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Accuracy of Transperineal Sonography and Distal Loopogram in Determining Distal Rectal Pouch-Perineal Distance and Fistula on Anorectal Malformation Post Colostomy Patients in Dr. Mohammad Hoesin General Hospital Palembang

Halomoan Sejahtera[1], Saksono Sindu[1*], Uli Hanna Marsinta[2], Bahar Erial[3]

[1] Department of Surgery, Dr. Mohammad Hoesin General Hospital, Palembang, Indonesia

[2] Department of Radiology, Dr. Mohammad Hoesin General Hospital, Palembang, Indonesia

[3] Department of Anatomy Faculty of Medicine Sriwijaya University, Palembang, Indonesia
Corresponding author: sindusaksono92@gmail.com

Abstract

Background: Imperforated anus is a congenital abnormality which is better known as anorectal malformation. The incidence of anorectal malformations is 1 in 4000 to 5000 live births. The incidence of anorectal malformations in Europe varies from 1.14 to 5.96 per 10,000 people and can change each year.[1-3] The surgical approach in the management of anorectal malformations depends on the classification of anorectal malformations based on low, intermediate and high location. The distal loopogram or distal colostrography / colostrogram examination is an examination that is very important to determine the location of the distal rectum before definitive repair, but this examination has the disadvantage of using contrast and x-ray radiation. One alternative radiological examination that can be used for faster initial management is sonographic.[7]

Methods: This study was a diagnostic test study to assess the diagnostic accuracy of the distal loopogram / colostrography examination and sonographic examination with the transperineal approachon post-colostomy Anorectal Malformations patients at Mohammad Hoesin Hospital, Palembang in August 2019-October 20120 or until the number of research samples is met.

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Results: There were 21 samples in this study, 11 (52.4%) samples were dominated by clinical appearance without fistula. On the results of ultrasound examination, the majority of samples of rectouretral fistulas were 7 (33.3%). Loopogram examination found the majority of samples without fistulas as many as 10 (47.6%) samples. On intraoperative examination, 7 (33.3%) samples were obtained for rectouretral fistulas. The result of pouch-perineum measurement on ultrasound examination was 1.15 ± 0.6 cm, loopogram was 0.7 ± 1.5 cm, intra-operasionem was 1.8 ± 1.6 cm. In this study, USG has a very high accuracy value with a sensitivity of 100% and a specificity of 93.8% while the Loopogram has a sensitivity of 100% and a specificity of 68.8%.

Conclusion: Transperineal ultrasound has a very high accuracy value with a sensitivity of 100% and a specificity of 93.8% while the Loopogram has a sensitivity of 100% and a specificity of 68.8%. Although there is a significant difference between the distal rectum -perineal distance calculation with a difference of -0.38 ± 0.70 cm, these two methods are equally accurate in assessing the distal rectal pouch -perineal distance and the location of the fistula.

Keyword: transperineal sonography, distal loopogram, intra operatif, anorectal malformation.

1. Introduction

Imperforated anus is a congenital abnormality which is better known as anorectal malformation. The incidence of anorectal malformations is 1 in 4000 to 5000 live births. The incidence of anorectal malformations in Europe varies from 1.14 to 5.96 per 10.000 people and can change every year.[1-3] Anorectal malformations 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 very small and easily treatable with an excellent functional prognosis, to complex, difficult to manage, often associated with other anomalies, and have a poor functional prognosis.[1-3]

The results of research conducted at Sardjito Hospital by Pratomo in 1998-2003 reported that the ratio between male and female anorectal malformation patients was 21:19. Of the 93 cases, there were an estimated 34 cases of anorectal malformation each year. Most patients with anorectal malformations were male with a ratio of 23:14 cases.[4-5]

Delay in the treatment of anorectal malformations can increase morbidity and mortality. Beudeker in 2013 reported that the mortality rate of malformations resulted from complications in the form of bowel perforation and postoperative septic complications in newborns and an increase in the most common morbidity such as constipation to severe such as fecal and urinary incontinence.

Rapid assessment and diagnostic accuracy are important in determining the course of action. The surgical approach in the management of anorectal malformations depends on the classification of anorectal malformations based on low, intermediate and high location. Examination of the distal loopogram or distal colostrography / colostrogram is an examination that is very important to determine the location of the distal rectum before definitive repair. Distal colostrography with augmented pressure should be performed in all male and all women patients undergoing colostomy with cloaca. This examination provides information regarding the location of the fistula between the rectum and the genitourinary, the length of the colon available from the colostomy to the fistula site, the distance between the rectum and the anal dimple, and the rectum to the sacrum. However, there are major drawbacks to using ionizing contrast under fluoroscopy or still using X-ray imaging. The exposure of young infants to radiation continues to cause concern. Although the absolute risk of radiation exposure from this procedure is minimal, there remains an even higher risk of cancer-related radiation injury in exposed children compared to adults. This is because children are more sensitive to radiation than adults because they are still growing and more actively dividing cells; their long life expectancy also increases the risk of radiation injury manifestations. In addition, the adjustment of radiation machines that do not fit their small body size can cause high radiation doses with the resulting cellular damage. One alternative radiological examination that can be used for faster initial management is sonographic examination.[1,7]

Takahiro et al in 2016 stated that the sonographic diagnostic accuracy of low-type imperforate anus based on pouch-perineal distance and fistula location was better than cross-table radiographs. If the pouch-perineal distance on the cross-table radiograph is greater than 10 mm, sonographic examination to determine the location of the fistula may be recommended. Hans et al in 2007 also stated that transperineal sonography is feasible for all children without special preparation. The mean distance between the distal rectal sac and perineum in 22 infants with low imperforate anus was 10 ± 4 (SD) mm compared with a mean of 24 ± 6 mm in 34 infants with moderate or high anomalies ($p < 0.001$). Transperineal sonographic sensitivity is 100%; All 34 cases of moderate or high imperforate anus were identified by a cutoff distance between the distal rectal sac and perineum of 15 mm. Hans also found sonography to have a specificity of 86% and an accuracy of 95% .[8-10]

The importance of accurate investigations for initial management to prevent complications led the investigators to asses the diagnostic accuracy of distal loopogram and sonographic examinations with the transperineal approach to anorectal malformations.

2. Methods

This study was a diagnostic test study to assess the diagnostic accuracy of the distal loopogram / colostrography examination and the sonographic examination with the transperineal approach to post colostomy Anorectal Malformations at Mohammad Hoesin Hospital, Palembang. This research was conducted in the Pediatric Surgery subdivision of the Department of Surgery when the study was conducted in August 2019- October 2020 or until the number of research samples was met.

The study population was all patients who were subjected to distal loopogram / colostrography examination and sonographic examination with the transperineal approach to post colostomy anorectal malformations who were treated in the Pediatric Surgery subdivision of the Department of Surgery RSMH Palembang and The research sample was all patients with post colostomy anorectal malformations who had met the inclusion criteria.

The inclusion criteria were all post colostomy Anorectal Malformations patient, had never been manipulated, and were willing to participate in the study and signed an informed concern sheet. Exclusion criteria were patients who had PSARP operation and were not willing to be sampled.

Based on the calculation of the sample size with predetermined limits, the minimum sample size was 34 patients. If add the possibility of dropping out of 10%,so get a sample numbers with rounds of 37 patients.

The Gold Standard variable in this study was the intraoperative result and the predictor variable was the transperineal ultrasound examination and the Distal Loopogram examination. Data were presented in 2x2 table form and analyzed using SPSS version 21 program.

3. Result

General Characteristics

Based on gender, there were 10 male and 11 female. The mean age in months was 13 months, the youngest was 3 months and the oldest was 70 months.

Table 1. Clinical Characteristics

Parameter	Clinical Characteristics (n=21)				
	Without Fistula	**Recto Vestibular Fistula**	**Perineal Fistula**	**Rectouretral Fistula**	**Cloaca**
Anorectal malformation	11(52.4%)	4(19%)	2(9.5%)	4(19%)	-
Transperineal Sonography	6(28.6%)	5(23.8%)	3(14.3%)	7(33.3%)	-
Loopogram	10 (47.6%)	3(14.3%)	1(4.8%)	5(23.8%)	2(9.5%)
Intra Operasionem	5(23.8%)	3(14.3%)	4(19%)	7(33.3%)	2(9.5%)

Table 2. Results of Measurement of Pouch-perineum

Parameter	Differences distance
Transperineal sonography	1.15 ± 0.6 cm
Loopogram	0.7 ± 1.5 cm
Intra operasionem	1.8 ± 1.6 cm
Transperineal sonography and Loopogram	0.38 ± 0.7 cm
Transperineal sonography and Intra Operasionem	0.6 ± 1.5 cm
Loopogram and Intra Operasionem	0.2 ± 1.2 cm

Table 3. Correlation of Rectal Pouch Distance Measurement and Fistula Location

	Transperineal Sonography		Loopogram		Intra Operasionem	
	R	P	R	p	r	P
Transperineal Sonography			0.474	0.030	0.684	0.001
Loopogram	0.474	0.030			0.839	0.000
Intra Operasionem	0.684	0.001	0.839	0.000		

*Spearman's rho correlation test, p value means if p <0.05, r value is very weak if r <0.2, weak if r = 0.21-0.4, middle if r = 0.41-0.6, strong if r = 0.61-0.8 and very strong if r > 0.8

Table 4. Comparison of Transperineal Sonography. Distal Loopogram and Intraoperationem

		Paired Differences					t	df	Sig. (2-tailed)
		Mean	Std. Deviation	Std. Error Mean	95% Confidence Interval of the Difference				
					Lower	Upper			
Pair 1	USG – Loopogram	-0.38	0.70	0.15	-0.70	-0.06	-2.5	20	0.020
Pair 2	USG – intra operasionem	-0.65	1.53	0.33	-1.35	0.04	-1.9	20	0.064
Pair 3	Loopogram - intra operasionem	-0.26	1.24	0.27	-0.83	0.29	-0.9	20	0.336

* Paired T test p value is significant if p > 0.05

Table 5. The accuracy of transperineal ultrasound results and loopogram in assessing fistulas compared to Intraoperative as a Gold Standard

	Intra Operasionem		**P**
	Without Fistula	With Fistula	
Transperineal Sonography			0.000
Without Fistula	5(100%)	1(6.3%)	
With Fistula	0(0%)	15 (93.8%)	
Loopogram			
Without Fistula	5(100%)	5(31.3%)	0.001
With Fistula	0(0%)	11(68.8%)	

Table 6. Diagnostic Accuracy Transperineal Ultrasound and Loopogram

Accuracy	**Anorectal Malformation Post Colostomi**	
	Transperineal sonography	Loopogram
Sensitivity	100%	100%
Spesificity	93.8%	68.8%
Positive predictive value	83.3%	50%%
Negative predictive value	93.8%	100%
Positive false value	16.7%	50%
Negative false value	0%	0%

Table 7. Transperineal sonography and Loopogram suitability

	Loopogram		P	Kappa
	Without Fistula	With Fistula		
Transperineal Sonography			0.000	0.611
Without Fistula	6(100%)	4(40%)		
With Fistula	0(0 %)	11 (73.3%)		

Kappa test. p value is significant if p <0.05. The Kappa value is very weak if <0.2 . weak if 0.21-0.4. moderate if 0.41-0.6. strong if 0.61-0.8 and very strong if > 0.8

4. Discussion

In this study, Transperineal Sonography has a very high accuracy value with a sensitivity of 100% and a specificity of 93.8% while the Loopogram has a sensitivity of 100% and a specificity of 68.8%. Although there is a significant difference between the distal rectum-perineal distance calculation with a difference of -0.38 ± 0.70 cm, these two methods are equally accurate in assessing the distal rectum and fistula distances. Examination of the distal loopogram or distal colostrography / colostrogram is an examination that is very important to determine the location of the distal rectum before definitive repair. Distal colostrography with augmented pressure should be performed in all male and all women patients undergoing colostomy with cloaca. This examination provides information regarding the location of the fistula between the rectum and the genitourinary, the length of the colon available from the colostomy to the fistula site, the distance between the rectum and the anal dimple, and the rectum to the sacrum. However, there are major drawbacks to using ionizing contrast under fluoroscopy or still using X-ray imaging.[1-7]

Takahiro et al in 2016 stated that the sonographic diagnostic accuracy of low-type imperforate anus based on pouch-perineal distance and fistula location was better than cross-table radiographs. If the pouch-perineal distance on the cross-table radiograph is greater than 10 mm, sonographic examination to determine the location of the fistula may be recommended. Hans et al

in 2007 also stated that transperineal sonography is feasible for all children without special preparation. The mean distance between the distal rectal sac and perineum in 22 infants with low imperforate anus was 10 ± 4 (SD) mm compared with a mean of 24 ± 6 mm in 34 infants with moderate or high anomalies ($p < 0.001$). Transperineal sonographic sensitivity is 100%; All 34 cases of moderate or high imperforate anus were identified by a cutoff distance between the distal rectal sac and perineum of 15 mm. Hans also found sonography to have a specificity of 86% and an accuracy of 95% .[8-10]

Ekwunife,Okechukwuhyginus, et al (2017) compared distal loopography and transperineal ultrasound in patients with colostomy anorectal malformations. There are 13 infant, 9 male and 4 female. Age 2-12 months with a median age of 9 months. Using a t-test confidence interval of 95%, the P value when compared with the distal loopogram was 0.19 and the ultrasound with intraoperative was 0.06. Ability to detect the presence / absence of fistulas; USG has a sensitivity of 50%, a specificity of 100%, an accuracy of 69.2%, a negative predictive value of 55.6% and a positive predictive value of 100%.[7]

Complex genitourinary anomalies that can replace pouches can also be quickly recognized. Ultrasound carries no radiation risk and is not as expensive as an MRI or CT scan. Using ultrasound, internal fistulas can be identified as well. Donaldson evaluated 18 children with imperforate anus using ultrasound mostly using a suprapubic approach, and in some cases via the perineum. Ultrasonography correctly predicted pocket location in all 12 children who had pocket-level confirmation surgically or a distal colostogram. However, definitive surgery to confirm the placement of the pouch was performed only in one of the 7 cases with a P-P distance of more than 15 mm. In the study of Alehossein, et al, all examinations were carried out with a transperineal approach. Of our 23 cases, five children, the pouch to perineum (P-P) distance was less than 10mm. All of these low lesions were safely treated by a simple perineal anoplasty (minimal posterior sagittal anorectoplasty; i.e. minimal PSARP) and were confirmed as low type. Seven children had P-P distance of 10-15 mm. In the follow up definitive surgery, 5 cases were intermediate and two cases were high. Eleven children had a P-P distance of more than 15 mm, of which ten cases were high lesions and had colostomy at birth. During follow up, 8 cases underwent definitive surgery

of PSARP and two of these high cases were confirmed by distal colostogram and one case was categorized as intermediate by definitive PSARP. Measure of agreement (Kappa) was calculated to be 0.791 (P = 0.001) Therefore, Ultrasound correctly predicted the level of the distal pouch in 20 of 23 patients.[9,20]

5. Conclusion

Transperineal ultrasound has a very high accuracy value with a sensitivity of 100% and a specificity of 93.8% while the Loopogram has a sensitivity of 100% and a specificity of 68.8%. Although there is a significant difference between the distal rectum -perineal distance calculation with a difference of -0.38 ± 0.70 cm, these two methods are equally accurate in assessing the distal rectal pouch -perineal distance and the location of the fistula.

6. References

1. Pena A., Levitt M. 2012. Anorectal malformations. In: A.G. Coran (Ed.): Pediatric Surgery 7th ed - Vol 1. Elseviersurgery: Mosby, Inc, New York pp: 1289- 310.
2. Moore, S W, et al. “Anorectal Malformations in Africa.” South African Journal of Surgery, South African Journal of Surgery, Nov. 2005, www.ajol.info/index.php/sajs/article/view/34352.
3. Levitt, Marc A, and Alberto Peña. “Anorectal Malformations.” Orphanet Journal of Rare Diseases, vol. 2, no. 1, 2007, doi:10.1186/1750-1172-2-33.
4. Pratomo S. Evaluasi posterosagittal anorectoplaty pada atresia ani di RSUP Dr.Sardjito Yogyakarta 1998 – 2003. Universitas Gajah Mada. 2003.
5. Kurniawan, W. Gambaran pasien malformasi anorektal di bangsal bedah RSUD Arifin Achmad Provinsi Riau periode Januari 2007 – Desember 2009. Universitas Riau. 2009.
6. Beudeker N, Broadis E, Borgstein E, Heij HA. The hidden mortality of imperforate anus. Afr J Paediatr Surg. 2013 Oct-Dec. 10(4):302-6. [Medline].

7. Ekwunife, Okechukwuhyginus, et al. "Comparison of Trans-Perineal Ultrasound-Guided Pressure Augmented Saline Colostomy Distension Study and Conventional Contrast Radiographic Colostography in Children with Anorectal Malformation." African Journal of Paediatric Surgery, vol. 13, no. 1, 2016, p. 26., doi:10.4103/0189-6725.181703.
8. Hosokawa, Takahiro, et al. "Comparison of Diagnostic Accuracy for the Low-Type Imperforate Anus Between Prone Cross-Table Radiography and Sonography." Journal of Ultrasound in Medicine, vol. 36, no. 8, 2017, pp. 1679–1686., doi:10.7863/ultra.16.07048.
9. Donaldson JS, Black CT, Reynolds M, Sherman JO, Shkolnik A. Ultrasound of the distal pouch in infants with imperforate anus. J Pediatr Surg 1989; 24:465–468.
10. Haber, Hans P., et al. "Transperineal Sonography for Determination of the Type of Imperforate Anus." *American Journal of Roentgenology*, vol. 189, no. 6, 2007, pp. 1525–1529., doi:10.2214/ajr.07.2468.
11. Moore S. Genetics, *Pathogenesis and Epidemiology of Anorectal Malformation and Caudal Regression Syndrome*. In: Holscheneider AM, Hutson JM, editors. *Anorectal Malformation in Children*. New York: Springer 2006;3: 48-64
12. Texas Departement of State Health Services. *Birth Defect Epidemiology and Surveillance: Anal atresia/stenosis*. Texas 2007. Diakses Juni 2017
13. Levit MA, Pena A. *Complication of Surgery for Anorectal Malformations*. In: Caty MG, Glick P, Levit MA, Haynes JH, editors. *Complication in Pediatric Surgery*. New York: Informa 2008;17:299-316
14. Paidas C, Levit MA, Pena A. *Rectum and Anus*. In: oldham, Keith T, Colombani, Paul M, Robert et al, editors. *Principles and Practice of Pediatric Surgery, 4th Edition*. Baltimore: Lippincot Williams & Wilkins.2005;89:1395-1438
15. Nichol, Petter F, et al. *Fundamentals of Pediatric Surgery*. 2nd ed., Springer, 2018.
16. Chouikh, Taieb. Pediatric Surgery Handbook for Residents and Medical Students. Nova Biomedical, 2017.

17. Westgarth-Taylor, Christopher, et al. “Imaging in Anorectal Malformations: What Does the Surgeon Need to Know?” South African Journal of Radiology, vol. 19, no. 2, 2015, doi:10.4102/sajr.v19i2.903.
18. Rahalkar, Mukundd, et al. “Pictorial Essay: Distal Colostography.” Indian Journal of Radiology and Imaging, vol. 20, no. 2, 2010, p. 122., doi:10.4103/0971-3026.63054.
19. Levit MA, Pena A. *Imperforate Anus and Cloacal Malformation*. In: Holcomb III GW, Murphy PJ, editors. *Ashcraft’s Pediatric Surgery, 6th Edition*. Philadelphia: Saunders Elsevier 2014;35:492-514
20. Alehossein, M, et al. *Validity of Transperineal Ultrasound in Infants with Imperforate Anus.* Iran Journal of Radiology, Dec. 2004, pp. 43–46.
21. Hosokawa T,Tanami Y, Tamada Y et al. Sonography fo An Imperforate Anus. J Ultrasound Med 2017; 36:1747–1758