last decade some authors questioned the validity of the sacral ratio to detect sacral anomalies as a tool to predict future continence [18, 19]. The current survey revealed that the determination of the sacral ratio is not considered an important part of the preoperative workup in Europe, as only 32 % of the respondents routinely measure it.

In recent years the prone cross-table lateral X-ray has been favored over the upside-down inversion X-ray, which was described by Wangensteen and Rice [20], to assess the level of the rectal pouch and decide on the appropriate surgical approach in the neonatal period. Advantages of the prone cross-table lateral X-ray include easy positioning, better cooperation of the patient, elimination of the effect of gravity, and better delineation of the rectal gas shadow [21]. However, our survey shows that although 60 % of respondents perform cross-table lateral X-rays, the upside-down inversion X-ray is still routinely performed in 8 % of the participating centers. In the last years transperineal ultrasonography has been proposed as an alternative method to determine the level of the rectal pouch and to identify a possible fistula [22, 23]. This method is used in 52 % of European institutions: 16 % would only perform ultrasound alone and 36 % in combination with an X-ray. As studies comparing ultrasound versus cross-table lateral X-rays are lacking, the future role of these radiologic techniques has to be determined.

Voiding cystourethrography (VCUG) to detect a rectourinary fistula has been suggested [24, 25]. This survey showed that 40 % of the participants regularly perform VCUGs in male patients with ARM, although there is no data on the specificity and sensibility of VCUG in detecting fistulas.

When a diverting enterostomy is required, Peña et al. [26] recommend the opening of a descending colostomy with separated stomas and distal mucous fistula. In a large retrospective study he showed disadvantages of loop colostomies including spilling of stool into the distal stoma, which may promote urinary tract infections and fecal impaction in the distal pouch with corresponding megarectum. Another downside is the possible absorption of urine by the colon if a rectourinary fistula is present. Finally, the prolapse rate of loop ostomies was significantly higher, which was also described by others [27].

This survey clearly indicates that in European centers the divided descending colostomy (Peña stoma) is the most popular technique, which is preferred by 84 % of the participants. Only 16 % of participating surgeons bring out the transverse or descending colon as a loop-enterostomy.

Moreover, our survey revealed that the decision on when to attempt a primary repair or create a diverting

enterostomy is very variable between centers. One would expect that institutions with a high volume of children with ARM treated per year create the smallest number of stomas and prefer primary repair in the newborn period. However, this was not confirmed by our survey (Table 3) as the preference for a protective ostomy did not correlate with the caseload per year.

Almost every operation has been performed in children using minimally invasive techniques. Correspondingly, minimally invasive surgery has also influenced the surgical treatment of ARM [28–34]. LAARP was proposed especially for "high" types of ARM such as rectobladderneck and rectoprostatic fistulas [31] in which the rectum is not reachable posterior-sagittally. This corresponds with the answers in the current survey. Despite the fact that the majority of the participants believe that LAARP offers equal (72 %) or even better (12 %) results when compared to the traditional open approach, laparoscopy for ARM is not routinely used all over Europe. Our survey shows that it is performed by less than half of the participants, and almost one-third had never performed laparoscopy for ARM (Table 4). Despite enthusiastic reports on laparoscopy for ARM, it is important to recognize that advantages have never been proven by a randomized study. Bischoff et al. [31] performed a literature review on 47 publications and concluded that although LAARP is a less invasive procedure compared to open surgery, there is no evidence that the functional results are better. In addition in 2013 Bischoff et al. [34] described the use of laparoscopy for fistula ligation followed by PSARP as a technical modification for ARM with rectobladderneck and high prostatic fistulas. The authors concluded that this approach may avoid injury to the urinary tract and vas deferens due to the semi-blind introduction of a trocar through the perineum and may more precisely place the rectum in the center of the sphincter complex. However, our survey did not assess the application of this technique.

The protocols used for postoperative management of ARM in this survey were heterogeneous. The role of postoperative anal dilations has been discussed controversially, as studies indicated that anal dilations may act as a chronic stressor for children and parents [35]. Both, mental health and psychosocial functioning correlated significantly with the duration of anal dilations [35, 36]. However, despite all these side effects of routine anal dilations, 88 % of participants believe that this procedure is useful in order to reduce the incidence of anal stenosis. The majority of participating centers (74 %) regularly initiate dilations 2 weeks after surgery, as suggested by Peña and Levitt. In contrast to the recommendations by Peña [9], Temple et al. suggested that weekly calibration by the surgeon is associated with similar outcomes compared to daily dilation by the parents [37]. However, this survey clearly shows that the majority of the centers prefer daily dilations by the parents (52 % twice daily, 31 % once daily) with 68 % of the participants following the Peña protocol [9] in Europe.

The treatment of complications after surgery for ARM is challenging. General agreement was reached on the conservative treatment for a superficial dehiscence of the perineum during dilations. However, this survey revealed a great variety of answers for more complex complications, such as therapy-resistant rectal stenosis and urethral diverticulum. The surgical strategies for such complications are not standardized.

Genetic information of children with ARM is increasingly reported in recent years. However, genetic investigations are still not part of the routine diagnostic workup of patients with ARM, as they are regularly performed by only 28 % of the participants.

Despite offering a general picture of the current practice in the management of ARM in European centers, we are aware of limitations of our study. There was a bias in selecting the centers as we did not adopt a clear definition of "center for pediatric colorectal surgery". For this reason some centers of excellence might have been missed. In addition, we cannot conclude that the data are really representative of the European practice, as the questionnaire was completed by only one representative expert per center. Therefore, we cannot exclude that multiple protocols exist within one institution.

The diagnostic and therapeutic concepts for the management of children with ARM are highly variable in European centers of excellence. There is no generally accepted protocol which mirrors the lack of comprehensive data to support superiority of any approach (including the PSARP) in the management of anorectal malformations in the literature. In the future, a multicenter prospective approach is needed to provide evidence on outcome of the different surgical strategies which are currently in use.

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