Case Report

# Female Anorectal Malformation with Genitourinary Prolapse: A Rare Association

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## Abstract

Genitourinary prolapse in newborn females as an introital mass is an uncommon entity. The usual causative mechanisms are poor pelvic innervation, damage or pressure on pelvic musculature and ligaments etc. Different methods of reduction as treatment were proposed in the past. Apart from uncommon occurrence of genitourinary prolapse in newborns, its association with anorectal malformation is not reported in English literature after searching on PubMed and Google Scholar. We report three cases of genitourinary prolapse with anorectal malformation in newborn females where decompressing colostomy was curative for the condition reflecting increased intra-abdominal pressure as causative mechanism.

**Keywords:** Abdominal distension, colostomy, introital mass, paediatric, urinary retention

## Introduction

A mass protruding in between labia in newborns with anorectal malformation is a cause of concern for the parents because it creates fear of abnormality of genitourinary area, which is worsened when the anal opening is absent or abnormal. In normal newborns, the common differential diagnoses are urethral prolapse, ectopic ureterocele, paraurethral duct cyst, imperforate hymen, rhabdomyosarcoma of vagina etc.[1] The most common congenital anomaly associated with infantile genitourinary prolapse is spinal dysraphism.[2] This is mostly due to defective pelvic innervation. Association of anorectal malformation with genitourinary prolapse as introital mass is very rare and not reported in English literature. We report three cases of female newborns with anorectal malformation who presented to us with abdominal distension and introital mass due to genitourinary prolapse.

## Case Reports

### Case 1

A 5-day-old female was brought to us with complaints of absent anal orifice since birth. Baby was passing stool through abnormal opening at the introital area in small amounts. On clinical examination, general condition of the child was fair, abdomen was moderately distended, genitalia was female type with normal looking clitoris, labia majora and minora, urethral opening and absent anal orifice. Detailed genital examination in the presence of a gynaecologist showed normal urethral opening, prolapsed tissue from vaginal orifice and anal opening below it [Figure 1]. Routine blood investigations like complete blood counts, serum electrolytes and renal function tests were carried out. A high divided sigmoid colostomy was done in view of abdominal distension due to partially decompressing anorectal malformation. The patient was discharged on post-operative day 7 without any complication. In the follow-up period after 3 months, it was observed that the prolapsed vaginal mucosa had reduced spontaneously.

### Case 2

A 5-month-old female presented to us with complaints of passage of stool from abnormal opening in the genital area. On detailed perineal examination, there was an anal orifice in the vestibule and a polypoidal mass protruding from the urethral opening compressing the normal vaginal opening [Figure 2].

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**Figure 1:** Female anorectal malformation with genitourinary prolapse



**Figure 2:** Female anorectal malformation with genitourinary prolapse



**Figure 3:** Female anorectal malformation with genitourinary prolapse

Labia majora and minora appeared normal. Ultrasonography of the abdomen showed dilated rectosigmoid colon and absent/non-visualised right kidney. Micturating cystourethrogram was normal. After routine workup, the patient was worked up for a high divided sigmoid colostomy. During surgery, prolapsed urethral tissue was reduced and per urethral catheterisation done. High divided sigmoid colostomy was performed. Per urethral catheter was removed on day 7. Following this, there was spontaneous reduction of prolapsed urethral tissue. The patient was discharged in stable condition.

### Case 3

A 6-month-old female presented to our outpatient department with features of urinary retention and absent anal opening since birth. On general examination, the patient's condition was poor with gross abdominal distension. The patient was admitted and resuscitation commenced. On local examination of the perineum, there was absent anal orifice and a soft pink prolapsed mass appearing in the introitus [Figure 3]. The patient was catheterised with difficulty. After catheterisation, a small opening was seen in the lower part of the vestibule from where the patient was passing stool. The patient had a high divided sigmoid colostomy after stabilisation. The patient did well in the post-operative period and the mass reduced gradually. Per urethral catheter was removed on post-operative day 5 and the patient was discharged in stable condition.

Posterior sagittal anorectoplasty and finally colostomy closure was done for all three patients during follow-up as standard procedure. There was no prolapse or any introital mass during follow-up.

## Discussion

The occurrence of genitourinary prolapse in newborns is rare.[3,4] The first description was provided in the Ebers papyrus (1500 BC).[5] Findley in 1917 published a series on neonatal genitourinary prolapse having association with spinal cord malformation in 86% of cases.[6] Malpas (1955) classified the causative factor into primary and secondary.[7] The primary causes include congenital abnormality leading to weakness of pelvic floor. The secondary causes include factors producing abnormal stress to normally suspended genitalia. In primary cause, the most common pathology is spinal cord malformation whereas birth trauma, abnormalities of cervix, prolonged labour, especially in breech presentation etc. fall into secondary causes. The aetiology of genitourinary prolapse in spinal cord malformation is due to defective pelvic innervation leading to poor pelvic floor musculature and ligamentous support. It leads to flaccid paralysis of these supports causing downward protrusion of the abdominal and pelvic organs. The diagnosis of this clinical entity is simple, usually with clinical examination only.[8] Treatment of genitourinary prolapse in this age group is varied and includes simple digital repositioning, vaginal plug, pessary placements, temporary labial fusion etc.[9-12]

After rigorous review of English literature on PubMed and Google Scholar, we found that the presence of genitourinary prolapse in cases of anorectal malformation in female newborns had not been reported. The causative mechanism for genitourinary prolapse in these cases of anorectal malformation may fall into primary as well as secondary. Due to known

association of anorectal malformation with spinal cord abnormality, it may fall into the primary group. The causative mechanism may also be included as secondary as increased intra-abdominal pressure due to non-decompressing anal opening in vestibule or vagina leads to downward pressure on suspended genitalia. However, we found that the cause was more likely due to secondary factor because the prolapse was reduced or cured spontaneously after decompressive high divided sigmoid colostomy.

## Conclusion

Genitourinary prolapse in cases of anorectal malformation in female newborns is a rare clinical entity. Causative mechanism may fall into primary, secondary or combination of causes of genitourinary prolapse. It may be a sign of non-decompressing anal orifice and may suggest raised intra-abdominal pressure. Diagnosis is clinical and decompressive colostomy with or without manual repositioning of prolapse is curative.

### Consent

Informed consent taken from parents.

### Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form, the patients' parents have given their consent for the patients' images and other clinical information to be reported in the journal. The patients' parents understand that the patients' name and initials will not be published and due efforts will be made to conceal identity, but anonymity cannot be guaranteed.

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### Conflicts of interest

There are no conflicts of interest.

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