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Incarcerated gravid uterus – A systematic review

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ABSTRACT

Incarcerated gravid uterus (IGU) is a serious complication of pregnancy that leads to adverse obstetric outcomes. The aim of this review was to describe this entity in detail. We also aimed to understand if pregnancies with predisposing risk factors that increase the risk of developing IGU had a difference in their clinical manifestations, treatment, and obstetric outcomes. The PubMed, MEDLINE, Embase, Scopus databases and clinicaltrials.gov were searched from inception to July 2023. Case reports and series that provided all the details of the pregnancy and IGU outcome were included. Study quality and risk of bias were assessed using a tool that is an adaptation from criteria listed by Pierson, Bradford Hills and Newcastle Ottawa scale modification. Patients with the condition of interest included in this review were grouped into those with documented, identified risk factors and no risk factors. The two groups were compared to understand the difference in obstetric outcome and presentation of IGU. Data were analyzed and summarized descriptively, categorical variables were assessed by chi-squared test or Fisher's exact test, and continuous variables by the Wilcoxon Mann Whitney test. Of 236 articles found, 62 articles with 80 cases were included in the final analysis. The median age was 32 [27–35] years. The median gestational age of diagnosis was 17 [14–26] weeks. The most common risk factor was fibroids (N = 22, 27.5 %). Most common presentation was urinary complaints and lower abdomen pain (N = 47, 58.6 %). Twenty-seven patients (33.6 %) needed more than one visit for the diagnosis to be made. Conservative management was the first step to treat IGU in most patients. Most common complication was fetal malpresentation (N = 13, 40.6 %). Patients with or without risk factors developing IGU had no statistical difference in parity, median gestational age of diagnosis, delay in diagnosis, increased chance of misdiagnosis, management of IGU or in obstetric outcome (all $p > 0.05$). It is important to recognize this entity early to prevent obstetric complications especially when patients report urinary retention and abdomen pain. The presence of risk factors does not change the management course or obstetric outcome in patients with IGU. Hence it is reasonable to start with conservative management of IGU regardless of presence of risk factors or the gestational age of diagnosis, in clinical practice.

1. Introduction

Incarcerated gravid uterus (IGU) is a rare, but serious, condition complicating 1 in 3000 pregnancies that leads to adverse maternal and fetal outcomes [1]. The retroverted uterus is a normal variant that is seen in 15 % of the pregnancies [2]. With uncomplicated pregnancies, the axis of the uterus corrects itself and no adverse pregnancy outcomes are found. However, a pregnant retroverted uterus can persist when present with certain predisposing factors (e.g., pelvic adhesions, fibroids, pelvic, or uterine structural anomalies), which can lead to its entrapment within the sacral hollow [3]. This in turn leads to the clinical

manifestation of an IGU. With the rise in high-risk pregnancies complicated by the above-mentioned predisposing factors, we aimed to review the clinical presentation, diagnosis, management, obstetric outcomes, and complications of patients with IGU. We also aimed to compare the clinical manifestations, treatment and obstetric outcomes, occurrence of complications and see if it made a difference in those who had developed IGU during pregnancy with and without known predisposing risk factors.

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2. Methods and materials

This review follows the PRISMA (Preferred Reporting Items for Systematic Reviews and Meta-analyses) and was registered with the PROSPERO International Prospective Register for Systematic Reviews (registration no. CRD42022298610).

2.1. Sources

The PubMed (National Library of Medicine), MEDLINE, Embase, and Scopus databases and clinicaltrials.gov were searched from inception to July 2023 for English language studies with no restrictions on date or geographic location. Search headings included, 'incarcerated uterus', 'gravid', 'pregnancy', 'incarceration of uterus' and 'risk factor'.

2.2. Study selection

Eligibility for inclusion was articles providing details of pregnant patients who were diagnosed with IGU during pregnancy or intra-operatively during delivery and the management provided and their subsequent pregnancy course. Studies were excluded if the articles provided no details on the method by which IGU was corrected or on the obstetric outcome after the IGU was corrected. The literature search identified case reports and series on IGU (Table 1). No prospective or retrospective studies providing information of interest were identified in our search. Two authors, AH and RP, screened the abstracts of the published manuscripts, and a third author SN resolved conflicts. Full-text articles were obtained for all included abstracts. Full-text article review was conducted by the same two authors independently, with conflicts resolved in the same manner.

The methodological quality of case reports and series included in the review was assessed, based on the tool described by Murad et al. [64]. This tool assesses case reports and series on the domains of selection, ascertainment, causality, and reporting, and was an adaptation from the criteria listed by Pierson, Bradford Hills and Newcastle Ottawa scale modification. The aggregate score for each series and report included in the study is provided in Table 2. To ensure good inter-rater reliability correlation two authors independently assessed the reports and series. We considered the quality of the report as good (low risk of bias) when all 5 criteria were fulfilled, moderate when 4 were fulfilled, and poor (high risk of bias) when 3 or less were fulfilled. The same two reviewers assessed the risk of bias of the included studies with discussion between them in case of disagreement.

All data were collected independently by co-investigators, AH and RP, into a study spreadsheet and verified by a separate author, SN. The following data was collected (Supplementary material 1).

- Baseline characteristics of the study including year of study, country of origin, study design (Table 1).
- Patient characteristics: age, parity, number of previous vaginal deliveries and cesarean deliveries, risk factors that may predispose patient to developing IGU.
- Details of IGU: presenting complaint, gestational age at time of presentation, number of visits made by patient with same complaint before diagnosis was made, methods to diagnose the condition (pelvic examination, ultrasound, magnetic resonance imaging (MRI)), the misdiagnosis made.
- Management provided such as conservative or surgical management, number of attempts in conservative management before proceeding with surgical management.
- Obstetric outcomes: mode of delivery, gestational age of delivery.

Risk factors for the development of IGU mentioned were the presence of uterine fibroids, adhesions, uterine or pelvic anatomical anomaly, pelvic infection, prior pelvic surgery, and prior pregnancy with IGU. We grouped the patients included in this review into two categories- one in

Table 1
Characteristics of case reports/series included.

First author	Publication year	Country	Number of IGU cases reported
Calvert et al. [4]	1909	India	1
Freeth et al. [5]	1953	Australia	1
Carty et al. [6]	1954	USA	3
Evans et al. [7]	1986	UK	1
Nelson et al. [8]	1986	USA	1
Edminster et al. [9]	1987	USA	1
Meislin et al. [10]	1987	USA	1
Hankins et al. [11]	1989	USA	2
Smalbraak et al. [12]	1991	Netherlands	4
Van Winter et al. [13]	1991	USA	3
Keating et al. [14]	1992	UK	1
Gunn et al. [15]	1993	Australia	1
Patterson et al. [16]	1997	USA	1
Hirsch et al. [17]	1997	USA	1
Feusner et al. [18]	1997	USA	1
O'Connell et al. [19]	1999	UK	1
Algra et al. [20]	1999	USA	2
Love et al. [21]	2000	USA	2
Li et al. [22]	2000	Taiwan	1
Hamod et al. [1]	2002	UK	1
Matsushita et al. [23]	2004	Japan	1
Inaba et al. [24]	2005	Japan	1
Barton-Smith et al. [25]	2007	UK	1
Singh et al. [26]	2007	UK	1
Charova et al. [27]	2008	UK	1
Chauleur et al. [28]	2008	France	2
Sweigart et al. [29]	2008	USA	1
Hooker et al. [30]	2009	Netherlands	1
Van der Tuuk et al. [31]	2009	Netherlands	1
Dierickx et al. [32]	2010	Belgium	1
Dierickx et al. [33]	2011	Belgium	4
Grossenburg et al. [34]	2011	USA	1
Hachisuga et al. [35]	2012	Japan	1
Wang et al. [36]	2012	China	1
Katopodis et al. [37]	2013	Canada	1
Newell et al. [38]	2014	UK	2
Matsushita et al. [39]	2014	Japan	1
Policiano et al. [40]	2014	Portugal	2
Dierickx et al. [41]	2014	Belgium	1
Slama et al. [42]	2015	USA	3
Ozyurek et al. [43]	2015	Turkey	1
Takami et al. [44]	2016	Japan	1
Hassanin et al. [45]	2016	Egypt	1
Sadath et al. [46]	2016	USA	1
Yamamoto et al. [47]	2017	Japan	1
Gardner et al. [48]	2018	USA	1
Lawrence et al. [49]	2018	USA	1
Alhousseini et al. [50]	2018	USA	1
Soyama et al. [51]	2018	Japan	1
Hsu et al. [3]	2018	Taiwan	1
Kim et al. [52]	2018	Korea	1
Tong et al. [53]	2019	China	1
Han et al. [54]	2019	China	1
Ouchi et al. [55]	2019	Japan	1
Hire et al. [56]	2019	USA	1
Lackey et al. [57]	2019	USA	1
Suzuki et al. [58]	2020	Japan	1
Kocher et al. [59]	2020	USA	1
Ntafam et al. [60]	2022	USA	1
Abelman et al. [61]	2022	USA	1
Morais et al. [62]	2022	Portugal	1
Tanimura et al. [63]	2023	Japan	1

USA, United States of America; UK, United Kingdom.

Table 2

Tool for risk of bias assessment of case reports and case-series.

First author	Year of publication	Country of publication	Selection ^a	Ascertainment ^b	Causality-A ^c	Causality B ^d	Reporting ^e	Score	Bias
Calvert et al. [4]	1909	India	Y	Y	Y	Y	Y	5	Good
Freeth et al. [5]	1953	Australia	Y	Y	Y	Y	Y	5	Good
Carty et al. [6]	1954	USA	Y	Y	Y	Y	Y	5	Good
Evans et al. [7]	1986	UK	Y	Y	Y	Y	Y	5	Good
Nelson et al. [8]	1986	USA	Y	N	Y	Y	Y	4	Moderate
Edminster et al. [9]	1987	USA	Y	Y	Y	Y	Y	5	Good
Meislin et al. [10]	1987	USA	Y	N	Y	Y	Y	4	Moderate
Hankins et al. [11]	1989	USA	Y	Y	Y	Y	Y	5	Good
Smalbraak et al. [12]	1991	Netherlands	Y	Y	Y	Y	Y	5	Good
Van Winter et al. [13]	1991	USA	Y	Y	Y	Y	Y	5	Good
Keating et al. [14]	1992	UK	Y	N	Y	Y	Y	4	Moderate
Gunn et al. [15]	1993	Australia	Y	Y	Y	Y	Y	5	Good
Patterson et al. [16]	1997	USA	Y	Y	Y	Y	Y	5	Good
Hirsch et al. [17]	1997	USA	Y	Y	Y	Y	Y	5	Good
Feusner et al. [18]	1997	USA	Y	N	Y	Y	Y	4	Moderate
O'Connell et al. [19]	1999	UK	Y	Y	Y	Y	Y	5	Good
Algra et al. [20]	1999	USA	Y	Y	Y	Y	Y	5	Good
Love et al. [21]	2000	USA	Y	Y	Y	Y	Y	5	Good
Li et al. [22]	2000	Taiwan	Y	Y	Y	Y	Y	5	Good
Hamod et al. [1]	2002	UK	Y	Y	Y	Y	Y	5	Good
Matsushita et al. [23]	2004	Japan	Y	Y	Y	Y	Y	5	Good
Inaba et al. [24]	2005	Japan	Y	Y	Y	Y	Y	5	Good
Barton-Smith et al. [25]	2007	UK	Y	Y	Y	Y	Y	5	Good
Singh et al. [26]	2007	UK	Y	Y	Y	Y	Y	5	Good
Charova et al. [27]	2008	UK	Y	Y	Y	Y	Y	5	Good
Chauleur et al. [28]	2008	France	Y	Y	Y	Y	Y	5	Good
Sweigart et al. [29]	2008	USA	Y	N	Y	N	Y	3	Poor
Hooker et al. [30]	2009	Netherlands	Y	Y	Y	Y	Y	5	Good
Van der Tuuk et al. [31]	2009	Netherlands	Y	Y	Y	Y	Y	5	Good
Dierickx et al. [32]	2010	Belgium	Y	Y	Y	Y	Y	5	Good
Dierickx et al. [33]	2011	Belgium	Y	N	Y	N	Y	3	Poor
Grossenburg et al. [34]	2011	USA	Y	Y	Y	Y	Y	5	Good
Hachisuga et al. [35]	2012	Japan	Y	Y	Y	Y	Y	5	Good
Wang et al. [36]	2012	China	Y	N	Y	N	Y	3	Poor
Katopodis et al. [37]	2013	Canada	Y	Y	Y	Y	Y	5	Good
Newell et al. [38]	2014	UK	Y	Y	Y	Y	Y	5	Good
Matsushita et al. [39]	2014	Japan	Y	Y	Y	Y	Y	5	Good
Policiano et al. [40]	2014	Portugal	Y	Y	Y	Y	Y	5	Good
Dierickx et al. [41]	2014	Belgium	Y	Y	Y	Y	Y	5	Good
Slama et al. [42]	2015	USA	Y	N	Y	Y	Y	4	Moderate
Ozyurek et al. [43]	2015	Turkey	Y	Y	Y	Y	Y	5	Good
Takami et al. [44]	2016	Japan	Y	Y	Y	Y	Y	5	Good
Hassanin et al. [45]	2016	Egypt	Y	N	Y	N	Y	3	Poor
Sadath et al. [46]	2016	USA	Y	Y	Y	Y	Y	5	Good
Yamamoto et al. [47]	2017	Japan	Y	Y	Y	Y	Y	5	Good
Gardner et al. [48]	2018	USA	Y	Y	Y	Y	Y	5	Good
Lawrence et al. [49]	2018	USA	Y	Y	Y	Y	Y	5	Good
Alhousseini et al. [50]	2018	USA	Y	Y	Y	Y	Y	5	Good
Soyama et al. [51]	2018	Japan	Y	Y	Y	Y	Y	5	Good
Hsu et al. [3]	2018	Taiwan	Y	Y	Y	Y	Y	5	Good
Kim et al. [52]	2018	Korea	Y	N	Y	Y	Y	4	Moderate
Tong et al. [53]	2019	China	Y	Y	Y	Y	Y	5	Good
Han et al. [54]	2019	China	Y	Y	Y	Y	Y	5	Good
Ouchi et al. [55]	2019	Japan	Y	Y	Y	Y	Y	5	Good
Hire et al. [56]	2019	USA	Y	Y	Y	Y	Y	5	Good
Lackey et al. [57]	2019	USA	Y	Y	Y	Y	Y	5	Good
Suzuki et al. [58]	2020	Japan	Y	Y	Y	Y	Y	5	Good
Kocher et al. [59]	2020	USA	Y	Y	Y	Y	Y	5	Good
Ntafam et al. [60]	2022	USA	Y	Y	Y	Y	Y	5	Good
Abelman et al. [61]	2022	USA	Y	Y	Y	Y	Y	5	Good
Morais et al. [62]	2022	Portugal	Y	Y	Y	Y	Y	5	Good
Tanimura et al. [63]	2023	Japan	Y	Y	Y	Y	Y	5	Good

Y, Yes; N, No; USA, United States of America; UK, United Kingdom.

^aSelection: Did the patient(s) represent the case of the medical center?^bAscertainment: Was the exposure and outcome ascertained?^cCausality-A. Were other alternative causes that may explain the observation ruled out?^dCausality-B. Was follow-up long enough for outcomes to occur?^eReporting: Was the case(s) described with sufficient details to allow practitioners make inferences related to their own practice?

whom no risk factor could be identified and the other in whom risk factors were identified. The age, parity, median gestational age of diagnosis of incarcerated uterus, if there was delay in diagnosis, course of management of IGU diagnosis and obstetric outcome were compared

between these two groups. Delay in diagnosis was defined as needing more than the first visit to diagnose IGU and if it was misdiagnosed during the first visit.

Data analysis was conducted using STATA 16.0 software (Stata Corp,

College Station, TX, USA). Patient characteristics, IGU manifestations and its management, and obstetric outcomes were summarized descriptively. Demographic data, pregnancy outcomes, and management of IGU were compared between patients with and without risk factors. Categorical variables were assessed by chi-squared test or Fisher's exact test. Continuous variables were presented as median with interquartile range (IQR) and assessed by the Wilcoxon Mann Whitney test. A p value 0.05 was considered statistically significant.

3. Results

The initial database search identified 236 abstracts from which 62 articles with 80 individual cases were included in the final analysis (Fig. 1). Most of the articles were from the United States (N = 23, 37.1 %), followed by Japan (N = 10, 16.1 %), and the UK (N = 8, 12.9 %). The maximum number of articles included in the review were published between the years 2000 and 2023 (N = 45, 72.6 %) followed by years 1980–1999 (14, 22.6 %) (Table 3).

3.1. Patient demographics and risk factors (Table 3)

Of the 80 patients, the median age was 32 years, IQR [29–37] years. Nulliparous, multiparous with 2–4 deliveries and grand multiparous patients with 5–9 deliveries were 48 (60 %), 29 (36.3 %), 3 (3.7 %), respectively. Of the 29 patients with one or more parity, only 7 (1 %) reported history of prior cesarean delivery. Most of the patients were

diagnosed with IGU between 13 and 20 weeks (36, 45 %) with median gestational age of diagnosis being 17 weeks, IQR [14–26] weeks. The most common risk factor noted was fibroids (N = 22, 27.5 %) followed by presence of pelvic adhesions (N = 13, 16.3 %). The other risk factors reported were history of infertility (N = 8, 10 %), uterine and pelvic structural anomaly (N = 7, 8.5 %), prior pelvic surgeries (N = 6, 7.5 %), infections such as pelvic inflammatory disease (N = 4, 5 %) and tuberculosis (N = 1, 1.3 %), prior IGU (N = 4, 5 %), and endometriosis (N = 1, 1.3 %). Patients had one or more risk factors present simultaneously (supplementary material 1).

3.2. Clinical Manifestation of incarcerated gravid uterus (Table 3)

Majority of the patients presented with urinary complaints and lower abdominal pain (N = 47, 58.6 %). Five (6.3 %) of the patients presented with preterm labor (PTL), preterm premature rupture of membranes (PPROM), or abruption. Seventeen (21.3 %) patients remained asymptomatic, however 'inverted polarity' of the uterus with the fundus being caudal rather than cephalad was noted intraoperatively during cesarean delivery. One article reported a rare presentation of rectal prolapse as the presenting complaint. Misdiagnosis made while diagnosing IGU was mentioned for 28 patients. Common misdiagnosis noted were a degenerating fibroid (N = 5, 17.6 %), urinary tract infection (N = 7, 25 %), hydronephrosis in acute kidney injury (AKI) (N = 1, 3.6 %), low lying placenta/placenta previa (N = 11, 39.3 %), abdominal pregnancy (N = 1, 3.6 %), pelvic inflammatory disease (PID) (N = 1, 3.6 %),

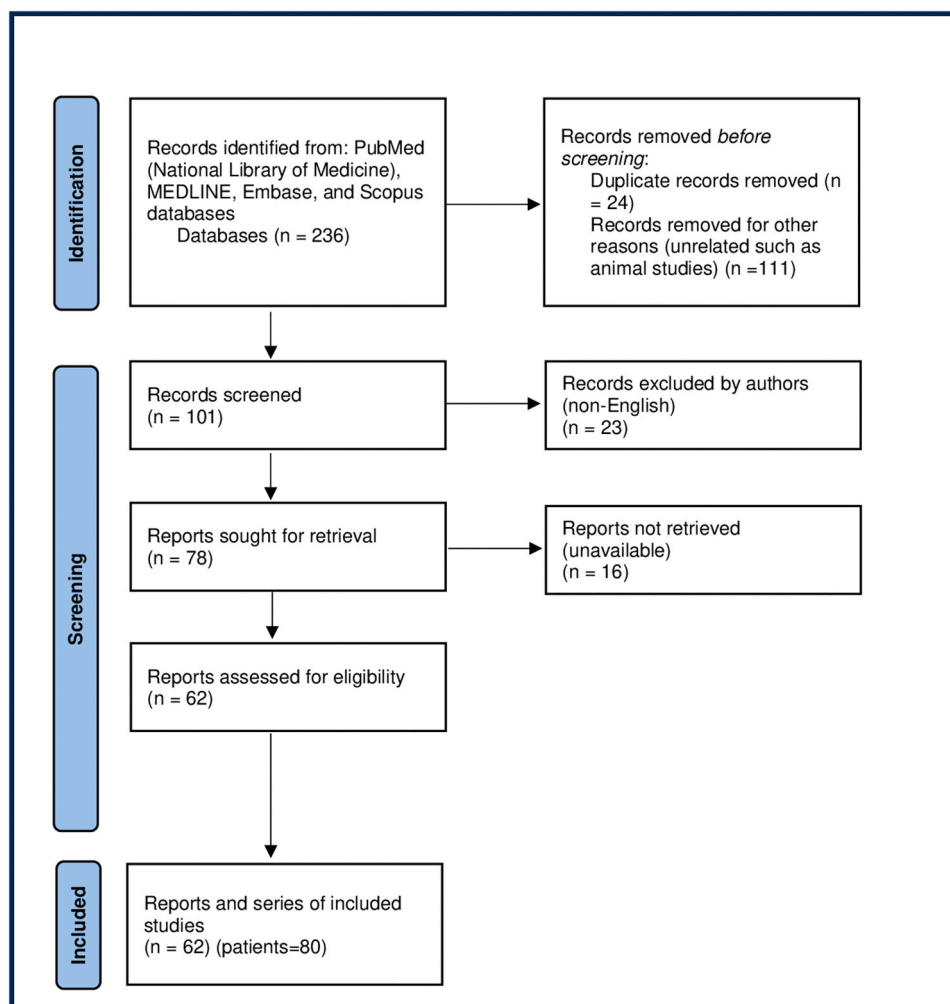


Fig. 1. Flow diagram for the database search.

Table 3
Summary descriptive statistics of the included case reports/series.

Demographics/characteristics	Number of cases N = 80
Country of publication	N = 62
Australia	2 (3.2)
Belgium	3 (4.8)
Canada	1 (1.6)
China	3 (4.8)
Egypt	1 (1.6)
France	1 (1.6)
India	1 (1.6)
Japan	10 (16.1)
Korea	1 (1.6)
Netherlands	3 (4.8)
Portugal	2 (3.2)
Taiwan	2 (3.2)
Turkey	1 (1.6)
United Kingdom	8 (12.9)
United States	23 (37.1)
Years of publication	N = 62
2000–2023	45 (72.6)
1980–1999	14 (22.6)
1950–1979	2 (3.2)
Before 1950	1 (1.6)
Median age, year	32 [27–35]
Parity	N = 80
Nulliparity	48 (60)
Multiparity	29 (36.3)
Grand Multiparity	3 (3.7)
Median Gestational age in weeks	17 [14–26]
Gestational age at diagnosis (weeks)	N = 80
< / = 13	14 (17.5)
14–20	36 (45)
21–28	10 (12.5)
29–34	10 (12.5)
35–37	3 (3.7)
Term (more than 37)	5 (6.3)
Unknown	2 (2.5)
Various risk factors*	36 (45)
No identified risk factors	13 (16.3)
Adhesions	7 (8.7)
Anomaly (uterine/pelvis)	22 (27.5)
Fibroid	8 (10)
Infertility	4 (5)
Pelvic Inflammatory disease	6 (7.5)
Prior pelvic surgeries	4 (5)
Prior history Incarcerated uterus	1 (1.3)
Endometriosis	1 (1.3)
Tuberculosis	
Presenting complaint	N = 80
No complaint	17 (21.3)
Urinary complaints* with abdomen pain	47 (58.6)
Lower abdomen pain	9 (11.3)
Obstetric complaints+	5 (6.3)
Rectal prolapse	1 (1.3)
Pedal edema	1 (1.3)
Missed diagnosis	N = 28
Fibroid degeneration	5 (17.6)
Low lying placenta/placenta previa	11 (39.3)
Urinary tract infection	7 (25)
Labor	1 (3.6)
Abdominal pregnancy	1 (3.6)
Hydrourteronephrosis with acute kidney injury	1 (3.6)
Pelvic Inflammatory disease	1 (3.6)
Ovarian torsion	1 (3.6)
Delay in Diagnosis	27 (48.2)
Patients needing more than one visit for diagnosis (N = 56)	
Diagnosis	N = 80
Clinical (physical exam alone)	14 (17.5)
Clinical with US	28 (35)
Clinical with MRI	5 (6.3)
Clinical with US and MRI	10 (12.5)
US alone	10 (12.5)

Table 3 (continued)

Demographics/characteristics	Number of cases N = 80
US with MRI	6 (7.5)
Intraoperative diagnosis	2 (2.5)
Unknown	5 (6.3)
Management	61 (76.3)
Conservative (N = 80)	7 (11.5)
Failed conservative management (N = 61)	7 (8.6)
Surgical management (N = 80)	13 (16.3)
No attempt to correct incarcerated uterus (N = 80)	6 (7.5)
Unknown management (N = 80)	
Initial conservative management	N = 61
Foley catheter with expectant management	18 (29.5)
Foley catheter + maternal positional change	10 (16.4)
Foley catheter + manual reduction (vaginal or rectal)	26 (42.6)
Foley catheter + maternal positional change + manual reduction with pelvic examination	7 (11.5)
Obstetric outcome	43 (53.7)
Uneventful (N = 80)	5 (6.3)
Unknown (N = 80)	12 (37.5)
Preterm delivery/ Ruptured membranes (N = 32)	13 (40.6)
Malpresentation (N = 32)	4 (12.5)
Fetal growth restriction (N = 32)	4 (12.5)
Fetal death and miscarriage (N = 32)	1 (3.1)
Non reassuring fetal heart tracing (N = 32)	1 (3.1)
Accreta (N = 32)	
Mode of delivery	N = 80
Unknown	6 (7.5)
Vaginal delivery (N = 74)	36 (48.6)
• Preterm (N = 36)	4 (11.1)
• Term (N = 36)	32 (88.9)
Cesarean delivery (N = 74)	36 (48.6)
• Preterm (N = 36)	
• Term (N = 36)	16 (44.5)
	20 (55.5)
Dilation and Curettage (N = 74)	1 (1.3)
Hysterectomy (N = 74)	1 (1.3)
Gestational Age of delivery	N = 80
37 and above	49 (61.3)
34–36	4 (5)
28–33	11 (13.8)
20–27	5 (6.3)
< 20	3 (3.6)
Unknown	8 (10)

*dysuria, frequency, dribbling, hesitancy. +contractions, ruptured membranes.

Data represented as N (%) or median [IQR]. US: Ultrasound, MRI: Magnetic Resonance Imaging.

* Patient could have one or more risk factors.

preterm labor (N = 1, 3.6 %), and ovarian torsion (N = 1, 3.6 %).

3.3. Methods of diagnosis (Table 3)

Of the 80 patients, 24 reports had no mention on how many recurring visits it took to diagnose incarcerated gravid uterus. Of the 56 remaining patients, 27(48.2 %) needed more than one visit with presenting complaints for the diagnosis to be made. Of the 80 patients, 57 (71.3 %) had a pelvic examination as the first step to approach to diagnosis. This revealed a marked anterior displacement of the cervix with retroverted uterus and a fullness of the pouch of Douglas. Ultrasound was used in 54 patients (67.5 %), of which 10 (12.5 %) had the ultrasound as the first step to diagnosis. The use of MRI for diagnosis over the years was noted to be 1999–2010 (4/18) – 2011–2023 (17/38) (p = 0.14).

3.4. Clinical management (Table 3)

Of the 80 included patients, 61 (76.3 %) received conservative management as the first step to treat IGU, of which 26 (42.6 %) had more than one attempt in using different types of conservative management. Initial conservative management performed on patients were Foley catheter placement alone to allow rectification of the uterine position via bladder decompression (N = 18, 29.5 %), bladder catheterization with positional change such as knee chest or Sim's position (N = 10, 16.4 %), manual reduction attempted either vaginally or rectally using colonic insufflation (N = 26, 42.6 %). Studies noted that only 7 patients (11.5 %) had all the 3 types of conservative management together. No patients had a reported spontaneous resolution of IGU. The median number of attempts of conservative management was 1, IQR [1–2]. Of the 61 patients, 7 (11.5 %) failed conservative management and hence underwent surgical management such as laparotomy to manually release the uterus entrapped under the sacral promontory at 16 weeks (N = 1, 14.3 %). Three patients had laparotomic myomectomy at 7, 9, and 22 weeks of gestation, respectively (42.9 %). Two patients underwent lysis of adhesions between pelvic wall and posterior uterine surface: one patient at 25 weeks and the other at an unknown gestational age (N = 2, 28.6 %). One patient's report did not mention the details of the surgical management performed to rectify the IGU. It is important to note that 13 (16.3 %) patients had no attempts to correct IGU.

Of the 80 patients, 43 (53.7 %) had an uneventful pregnancy following management of IGU. Five (6.3 %) patients had incomplete obstetric outcome details in the articles. Of the remaining, 32 patients reported obstetric outcomes. The complications noted in this review were malpresentation (e.g., breech and brow) (N = 13, 40.6 %), preterm labor/PPROM (N = 12, 33.4 %), intrauterine fetal death (IUFD)/miscarriage (N = 4, 12.5 %), fetal growth restriction (FGR) (N = 4, 12.5 %), non-reassuring fetal heart (N = 1, 3.1 %), and placenta accreta (N = 1, 3.1 %).

Of the 80 included patients, 74 (92.5 %) reported mode of the delivery. Of these 74 patients, 36 (48.6 %) had a vaginal delivery (VD), 36 (48.6 %) underwent a cesarean delivery, and 1 (1.3 %) underwent a cesarean hysterectomy. One patient (1.3 %) had a miscarriage at 12 weeks and underwent suction curettage. Of the 36 patients who had a VD, 32 (88.9 %) had term VD and 4 (11.1 %) had preterm VD. Of the 36 patients who underwent a cesarean delivery, 20 (55.5 %) had a term delivery and 16 (44.5 %) had a preterm delivery. Three articles reported on recurrent IGU in a subsequent pregnancy.

3.5. Comparison of patients with and without risk factors (Table 4)

We grouped the patients included in this review (N = 80) into two categories- one in whom no risk factor could be identified to develop IGU (N = 44, 55 %) and patients in whom risk factors such as fibroids, adhesions, uterine or pelvic anatomical anomaly, pelvic infection, prior pelvic surgeries, and prior IGU were identified and developed IGU (N = 36, 45 %). Patients with risk factors were older than those without risk factors (33 vs 28 years, $p = 0.02$). There was no statistically significant difference in parity or median gestational age of diagnosis between the two groups (all $p > 0.05$). There was no delay in diagnosis, or increased chance of misdiagnosis in either group. No difference was noted in performing conservative measures as an initial treatment to correct IGU or in the number of attempts needed to correct IGU. No statistical difference was noted in the mode of delivery, obstetric outcome, or preterm delivery (all $p > 0.05$).

4. Discussion

With the advent of modern obstetrics and assisted reproductive techniques, there is an increase in pregnancies complicated by adhesive conditions like endometriosis, fibroids, PID, and multiple prior pelvic surgeries. This leads to the need for addressing the clinical epidemiology

Table 4

Patients with and without identified risk factors who developed IGU.

	Patients with identified risk factors (N = 44)	Patients without identified risk factors (N = 36)	P value
Age (N = 78), years	33 [29–37]	28 [26–33]	0.02
Parity			0.06
Nulliparity	30(68.2)	18(50)	
M	14(31.8)	15(41.7)	
GM	0	3 (8.3)	
Median Gestational age (N = 80), weeks	17 [13–28]	15.5 [14–21]	0.83
Patients diagnosed during 1st visit	21 (47.7)	25 (69.4)	0.06
Patients misdiagnosed initially	10 (22.7)	13 (36.1)	0.17
Patients in whom conservative management was performed first	34 (77.3)	27 (75)	0.09
If more than one attempt was performed (N = 57)	15 (34.1)	10 (27.8)	0.76
Patients with successful conservative management (N = 70)	11 (25)	10 (27.8)	0.12
Surgical measure performed to correct incarcerated uterus	8 (18.2)	9 (25)	0.10
Complicated obstetric outcome	20 (45.5)	15 (41.7)	0.64
Preterm vaginal delivery	4 (9.1)	2 (5.6)	0.12
Preterm cesarean delivery	7 (15.9)	8 (22.2)	0.50

M, multipara; GM, grand multipara.

Data presented as median [IQR] or N (%), N/N (%) if missing.

of IGU. Several risk factors have been noted by case reports that prevent the ascend of the gravid uterus out of the sacral hollow [13,28,30,33,6,7]. In this systematic review, we found that the presence of risk factors neither made a difference in the gestational age at which patients presented with complaints, diagnosis, and clinical management of IGU, gestational age of delivery, nor the mode of delivery. A gravid retroverted uterus typically enlarges during early second trimester and may not correct its axis to become an abdominal organ regardless of the presence of a risk factor. This leads to compression or obstruction of surrounding structures, like the bladder and bowel and precipitate symptoms.

Our review emphasizes the importance of clinical examination in any pregnant patient with abdominal pain and complaints pertaining to urinary or bowel habits. A severely anteriorly displaced cervix, which is unpalpable and difficult to visualize using a sonogram should raise the suspicion for IGU [4,6,5]. While reports noted the usefulness of ultrasound findings of IGU [9,12,15], some cases stated that MRI helped to better delineate anatomy and in preoperative planning to perform a cesarean delivery in inverted polarity [2].

The literature suggests that it is preferable to do a surgical correction for patients who developed IGU beyond 20 weeks [65]. Based on this review, it would be reasonable to state that one could attempt conservative measures as an initial management to rectify IGU regardless of the gestational age. However, the number of attempts or a type of conservative method cannot be definitively recommended based on this review. A novel method of conservative management such as using an inflated Bakri balloon in the posterior fornix has recently been added to literature [61]. However further prospective studies are required to establish new methods of management of IGU. Case reports have stated that when IGU is corrected, pregnancy can proceed until term [16,17,20]. Though there were reports of preterm deliveries or complications such as abruption and growth restricted fetus, we did not find a statistically significant increased occurrence among patients with identified risk factors for IGU. This finding, however, depends on the accuracy of

the reports.

4.1. Limitations and strengths

This is a recent and large review of IGU. We were able to compare the clinical characteristics of patients, management, and obstetric outcomes of IGU between patients with and without identified risk factors. However, our comparison depends solely on the publisher's mention of a risk factor. Risk factors other than IGU that could contribute to complicated obstetric outcomes were not mentioned in the articles. Cases included were identified from a comprehensive search of databases using a systematic search strategy. However, we are unable to rule out the possibility of missing some important cases aggregated in larger series, given that some individual patient data was unavailable in these series. Publication bias is another limiting factor as case reports of rare or atypical observations are more likely to be published, thus excluding the more common findings. The tool applied for assessing risk of bias of case reports and case-series is derived from a tool that has been used in many previously published systematic reviews compiling case reports and series.

4.2. Practice and research implications

Findings from the current review suggest that it is difficult to make an association between various predisposing risk factors and IGU. Clinical examination and ultrasound remain the most common means of diagnosis, but studies report a good utility of MRI to diagnose IGU, and specifically inverted polarity of the uterus, thereby avoid misdiagnosis of placenta previa and breech and plan the uterine incision preoperatively.

5. Conclusion

It is important to consider IGU in pregnancies with lower abdominal pain and urinary retention. Obstetric outcomes and management of IGU does not differ between patients with and without identified risk factors. Despite advances in imaging technology, IGU is frequently misdiagnosed. It is reasonable to start with conservative management to rectify IGU regardless of the gestational age of diagnosis.

Declaration of Competing Interest

The authors declare that they have no known competing financial interests or personal relationships that could have appeared to influence the work reported in this article.

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Appendix A. Supporting information

Supplementary data associated with this article can be found in the online version at [doi:10.1016/j.eurox.2023.100227](https://doi.org/10.1016/j.eurox.2023.100227).

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