Case Report

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A Rare Case of Labial Adhesion with Ectopic Urethral Orifice in A Pregnant Female

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1. Abstract

Labial fusion is a rare condition that is defined as the complete or partial fusion of the labia minora or majora. Hypo-estrogenic state routes the cause of such adhesions and hence, its incidence is seen in the pre-pubertal and, to some extent, in post-menopausal women Being a rare case, with few references in literature, we intend to analyse the cause, symptoms and management plan for one such case. We present a case of 32 years old second gravida, with fused labia minora and an ectopic urethral orifice draining into the vagina. The patient presented to our OP at 38 weeks of gestation for antepartum hemorrhage when was found to have a partially fused labia minora covering the uretheral orifice. Later, when she was posted for elective repeat caesarean section, difficulty was encountered during catheterisation where urethra was not visualised underneath the fused labia minora after partial adhesiolysis. On careful examination, urethral orifice was seen in the anterior vaginal wall and was serially dilated. She gave history of dyspareunia, recurrent UTI and poor stream of urine along with prolonged period of subfertility. This patient was treated by adhesiolysis with topical steroids and has been counselled for urethral orifice relocating surgery.

Labial fusion in a reproductive female should evoke prompt evaluation, as hypoestrogenic state cannot be a cause. This case is being presented for its rarity and to emphasise on treatment modalities for different types of fusion.

2. Keywords:

Labial fusion, labial adhesion, female hypospadiasis, ectopic urethral orifice, dyspareunia

3. Introduction

Labial adhesion is a rare condition that is defined as the complete or partial fusion of the labia minora or majora. The disorder is stated by different titles in the literature such as labial fusion, labial agglutination, or labial synechia. Labial adhesion is not present at birth, and it is thought to develop during re-epithelization of microtraumatizedhypo-estrogenised labial skin. Labial adhesion is one of the most common causes of presentations to paediatric surgery clinics among prepubertal girls. Labial adhesions are usually asymptomatic and are detected incidentally by a meticulous paediatrician. Because they are usually asymptomatic, follow-up is sufficient. However, sometimes blockage of urine flow might predispose to different symptoms such as postvoid dribbling, strain, and irritation during urination, vaginal pain or discharge, and recurrent urinary tract infection. Treatment of such symptomatic cases is indicated. And hence, its incidence is seen in the pre-pubertal and, to some extent, in post-menopausal women as hypo-estrogenic state routes the cause of such adhesions. It is extremely rare in the reproductive population, with only a few cases reported in literature. This is an atypical presentation of labial adhesion in a pregnant female, wherein the cause of adhesion is not hypo-estrogenic state nor micro-trauma.

4. Case Report

A 32-year-old second gravida, with previous caesarean section, visited our OP for the first time at 38 weeks with history of antepartum hemorrhage. On local examination, she had a fused labia minora leaving a 2-3 cm introitus open. It was only then, when her first per speculum examination was attempted and she had minimal fresh bleed when the tip of speculum was negotiated. She gave a history of coitus followed by bleeding. All initial evaluations were done for antepartum hemorrhage. By ultrasonogram, placenta was posteriorly located without any evidence of retroplacental clot. She was given a pad-watch for 12 hours and didn't have any further episodes of bleeding since then.

Previously she used to have frequent urinary tract infection, severe dyspareunia, poor voiding stream along with three years of subfertility. During her evaluation of subfertility, she gave a history of introital adhesion release in an OPD after which she conceived. She underwent elective cesarean in view of twins with non-cephalic presentation. A year later, her symptoms reappeared, however, she never noticed her fused labia minora.

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She gave a history of non-penetrative ejaculatory intercourse. Currently, when she was posted for elective repeat cesarean section, difficulty was encountered while catheterization. After partial labial adhesiolysis, urethra was not seen underneath. On careful examination, urethral orifice was seen in the anterior vaginal wall about 4cm from the introitus with mild stenosis. Urethra was serially dilated and catheterised. Intranatal and postnatal course was uneventful.

5. Management

Patient was managed with partial blunt labial adhesiolysis with serial dilatation of urethra. Catheter was removed with ease on post-operative day-1 after which patient was advised topical emollients for local application. On discharge, patient was advised to maintain perineal hygiene, local emollients, continue coital function to maintain introitus diameter.

6. Follow Up

Patient was reviewed on third week post-procedure. Labia minora was healthy and not united. Symptomatically, her voiding stream had improved. Coital function not initiated. As our patient was symptomatically improving, she was advised to be under follow up and report if she develops recurrent cystitis, post coital UTI, dyspareunia.

7. Discussion

In females, hypospadias is a rare entity. The urethral meatus opens on the anterior vaginal wall anywhere between the introitus and the fornix. This may be associated with other anomalies like 46XX disorders of sexual development (DSD), non-neurogenic neurogenic bladder and urethral duplication. Failure of both urethral-fold fusion and distal urethral migration result in female hypospadias. Embryologically this could represent an arrest during a late stage of urogenital sinus development. (Figure 1)

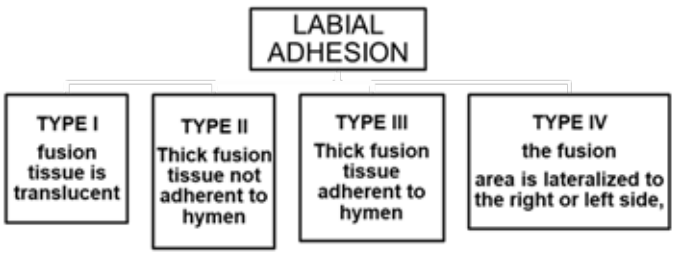

Figure 1

Management of labial adhesion depends upon patient's symptoms and age group. Topical oestrogen cream is a common therapy for childhood or postmenopausal labial adhesions. These adhesions are very superficial and are the result of mild irritation of the vaginal mucus membranes and subsequent repair. Oestrogen for several days to a few weeks, along with gentle massage, is often enough to disrupt adhesions in these groups and surgical treatment is reserved for refractory cases.[2] As in literature, patients may be corrected surgically by transpositioning urethra in the sub-clitoral region. Other causes of labial adhesion in reproductive age-group includes vulvar infections, poor hygiene, dermatitis, trauma, female circumcision and lichen sclerosis. (Figure 2)

Treatment Protocol for Labial Adhesion

Labial Fusion Type	Response to 0.1% Betamethasone treatment	Treatment Preference
Type I (48%)	100%	0.1% Betamethasone 2 × 1
Type II (20%)	80%	0.1% Betamethasone 2 × 1 If no response after 3 weeks, surgical separation
Type III (24%)	0%	Surgical separation
Type IV (8%)	0%	Surgical separation

Figure 2

In conclusion, labial fusion should be kept in mind in the differential diagnosis of urinary retention presenting in reproductive aged women without discernible etiological factors. Furthermore, it is important to perform a gynaecological examination for the evaluation of voiding symptoms even if the patient does not have a history of sexual intercourse. Otherwise, the diagnosis of labial fusion may be delayed until the reproductive years.

Labial fusion in a reproductive female should evoke prompt evaluation, as hypoestrogenic state cannot be a cause. In our case, urological anomaly was the cause. Unlike a pre-pubertal labial synechiae, topical oestrogen alone will not suffice and cause should be evaluated.

Reference

1. Huseynov M, Hakalmaz AE. Labial Adhesion: New Classification and Treatment Protocol. J Pediatr Adolesc Gynecol. 2020; 33(4): 343-348.
2. Emre E, Demirel C, Tahaoğlu AE, and Özdemir A. "Labial fusion: A rare cause of urinary retention in reproductive age woman and review of literature." Turk J Urol. 2017; 43(1): 98-101.
3. Liang Z, Chen J, Yu X, Zhu L. Persistent Labial Minora Fusion in Reproductive Age Women: A Retrospective Case Series of Nine Patients and Review of Literature. Organogenesis. 2021; 17(1-2): 20-25. doi: 10.1080/15476278.2021.1905477. Epub 2021 May 20. PMID: 34014808; PMCID: PMC8162252.
4. Tug N, Sargin MA, Yassa M, Toklucu G. An unusual cause of female secondary infertility: Hypospadias. Turk J Obstet Gynecol. 2020; 17(3): 233-235. doi: 10.4274/tjod.galenos.2020.30049. Epub 2020 Oct 2. PMID: 33072429; PMCID: PMC7538824.
5. Ze Liang, Juan Chen, Xin Yu & Lan Zhu. Persistent Labial Minora Fusion in Reproductive Age Women: A Retrospective Case Series of Nine Patients and Review of Literature, Organogenesis, 2021; 17: 1-2, 20-25, DOI:10.1080/15476278.2021.1905477