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Case Report

# Anorectal malformation in an adult: A case report

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**Abstract**
*Background:* Anorectal malformations (ARM) are usually diagnosed at birth, although, some patients have presented in the hospital beyond the newborn period without recognition of the anomaly. Late presentation in adulthood has also been reported. We report a case of adult ARM whose mother was instructed not to present in the hospital until she was old enough.
*Method:* An adult female patient was evaluated and investigated preoperatively for high ARM with rectovestibular fistula. She had an initial colostomy which was followed by a primary posterior sagittal anorectoplasty (PSARP). Post-operatively, continent level was assessed using the Kelly continent score.
*Results:* She had good post – operative recovery with an episode of faecal soilage. The Kelly continent score was 3 which later improved to 5.
*Conclusion:* Although the sphincteric tone may be weak in the adult patient with ARM with reduced continent level at the initial stage, overall outcome of management of ARM with primary PSARP is good and comparable to outcome in children.

**Keywords:-** *Adult, female, anorectal malformations, primary, continence*

**Résumé**
*Contexte:* Des malformations ano-rectales (ARM) sont généralement diagnostiquées à la naissance, même si, certains patients se sont présentés à l'hôpital au-delà de la période néonatale sans la présence de l'anomalie. La présentation tardive de l'anomalie à l'âge adulte a été également signalée. Nous signalons un cas d'ARM adultes dont on a demandé à la mère de ne pas se présenter à l'hôpital jusqu'à avant sa vieillesse.
*Méthode:* Une patiente adulte a été évalué et soumise à l'étude de manière préopératoire pour une grande ARM avec une fistule recto-vestibulaire. Elle a subi une colostomie initiale qui est suivie d'une anorectoplastie sagittale postérieure primaire (PSARP). En postopératoire, le niveau d'incontinence a été évaluée en se servant du score de « Kelly continent ».
*Résultats:* Elle avait une bonne récupération post - opératoire avec un épisode de saletés fécale. Le score était de 3 Kelly continent qui, plus tard a atteint 5.
*Conclusion:* Bien que le ton sphinctérien soit faible chez le patient adulte souffrant d'ARM avec le faible niveau de continent à l'étape initial, le résultat global de la gestion des ARM avec un bon PSARP primaire et comparable aux résultats obtenus chez les enfants est favorable.

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## Introduction

Anorectal malformations (ARM) are the commonest cause of intestinal obstruction in the newborn [1]. In the developed countries it is commonly diagnosed at birth and treated accordingly. The diagnosis may however be later in many centres in the developing countries where reasons like late presentation, poverty and ignorance about the anomaly as well as how to correct it have been suggested [2,3]. The late presentation is particularly more common with female patients who, besides an abnormal anal opening, may remain asymptomatic [4]. Majority of these anomalies are corrected in infancy [5] using posterior sagittal anorectoplasty (PSARP) a procedure that was first described by Pena *et al* in 1982 [6]. Very few adult cases of ARM have been reported and majority of them were in females presenting with rectovaginal or rectovestibular fistula [4,5] and each of these patients had PSARP as the procedure of choice to repair the anomaly with claims of success. No report has emanated from sub Saharan Africa where late presentation due to poverty and lack of awareness of the existence and management of the anomaly are commonly encountered. We present a case of an adult female patient with successful repair of rectovestibular fistula.

## Case report

A 25 year old unmarried female presented to the surgical outpatient clinic of the University College Hospital, Ibadan, Nigeria with a history of passage of stool from the vagina since birth. This was associated with history of occasional constipation which the patient learnt to control with dietary management. She had no urinary symptoms. Significant findings on examination revealed an absent anus and a fistulous opening in the vestibule

of the vulva. The sacrum and the gluteal cleft were well developed. She had no other associated congenital malformations. An initial colostomy was established with irrigation of the mucous fistula to evacuate retained faecal masses. Further preoperative investigations were done to determine the anatomy and type of ARM. These, however, revealed a high anorectal malformation with a rectovestibular fistula.

The patient underwent a PSARP the following month with good post-operative recovery. Post-operatively, she was commenced on anal dilatation. The colostomy was subsequently closed two weeks after the PSARP. The sphincteric tone was weak and she had an episode of faecal soilage with a Kelly's continent score of 3. This, however, improved as she was able to differentiate between faeces and flatus and she became continent of both subsequently with a Kelly's score of 5. She was discharged from the hospital and has been followed up for a year. At present, she is leading a normal life.

## Discussion

Anorectal malformations are usually diagnosed at birth, but some patients have presented in the hospital beyond the early newborn period without recognition of their anorectal malformations [7].

Others have presented in the adolescent age and adulthood [4,5]. Since 1982 when Pena *et al*,[6] described the procedure of PSARP to repair ARM, it has been used widely to repair variants of ARM and in secondary repair of persistent incontinence in adult patients with ARM with variable reports of success rates [3,8]. The procedure has also been reportedly used successfully in the management of adult patients presenting with ARM [4,5] however, it is important to adhere to the guidelines described by Pena *et al* [6] while performing the procedure on adult patients. Chakravartty *et al* [4] observed that, the vagina is closely adherent to the rectum in females and care must be taking to ensure that the walls of the rectum and vagina are not damaged while paying close attention to the muscles of continence.

In the presence of a well-developed sacrum, normal gluteal cleft with a normal gluteal muscle bulk and no other associated anomalies of the lumbosacral spine, the reported prognosis following PSARP to repair ARM in adult is good [4]. Although, the development of the peri-anal muscles is complete in the adult patients, the presence of a weak sphincteric tone may reduce their continent level. Our patient had an episode of faecal soilage and weak sphincteric tone but these improved with active exercises of the perineal muscles in which she was asked to tighten the anal opening intermittently by contracting these muscles. We conclude that the overall outcome of management of ARM in the adult is good and similar to the outcome in children provided there are no associated anomalies of the lumbosacral spine and careful attention is paid to operative details.

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