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Case Report
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Recurrent Pelvic Inflammatory Disease and Resistant Balanoposthitis Secondary to a Variant Low Female Anorectal Malformation: An Unusual Cause of Primary Infertility in an Adult Couple

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Abstract

Primary infertility in adult couples is most commonly attributed to ovulatory dysfunction, tubal pathology, or male factor abnormalities. Rarely, congenital Anorectal Malformations (ARMs) persisting into adulthood may create atypical urogenital contamination pathways, predisposing both partners to recurrent genital infections. We report an unusual case of a married couple presenting with recurrent Pelvic Inflammatory Disease (PID) in the female partner and chronic, treatment-resistant balanoposthitis in the male partner. After extensive evaluation, the underlying cause was identified as a previously unrecognised variant low anorectal malformation in the woman, resulting in a concealed recto-vestibular fistulous tract. Surgical correction led to complete resolution of infections in both partners and subsequent spontaneous conception. This case highlights the importance of considering congenital anorectal variants in adults with unexplained recurrent genital infections and infertility.

Keywords: Primary Infertility; Pelvic Inflammatory Disease; Balanoposthitis; Anorectal Malformation; Anovestibular Fistula; Congenital Anomaly; Anterior Sagittal Anorectoplasty; Enteric Contamination; Recurrent Genital Infections; Adult Presentation of ARM

Introduction

Anorectal Malformations (ARMs) represent a spectrum of congenital anomalies affecting the termination of the rectum and anal canal, with presentations ranging from subtle low defects to complex high malformations [1]. International consensus efforts have established classification and management standards, yet low-variant lesions may remain undetected into adulthood due to preserved continence and minimal perineal distortion [2]. Long-term follow-up studies confirm that late-presenting ARMs can manifest with chronic genitourinary or reproductive complications [3], and multicentre registry data highlight the heterogeneity of anatomical variants that may escape early diagnosis [4].

Recurrent Pelvic Inflammatory Disease (PID) remains a major cause of tubal factor infertility worldwide. Contemporary guidelines emphasise the polymicrobial nature of PID and the importance of identifying atypical sources of ascending infection [5]. PID contributes significantly to reproductive morbidity, including tubal occlusion, hydrosalpinx, and chronic pelvic pain [6]. In parallel, sexually transmitted infections and lower genital tract inflammation are recognised contributors to infertility in both women and men [7]. However, the simultaneous occurrence of recurrent PID in a woman and chronic balanoposthitis in her male partner - both driven by a shared anatomical contamination pathway - has rarely been described.

Case Presentation

Female Partner

A 30-year-old woman presented with a 3-year history of primary infertility and recurrent episodes of pelvic inflammatory disease. She reported 5-6 episodes per year, characterised by pelvic

Figure 1: Clinical photograph -Male: Note aftermath of recurrent mixed infections on the glans and the inner foreskin globally in all areas despite several courses of topical and oral antibiotics.

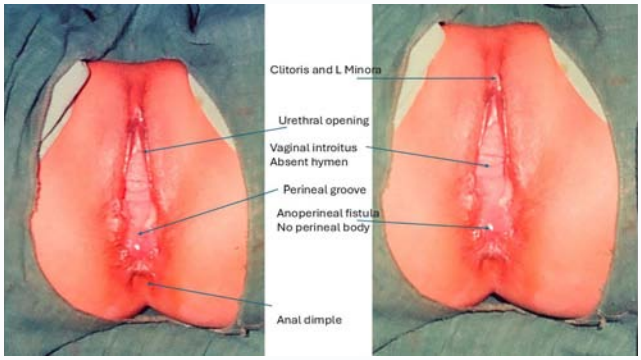

Figure 2: Clinical Photograph-Female: Clinical photoigraphs showing embryopathology.

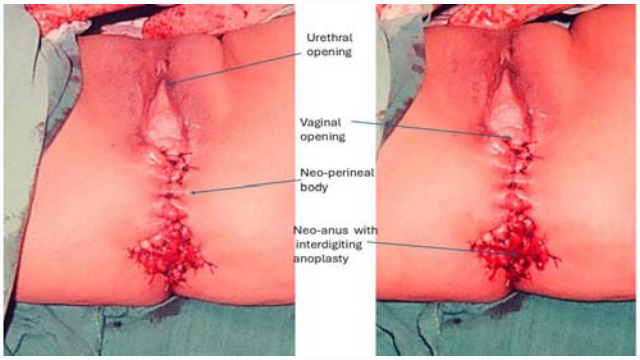

Figure 3: Post surgical correction photograph: Anterior Saggital Anorectoplasty (ASARP) reconstructed all three natural perineal orifices namely urethral, vaginal and anal in their proposed anatomical locations with reconstruction of the strong perineal body all in one stage as primary procedure without staging or diverting stoma.

pain, fever, malodorous discharge, and dyspareunia. Symptoms often recurred shortly after completing antibiotics. She denied bowel symptoms, incontinence, or perineal discomfort.

Menstrual cycles were regular. She had no prior surgeries, history of childhood perineal anal dilatations for a narrow anal opening, and no known sexually transmitted infections. She was in a monogamous marriage.

Male Partner

Her 32-year-old husband reported chronic balanoposthitis for 2.5 years, with erythema, fissuring, and pruritus of the glans and prepuce. Multiple courses of antifungals, topical steroids, and antibiotics provided only transient relief. He had no diabetes, immunosuppression, or dermatological conditions.

Investigations

Female Partner

Inspection revealed an absent perineal body with a narrow bridge of tissue between the vaginal fourchette and an ectopic anal opening (recto-ano-perineal fistula). A mucosal-lined perineal groove was noted extending from the posterior fourchette to the fistula. Perineal examination by specialist pediatric surgical colorectal team demonstrated blind ending anal pit within the anal external sphincter muscle complex, a variant of low anorectal malformation in the form of anoperineal fistula leading to the perineal groove which is the roof of the embryological perineal canal and continuity with vestibule and vagina without any skin bridge or barrier allowing contamination with stool all the time. Slightly anteriorly displaced anal opening and a pinpoint mucosal pit in the posterior vestibule with absent perineal body.

Blood tests were normal inflammatory markers between episodes; negative HIV, syphilis, hepatitis panel. Microbiology with high vaginal swabs repeatedly grew *E. coli* and mixed enteric flora. Transvaginal ultrasound showed mild hydrosalpinx bilaterally.

MRI pelvis depicted narrow fistulous tract (1.2 cm) connecting the distal rectum to the anoperineal fistula and the perineal groove, vestibule and posterior vagina. The findings were consistent with a variant low anorectal malformation with anoperineal fistula and perineal groove.

Male Partner

Erythema and excoriation of the glans penis and inner foreskin in all sectors globally (Figure 1-4); cultures were positive for enteric flora (*E. coli*, Enterococcus). Blood tests demonstrated normal HbA1c, fasting glucose, and immune profile. Dermatology review showed no evidence of lichen sclerosus or dermatoses. Urology review indicated normal genital anatomy; balanoposthitis attributed to chronic exposure to enteric organisms.

Differential diagnosis included recurrent sexually transmitted infections, chronic candidiasis**,** dermatological balanitis**,** tubal factor infertility secondary to PID**,** immunodeficiency and undiagnosed congenital anorectal malformation (confirmed).

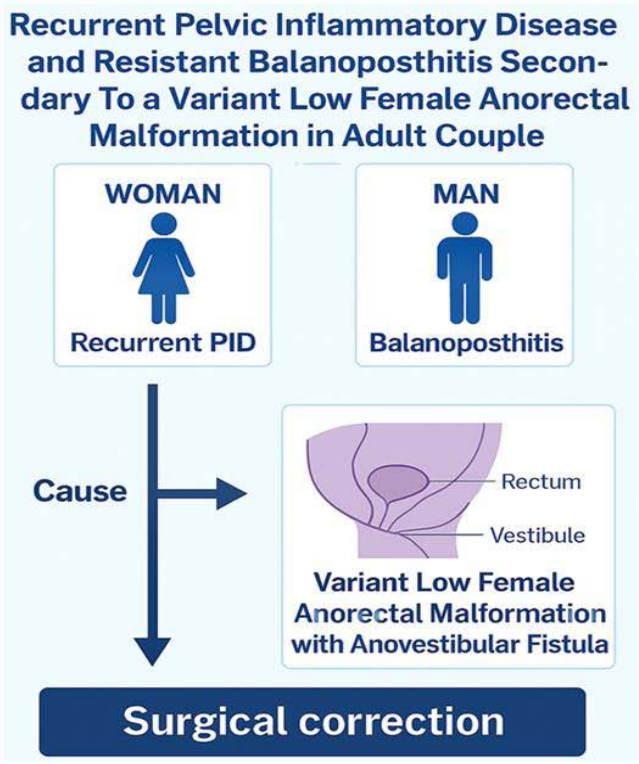

Figure 4: Graphical abstract summarising the clinical pathway: A schematic representation illustrating the dual-partner presentation of recurrent pelvic inflammatory disease in the female partner and resistant balanoposthitis in the male partner. The diagram highlights the underlying cause: a previously unrecognised variant low anorectal malformation with an anovestibular fistula, enabling chronic faecal contamination of the vestibule. Surgical correction is shown as the definitive intervention leading to resolution of infections and restoration of fertility.

Treatment

Female Partner

Surgical correction performed using technique of Anterior Sagittal Anorectoplasty (ASARP) with excision of the fistulous tract, perineal groove and reconstruction of the vagina, vestibule, perineal body and neoanal opening by cruciate interdigiting anastomosis requiring no postoperative anal dilatations and, gave anatomical physiological and cosmetic normal appearance which was very well appreciated by the couple.

Postoperative care included broad-spectrum antibiotics, pelvic floor physiotherapy, and stool-softening regimen.

Male Partner

Targeted antibiotics following partner's surgery with topical barrier therapy and sexual abstinence until postoperative healing was complete led to slow and steady complete recovery.

Outcome and Follow-Up

Both partners experienced complete resolution of infections within 3 months. The male partner's balanoposthitis did not recur. The female partner remained free of PID episodes. The couple achieved spontaneous conception 9 months after surgery. At 18-month follow-up, both remained asymptomatic, well and female had normal vaginal delivery with episiotomy under supervision of our team and the baby is well.

Discussion

Anorectal Malformations (ARM) are typically diagnosed and managed in the neonatal period. This report describes a rare case of a 30-year-old female presenting with primary infertility, recurrent Pelvic Inflammatory Disease (PID), and a spouse with resistant balanoposthitis. Detailed examination revealed a variant of a low female ARM characterized by a rectoperineal fistula, perineal groove, and absent perineal body. This anatomical defect allowed for chronic feco-vaginal contamination, serving as a reservoir for recurrent ascending infections in the female and cross-infection to the male partner. Surgical reconstruction of the perineal body and anoplasty led to the resolution of recurrent infections and successful spontaneous conception, highlighting the importance of thorough perineal inspection in adult infertility workups.

Balanoposthitis is commonly attributed to candidiasis, dermatological conditions, or poor hygiene, but persistent cases associated with enteric flora warrant evaluation for unusual contamination sources [8]. Clinical guidance stresses the importance of identifying underlying structural or infectious drivers when balanitis is recurrent or treatment-resistant [9]. In this case, the male partner's chronic balanoposthitis reflected repeated exposure to enteric organisms originating from the female partner's concealed anorectal anomaly.

This case illustrates a "silent" presentation of ARM in an adult. The ***absent perineal body*** acted as a conduit for enteric pathogens to enter the female reproductive tract, causing recurrent PID - a known major cause of tubal factor infertility. Simultaneously, the male's resistant balanoposthitis was a direct result of exposure to the same feco-vaginal reservoir. Surgeons should maintain a high index of suspicion for anatomical anomalies when faced with "global" recurrent genital infections in a couple.

While low-type ARMs such as rectoperineal fistulas are the most common variants in females, they may remain undiagnosed until adulthood in resource-limited settings or when symptoms are mild. Perineal grooves - a rare midline malformation - often coexist with ARMs and can be mistaken for simple dermatitis or trauma. The absence of a perineal body significantly reduces the distance between the anal and vaginal orifices, predisposing the couple to a cycle of polymicrobial infections that can impair fertility via tubal damage in the female and chronic urethral/glans irritation in the male.

Anovestibular fistula is a recognised subtype of low ARM, typically diagnosed in infancy; however, delayed presentation into adolescence or adulthood has been documented, particularly in settings where neonatal perineal examination may be limited [10]. Posterior Sagittal Anorectoplasty (PSARP) remains the gold-standard corrective procedure, offering excellent anatomical restoration when the fistulous tract is clearly delineated [11]. Reports of delayed ARM diagnosis emphasise the diagnostic challenges posed by subtle perineal findings and preserved continence [12].

Recurrent PID in this patient was driven by chronic vestibular contamination with enteric organisms, consistent with established pathophysiological models of ascending infection [13]. Subclinical or recurrent lower genital tract inflammation is known to contribute to tubal damage and infertility, even in the absence of classical PID symptoms [14]. The male partner's recurrent balanoposthitis further supports the concept of a shared contamination source, aligning with evidence that genitourinary infections can impair male reproductive

health [15].

This case underscores the importance of considering congenital anorectal variants in adults with unexplained, recurrent genital infections - particularly when both partners are affected. Surgical correction of the underlying malformation resulted in complete resolution of infections and restoration of fertility, demonstrating the profound reproductive implications of timely diagnosis.

This case describes a uniquely challenging presentation of primary infertility in an adult couple, in which both partners experienced recurrent genital infections - pelvic inflammatory disease in the female partner and resistant balanoposthitis in the male partner. After extensive multidisciplinary evaluation, the underlying cause was identified as a previously unrecognised variant low anorectal malformation with an anovestibular fistula, resulting in chronic faecal contamination of the vestibule. Surgical correction led to complete resolution of infections in both partners and ultimately to spontaneous conception.

We believe this case is of significant clinical value because adult presentation of low anorectal malformations is rare and often overlooked. The dual-partner infectious pattern provides an important diagnostic clue not widely recognised in the literature. The case highlights the importance of considering congenital structural anomalies in recurrent, treatment-resistant genital infections. Surgical correction restored reproductive potential, underscoring the impact of timely diagnosis.

We hope this case will contribute meaningfully to clinical awareness, diagnostic reasoning, and multidisciplinary management of unexplained infertility and recurrent genital infections.

This case illustrates a rare but clinically significant cause of recurrent genital infections and infertility. Low anorectal malformations with ano-vestibular fistula may present subtly, especially when the anal opening is functional and continence is preserved. In such cases, the fistulous tract allows intermittent leakage of faecal material into the vestibule, promoting chronic colonisation with enteric organisms.

Learning Points

- Low ARMs may remain undiagnosed into adulthood, especially in regions with limited neonatal screening.
- Adult presentation of low female ARMs can manifest as primary infertility rather than primary bowel complaints.
- Chronic feco-vaginal contamination due to an absent perineal body can cause recurrent PID and resistant balanoposthitis in the partner. Dual-partner presentation (PID + balanoposthitis). Simultaneous genital infections in both partners strongly suggest a shared source of contamination.
- Consider congenital anorectal variants in adults with recurrent genital infections of unclear origin.
- Enteric flora on genital swabs should raise suspicion for structural contamination pathways. Recurrent PID with enteric organisms should prompt evaluation for structural abnormalities.
- Multidisciplinary assessment (gynaecology, colorectal surgery, urology) is essential in complex infertility cases.
- A thorough perineal examination is mandatory in every infertility workup to rule out structural anomalies like anorectoperineal fistulas or perineal grooves.
- Surgical reconstruction (ASARP) in adults can successfully restore anatomy, resolve chronic infections, and potentially restore reproductive potential and fertility.

Conclusion

This case highlights a rare but clinically significant cause of primary infertility in an adult couple: a variant low female anorectal malformation with an anoperineal fistula, a perineal groove and a direct continuity of anoperineal fistula into the vestibule and vagina without any barrier of skin in between. The persistent contamination of the female genital tract with enteric flora led to recurrent pelvic inflammatory disease, while the male partner developed resistant balanoposthitis due to shared exposure. Diagnosis was delayed due to the subtle anatomical presentation and absence of bowel symptoms. Surgical correction *via* primary one stage anterior sagittal anorectoplasty without any diversion resulted in complete resolution of infections and restoration of fertility.

Clinicians should maintain a high index of suspicion for congenital anorectal anomalies in adults presenting with unexplained, recurrent genital infections - especially when both partners are affected. Early recognition and multidisciplinary management can prevent long-term reproductive consequences and improve quality of life.

Patient's Perspective

"For years we were treated separately for infections that kept coming back. It was frustrating and emotionally exhausting. Discovering that a birth defect was the cause was surprising, but after surgery everything changed. We finally feel healthy - and becoming pregnant felt like a miracle."

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