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Idiopathic Non-Puerperal Uterine Inversion in a Young Female: A Case Report

Amit Kumar*, Vinod Kumar, Priyam Sharma

Department of Radio-Diagnosis, IGIMS, Patna, Bihar, India

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ABSTRACT

Non-puerperal uterine inversion (NPUI) is an extremely rare condition. Our case is the second, where a young female (adolescent age group) presented with uterine inversion, and is the first without any identifiable cause of uterine inversion. Her chief complaints were chronic menorrhagia, abdominal pain, and episodes of acute urinary retention. Most of the reported cases have identifiable etiology for the inversion. The reporting of this case is worthwhile because of its rarity in such age groups and even without any defined cause (spontaneous occurrence), making the diagnosis of uterine inversion in this condition challenging, particularly in this very young age. In our case, she was initially misdiagnosed as having a cervical polyp clinically and radiologically, especially on ultrasonography (USG). Finally, it was diagnosed as uterine inversion by magnetic resonance imaging (MRI) of the pelvis and retrospective evaluation of USG. Magnetic resonance imaging (MRI) thus plays a pivotal role in the diagnosis of uterine inversion. Treatment of uterine inversion is either mostly by manual repositioning, and if not possible, surgery is contemplated.

Introduction

Uterine inversion is the folding of the fundus into the uterine cavity. Uterine inversion is classified into two types: puerperal (obstetric) and non-puerperal (gynecologic) [1]. Puerperal uterine inversion occurs with an incidence of about 1/2000 deliveries and is considered a medical emergency [2]. Non-puerperal uterine inversion (NPUI) is an extremely rare condition. Generally, non-puerperal uterine inversion presents after the age of 45 years and is mostly related to uterine fibroids and seldom associated with malignancies [3]. In some studies, uterine inversion is further stratified by extent: incomplete (no part of corpus past cervix), complete (inversion extends into the vagina), and prolapsed (protrudes past the introitus) [4]. The mechanism of non-puerperal uterine inversion has not been clearly identified [5]. It is often associated with the presence of a polypoid uterine tumor [6].

Non-puerperal uterine inversion is an infrequent condition, with only 190 cases reported in the literature from 1940 to 2020, and the mean age of all the patients is 47.0 ± 17.2 years [1]. Clinical features

* **Corresponding Author:**
Amit Kumar
Address: *Radio-diagnosis, Room No.-249, Department of Radio-diagnosis, IGIMS, Patna-14, Bihar, India.*
E-mail: *amitmd2008@gmail.com.*

include vaginal bleeding, a mass protruding through the introitus, lower abdominal pain, and urinary problems [7]. The inverted uterus forms an inverted pyriform swelling, which occupies the upper part of the vagina. In the case of total inversion, the mass will protrude out of the introitus. Imaging techniques such as ultrasound and magnetic resonance imaging (MRI) help in the diagnosis, especially MRI, while the diagnostic value of transvaginal ultrasound is limited. In case of uterine inversion, a "U-shaped" uterine cavity with a thickened and inverted uterine fundus on a sagittal image and a "bulls-eye" configuration on an axial image in MRI and USG are key imaging findings [6].

Uterine inversion is a medical emergency and needs early intervention. It is based on a medical examination and is done with manual reduction and surgical treatment using various techniques. Here we report a case of uterine inversion in a 12-year-old girl presenting without any definable etiology (idiopathic) and treated by conservative maneuver of manual uterine fundus repositioning. To our knowledge, this is the first such case in a 12-year-old girl presenting with spontaneous inversion without a cause.

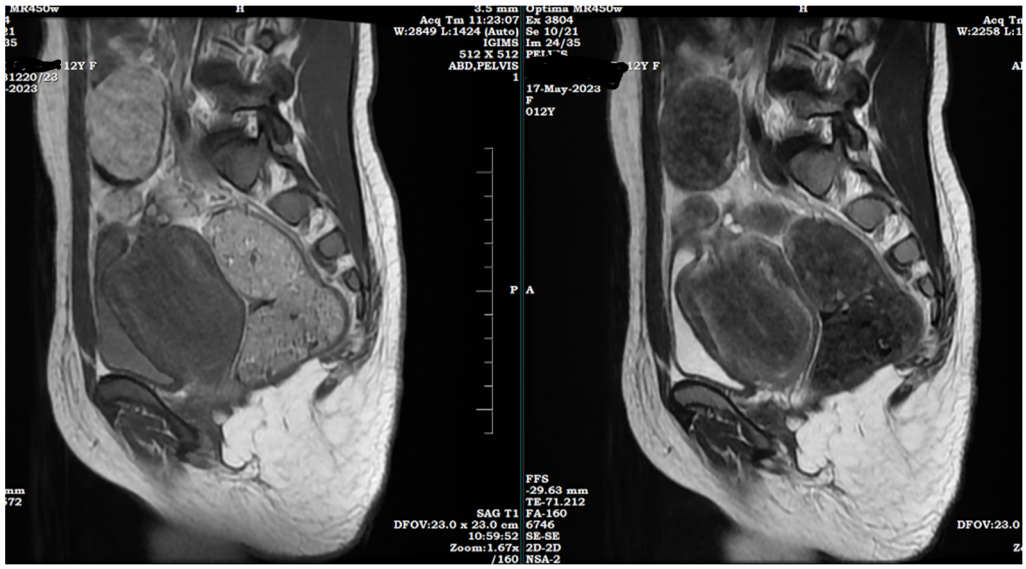
Case Report

The patient is a 12-year-old girl who was apparently asymptomatic until 3 months ago and presented with prolonged menstrual bleeding. The patient attained menarche 5 months ago (in December 2022), where she had regular cycles for two months that lasted for 4 days. In February 2022, she complained of excessive bleeding for 20 days, which then stopped until April. Now she has started to bleed continuously again. She had a history of continuous abdominal pain with urinary retention on 5th May 2023.

On 10th May 2023, she was admitted to a local hospital with a history of fever, foul-smelling vaginal discharge, urinary retention, and anemia. Examination under local anesthesia was done, and she received 2 units of blood transfusion. Local examination findings suggested a firm polypoidal lesion with necrotic tissue on the surface, and no demarcation between the mass and uterus; hence, the procedure was abandoned after packing the vagina, and she was referred to a higher center. There was no history of constipation or sexual abuse.

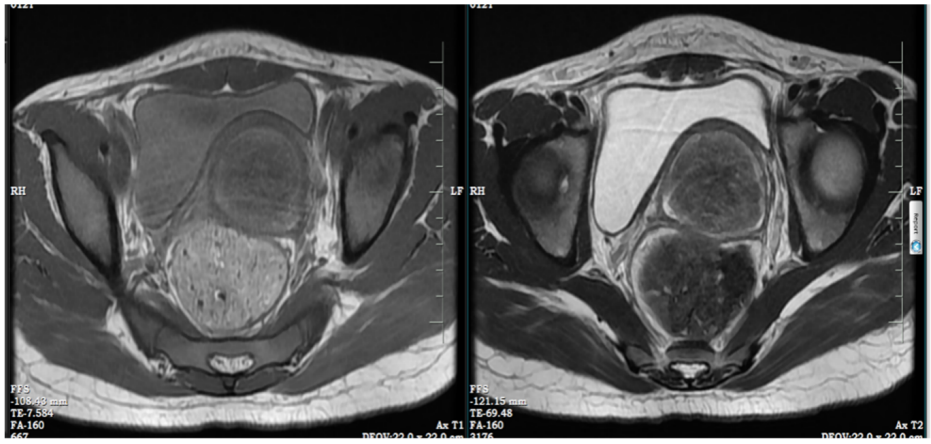
In IGIMS, Patna, on external examination vulva appeared normal. A firm mass was noticed protruding into the vagina, completely filling the vagina and fingers could not reach beyond mass. She was advised Pelvic MRI to know the nature of lesion, extent of lesion and anatomical state of uterus and adnexa.

Her MRI showed a U-shaped uterus with loss of normal convex fundal contour on sagittal T1 and T2 scans (Figure 1 A and B). The uterus is upside down, and the fundus is protruding into the cervical canal, reaching up to the upper vaginal canal, indicative of uterine inversion. Bilateral broad ligaments are also



(A) (B)

Fig. 1. (A) Sagittal T1. (B) Sagittal T2. U-shaped uterus with loss of normal convex fundal contour, fundus is protruding into cervical canal.



(A) (B)

Fig. 2. (A) Axial T1W. (B) Axial T2W. Axial section of pelvis shows target appearance of uterus.

pulled, and both adnexa are sitting onto the uterus, also visualized on axial T1 and T2 images (Figure 2 A and B). Coronal T1 and T2 images are also shown (Figure 3 A, B, C). The size of the uterus measures about 6 x 4 cm. Retrospectively (after MRI and its evaluation) , USG was done for the patient, and the findings were similar to the MRI findings (Figure 4 A, B, C). At IGIMS, Patna, she was advised on conservative management with antibiotics,blood transfusion, blood coagulants, etc. Unfortunately,in the meantime, the patient left the hospital without prior information for another higher center, Christian Medical College Vellore, Tamil Nadu, India.

There, she was admitted with the same diagnosis of idiopathic uterine inversion and was finally managed by manual repositioning of the fundus back under general anesthesia. The post-operative status of the patient was normal, and she was discharged. Follow-up was uneventful.

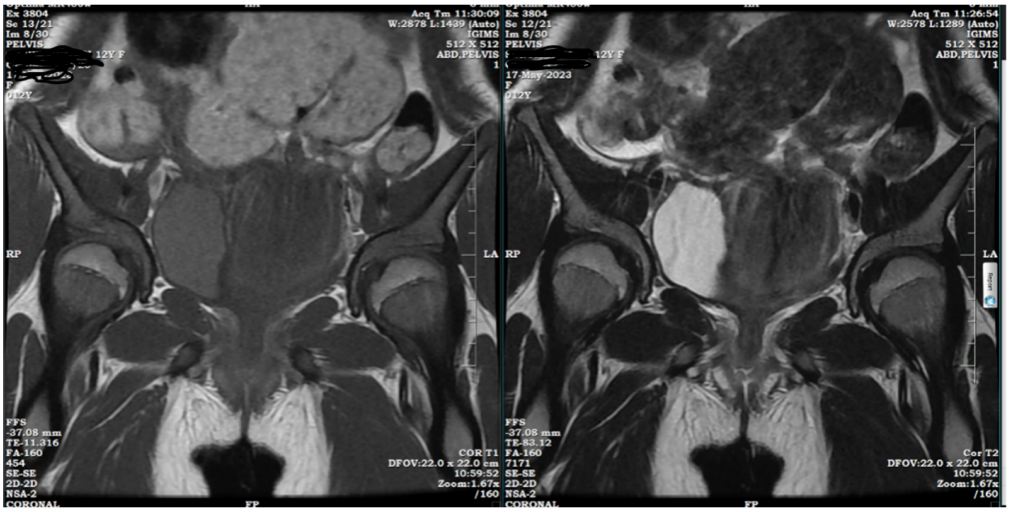
Discussion

Non-puerperal uterine inversions are rare, although some reports suggest that they comprise one-sixth of all inversions [6]. There have been several reported cases of non-puerperal uterine inversion over the past 10 years (2006–2017) [6]. According to a literature review, ours is only the second case where the youngest female presenting with uterine inversion has been identified. The youngest girl reported previously was an 11-year-old with an endometrial polyp [1]. In our case, no identifiable etiology causing the inversion of the uterus has been found, hence it is likely to be idiopathic, which has not yet been reported to the best of our knowledge.

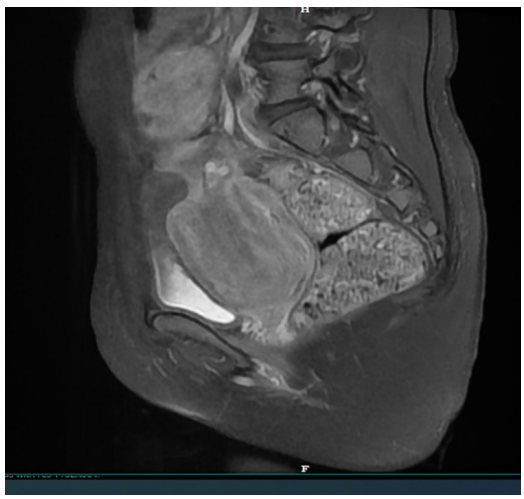
Non-puerperal uterine inversion may be idiopathic or associated with predisposing factors such as benign uterine tumors in 70–80 percent of cases (leiomyoma, endometrial polyps), or malignant tumors in the remaining cases, especially in young women [8]. Uterine inversion is classified into 4 stages: (1) incomplete inversion of the uterus with the uterine fundus in the cavity, (2) complete inversion of the uterine fundus through the cervix, (3) complete inversion with the fundus protruding through the vulva, and (4) complete inversion through the vulva of both the uterus and the vagina [9]. In our case, the fundus of the uterus was protruding out of the cervix into the upper vaginal canal on initial local clinical examination and was simulating a cervical polyp protruding into the vagina. However, radiological evaluation later showed inversion of the uterine fundus up to the cervical level in MRI and USG, likely retracted.

Strong symptomatic signs define the acute form, while the chronic form can be less symptomatic with pelvic pain, pelvic heaviness, or bleeding. Anemia, urinary symptoms, and vaginal mass are also reported. Hemorrhage is minimal, unlike in puerperal uterine inversion [5].

Uterine inversion in early age groups is not only rare but also poses great challenges to diagnose when nothing is obvious and mainly presents with menorrhagia and recurrent urinary retention. Including uterine inversion in the differential diagnosis in this early age female is crucial. This often leads to false or misdiagnosis, as with our case, which was falsely diagnosed as a cervical polyp. In this regard, imaging plays a key role in such patients to arrive at a definitive diagnosis. Ultrasound is usually the first line of investigation. The key finding is a “Y”-shaped uterine cavity in the longitudinal plane seen



(A) (B)



(C)

Fig. 3. (A) Coronal T1W. (B) Coronal section T2W. U shaped uterus with loss of normal contour, and fundus protruding into the cervical canal. (C) Sagittal T1W Fat sat sequence showing fundus protruding into cervical canal.

in incomplete uterine inversions on USG. In contrast, the longitudinal view in complete inversion shows a "U"-shaped configuration, with the limbs of the "U" representing the completely inverted endometrial lining both anteriorly and posteriorly, as noted in our case on USG (Fig. 4a, 4b) [10].

MRI is found to be more sensitive in the diagnosis of NPUI. Distinct observations identified are a U-shaped uterine cavity, a thickened and inverted uterine fundus on a sagittal section, and a "bull's-eye" configuration on the horizontal section [10], which we clearly noticed in our patient's MRI imaging of the uterus as well, definitely suggesting "Uterine Inversion."

In our case, no possible cause was identified clinically or even during the management maneuver.

The morbidity and mortality associated with uterine inversion correlate with the degree of hemorrhage, the rapidity of diagnosis, and the effectiveness of treatment [11]. Reposition procedures according to the reproductive desire of the patient or hysterectomy could be considered for surgical treatment [3].

If the female wishes to maintain fertility, repositioning is considered, as was done in our case. There are reports of successful pregnancies following the surgical correction of puerperal uterine inversion [10]. Surprisingly, although the literature stresses the importance of conserving the uterus if fertility is required, we could not find any evidence regarding the status of successful or unsuccessful pregnancies following repositioning of a non-puerperal uterine inversion [10].

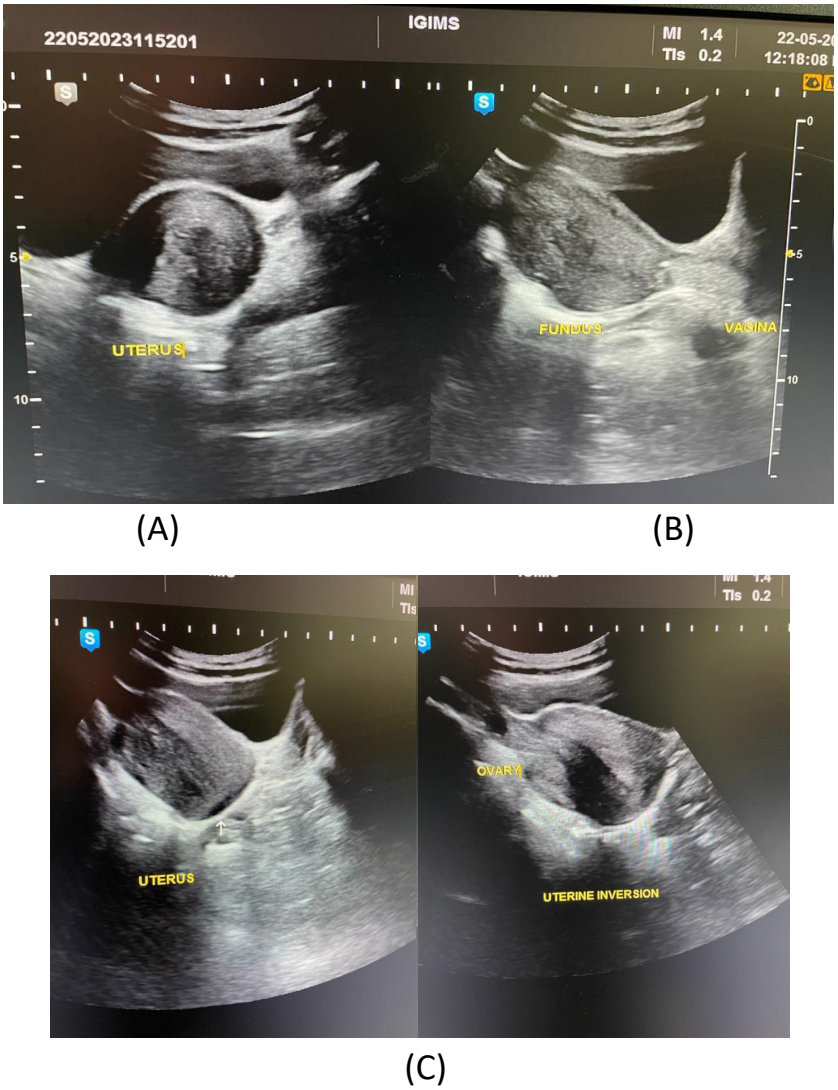


Fig. 4. (A) Axial and Sagittal section of uterus on ultrasound showing abnormal uterine contour and target sign in axial image. (B) Sagittal section of uterus on ultrasound. (C) Sagittal section of uterus and right ovary on ultrasound showing abnormal uterine contour with fundus protruding inside the cavity and right ovary separately visualised.

Conclusion

Non-puerperal uterine inversion is a rare entity and even rarer in young girls. Our case is unique in that we did not find any defined or identifiable etiology causing the inversion of the uterus, as inversion is mostly associated with identifiable uterine causes. In very young age groups, where clinical findings may be limited due to restricted local evaluation, radiological evaluation greatly aids in arriving at a definitive diagnosis, especially MRI evaluation of the pelvis or whole abdomen. In our case, initial clinical investigation led to a misdiagnosis, which was finally resolved by MRI evaluation of the pelvis. Therefore, MRI evaluation is always advised if there is any doubt in clinical studies. Treatment of such uterine inversion in this age group is conservative and managed by simply repositioning the inverted part in a proper setting.

Ethical Considerations

Compliance with ethical guidelines

There were no ethical considerations to be considered in this article.

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Conflict of Interests

The authors have no conflict of interest to declare.

Patient Perspective

Manual Repositioning was done in this patient and she was normal after treatment. Now she din't have menorrhagia, anaemia was corrected and no fresh retention of urine noted. Her parent was now ok with her.

Contribution

Amit K: Conceived and designed the case, Collected the data, Performed the editing, and contributed in writing the paper

Vinod K: Conceived and designed the case, Collected the data, and contributed in writing the paper

Priyam S: Conceived and designed the case, Collected the data, Performed the editing, and contributed in writing the paper

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