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ORIGINAL RESEARCH

Accuracy of Lateral Invertogram in diagnosing and classifying anorectal malformations

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Abstract

Background: The initial management of a new born baby with anorectal malformation could be made only after an accurate determination of the level of the anomaly.

Objective: To determine the accuracy of lateral invertogram in diagnosing and classifying anorectal anomalies.

Methods: This was a retrospective study carried out between January 2003 and December 2013. The accuracy, sensitivity, and specificity of lateral invertogram in diagnosing and classifying anorectal malformations in the cases studied were determined.

Results: A total of 62 children with anorectal malformations patients were seen during the study period. The age range was 4 hours to 6 years. A total of 50 (80.6%) had lateral invertogram while only 39 films of lateral invertogram were available for analysis. A total of 22 patients had high anomalies while 12 patients had low anomalies. The accuracy of lateral invertogram in identifying anorectal anomaly was 100% and its ability to differentiate between high and low anomalies was 76.5%. The Sensitivity in identifying high anomalies was 59% while the specificity was 31.8%. The sensitivity in identifying low lying lesions was 66.5% while the specificity was 50%.

Conclusion: The accuracy, sensitivity, and specificity of lateral invertogramin diagnosing and classifying anorectal malformations were found to be satisfactory in the present study.

Keywords: Accuracy, Anorectal malformations, Imperforate anus, Lateral Invertogram, Neonatal intestinal obstruction.

Introduction

The incidence of anorectal malformations is 1 in 5000 globally. They are more common in males than compared to females. The diagnosis is usually made shortly after birth and clinical evidence of anorectal malformations could be found in 80-90% of cases while about 10-20% remains doubtful. In the doubtful cases, the internal anatomy could be assessed using imaging techniques such as the invertogram radiology, erect plain abdominal radiography, ultrasound examination of the abdomen, pelvis, and spine, computed tomography (CT), micturating cystourethrogram, distal colostogram, magnetic resonance imaging (MRI) and muscle stimulation under general anaesthesia. [1-4]

Using the invertogram, the bowel-skin distance can be estimated and based on the findings, anorectal malformations could be divided into high and low types, depending on whether the termination of the hindgut is above or below the puborectalis sling. More than 1cm bowel-skin distance indicates a high anomaly while a bowel-skin distance of 1cm or less indicates a low anomaly. [5]

Though the lateral invertogram serves as an important modality of investigation, it could be associated with aspiration pneumonitis or cerebral congestion; the latter may arise while performing the procedure, as a result of positioning the baby in an inverted state. Also, the invertogram could be limited by its inability to accurately differentiate between the high and low types of anomalies as reported by some authors. [5,6]

The classification of anorectal malformations is important because it helps in planning and choosing the appropriate methods of treatment and also, provides a guide for prognosis. The earlier methods of classification of the malformations such as the Wingspread classification and the international classification did not standardize the indications and choices of intervention; however, Pena's classification includes some indications aimed at standardising the choices and types of treatment available to the patients. [7, 8]

The objective of this study was to determine the accuracy, sensitivity, and specificity of lateral invertogram in the diagnosis of anorectal malformations and to ascertain the role the lateral invertogram plays in the classification and definitive treatment of patients with anorectal malformations.

Methods

This was a retrospective study of the use of lateral invertogram in anorectal malformations for the period between January 2003 and December 2013 at the Olabisi Onabanjo University Teaching Hospital, Sagamu.

The hospital records of patients managed for anorectal malformations during the study period were retrieved from the Medical Records Department. The invertograms were done not less than 24 hours after birth; this was to allow the baby's swallowed air to reach the most distal part of the rectum. Relevant data such as age, sex, clinical and radiographic (lateral invertogram) findings were extracted from the hospital records. The anorectal malformation was classified as low-lying when the distance between the distal gas shadow in the rectal pouch and a radiopaque marker at the perineum (anal dimple) is about 1cm or less. High anorectal malformations were characterized by distances of more than 1cm. All the patients included in this study had lateral invertograms as the diagnostic modality following clinical suspicion or identification of an anorectal anomaly.

The gold standard against which the lateral invertogram was tested was intra-operative findings. The accuracy, sensitivity, and specificity of lateral invertogram were calculated using the following formulae:

   a. True positives were actual positives with anorectal malformations and those who had high or low malformations and were correctly identified through the lateral invertogram.
   b. True negatives were the actual negatives and were identified accordingly. These were proportions without anorectal malformations, those without high malformations who were accordingly identified and those without low malformations who were also accordingly identified.
   c. False positives were those who were tested to have the condition but at surgery, were found not to have anorectal malformations and cases

pre-operatively classified as high and low malformations, but with reverse findings at surgery.

d. False negatives were those which tested negative pre-operatively but were found to have the anorectal malformations at the surgery. These refer to the presence of anorectal malformations with high or low malformations which were correctly identified at surgery but missed pre-operatively.

Accuracy = (True positive + True negative)/(True positive+True negative+False positive+False negative)

Sensitivity = (True positive)/(True positive+False positive)

Specificity = (True negative)/(True negative +False positive)

The data obtained were analysed using SPSS version 22 and the results presented as frequencies and proportions.

Ethical considerations
Ethical approval for this study was obtained from the Health Research Ethics Committee (HREC) of the Olabisi Onabanjo University Teaching Hospital, Sagamu.

Results

A total of 62 children aged 4 hours to 6 years with anorectal malformations were seen during the study period. The subjects comprised 41 males and 21 females, giving a male-to-female ratio of approximately 2:1. Fifty children (80.6%) children had lateral invertogram while 12 (19.4%) did not have lateral invertogram and were not included in the data analysis.

Thirty-nine radiographic films of lateral invertogram were retrieved, 5 were of poor quality and excluded from further analysis. The remaining 34 radiographic films showed 22 children with high anomalies and 12 children with low anomalies.

Further findings from the 34 radiographic films showed an accuracy of 100% while its ability to differentiate between high and low anomalies was 76.5%. The Sensitivity of lateral invertogram in identifying high anomalies was 59% while the specificity was 31.8%. The sensitivity of the lateral invertogram for identifying low lying lesions was 66.5% while the specificity was 50%.

Figures 1 to 4 show the clinical and radiographic (lateral invertogram) manifestations of anorectal malformations in the newborn.

Discussion

Anorectal anomalies represent a wide spectrum of congenital malformations of the lower gastrointestinal tract (the rectum and anal canal) and may be extended to malformations and abnormal connections with the genitourinary tract. It may occur alone or in association with other life-threatening congenital anomalies involving the heart, vertebra and the musculoskeletal system (the VACTERAL syndrome). The outward appearance of the anorectum and perineum may not give a true picture of the complexity of anomalies affecting the anorectal region, neurovascular tissue, spinal cord, vertebra, and the genitourinary system. [9,10]

The diagnosis of anorectal malformation is made in 80% of cases through physical examination. However, in about 20% of cases, radiological studies would be required for the confirmation of the diagnosis. Radiological studies also contribute immensely towards classifying the malformations and planning for the ultimate care of these patients. The traditional radiological approach involves the

use of an invertogram to determine the level of the distal rectal pouch; the information obtained from the invertogram report is useful in determining whether the anorectal malformations would be safely repaired from the perineum or whether a colostomy would be needed. [11,12]

Figure 1: Lateral invertogram showing low-lying anomalies

Figure 2: Lateral invertogram showing the high type of anorectal anomalies

Figure 3: Gross appearances of the perineum of newborns with anorectal malformations

Figure 4: Anorectal malformation manifesting with neonatal intestinal obstruction

In our experience, the first investigative approach is the lateral invertogram and it has remained invaluable in the diagnosis and classification of anorectal malformations in our centre. It is very sensitive and specific in confirming the diagnosis and classifying anorectal anomalies. The results in the present study are similar to the findings of Akansha [13] and Horsirimanont [14] and their co-workers. Other investigations employed in the management of the cases included abdominal and perineal ultrasonography, plain abdominal radiography, chest radiography, and echocardiography as adjuncts to the lateral invertogram when indicated. None of the children in the present series had abdominal computerized tomography and magnetic resonance imaging for clinical evaluation. [13, 14]

In the study by Akansha and co-workers, [13] 33% of their patients with anorectal malformations were diagnosed with lateral invertogram while 20% were diagnosed with prone cross-table lateral radiography and 20% diagnosed with plain abdominal erect radiography. They also employed micturating cystourethrogram to diagnose some cases of recto-vesical fistula. [13] On the other hand, Horsirimanont and co-workers [14] reported a sensitivity of 66.7% for lateral invertogram in Thailand; the low sensitivity may be attributed to the limitations inherent in the procedure of lateral invertogram. [14]

The invertogram is a radiographic technique used for estimating the level of the rectal pouch in patients with anorectal malformations. Although the technique has been employed for the diagnosis of anorectal anomalies in the last eighty years, it is still an investigation of choice for neonatal evaluation of anorectal malformations. It has been considered the classic method for determining the distance from the blind rectal pouch to a marker placed on or within the anal dimple. The decision to use an invertogram to assess the level of the rectal pouch should be taken at least 24 hours after birth. The baby is carefully held upside down for at least 3 minutes during the procedure. Invertogram has been considered to be the most important early diagnostic modality in identifying and classifying newborns with anorectal malformations. [15,16]

Despite the usefulness of the lateral invertogram in the initial assessment and categorization of patients with anorectal malformations, it is associated with limitations such as difficulty in achieving optimum position, the inability of air to reach the end of the bowel due to impacted meconium, continuous crying during the procedure which leads to deceptive obliteration of lower rectum, significant hypoxia due to inverted position in babies with the imperforate anus and intestinal obstruction, upward displacement of the rectum due to the effect of gravity and escape of air through a fistulous tract which causes less distension of the blind-ending rectal pouch. [15-17] In addition, some of our patients presented late and could not be subjected to the lateral invertogram, some films were of poor quality and were not included in this study while others could not be retrieved at all. These are acceptable limitations of a retrospective study.

An alternative radiological procedure (prone cross-table lateral radiograph) could be used; this is an X-ray taken in the knee-chest position with the baby positioned prone, the buttocks elevated to about 45° and the x-ray taken laterally and centered at the greater trochanter. It provides equal or sometimes better information compared to the invertogram, in the demonstration of the level of rectal atresia. Its advantages include easy positioning, the better co-operation of the patient, elimination of the effect of gravity and better delineation of the rectal gas shadow. [18]

To improve the diagnostic accuracy of the lateral invertogram and lateral prone cross-table X-ray in diagnosing anorectal anomalies

and other associated congenital malformations, several investigative adjuncts have been employed. These newer diagnostic techniques include abdominal ultrasonography. The use of ultrasound to determine the position of the terminal colon is well known; initial reports involved scanning from the anal dimple and measuring the distance between the terminal colon and the skin at the anal dimple site. However, the application of pressure at the anal dimple distorts the distance between the anal dimple and the terminal colon. [19-21]

The computerized tomographic scan provides increased detail, particularly of the osseous structures as well as being able to demonstrate the pelvic floor musculature but has difficulty in distinguishing meconium from the rectal wall and adjacent musculature due to limitation in contrast resolution. There is also a risk of exposure to radiation in the use of a computerized tomography scan.[22] Magnetic resonance imaging has similar properties as CT but has improved contrast resolution which allows for easy differentiation of meconium from the rectal wall and levator musculature. MRI can accurately detect the level of the terminal bowel and the state of the pelvic floor musculature, and also, can demonstrate the presence of a fistula. [23]

The surgical management of an infant with anorectal malformation depends on the level of the anomaly as well as the presence or absence of a fistula between the rectal pouch and adjacent organs and other coexisting congenital malformations. Newborn primary anoplasty is the surgical method of choice for low-lying anomalies, while temporary colostomy is usually chosen for high-lying anomalies. [12,13,24]

Conclusion

Lateral invertogram is a useful diagnostic modality employed in the immediate evaluation and care of newborns with anorectal malformations. Its accuracy, sensitivity, and specificity in diagnosing and classifying these conditions were found to be satisfactory in the present study. It is attractive to recommend the routine use of lateral invertogram and lateral cross-table decubitus radiograph in the initial diagnosis and classification of anorectal malformations in children. The use of other diagnostic adjuncts such as abdominopelvic ultrasonography, computerized tomographic scan, magnetic resonance imaging and techniques for pelvic muscle stimulation improves the diagnostic accuracy, classification of these anomalies and identification of other associated congenital malformations.

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