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REVIEW

Round ligament varicosity: a rare but important differential diagnosis of groin lumps during pregnancy

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Abstract

Round ligament varicosity is rare and mainly reported during pregnancy. A systematic review of the literature identified 48 relevant studies reporting a total of 159 cases of round ligament varicosity, 158 of which were associated with pregnancy. Where reported, the mean age of the patients was 30.65 years, and 60.2% were of Asian ethnicity. The laterality of the condition was almost equally distributed, and nearly 50% presented with a painful groin lump. More than 90% of the patients were diagnosed via Doppler ultrasound scan of the affected groin. Conservative management was successful in more than 90% of the patients. Associated maternal complications are rare, with no mortality reported. No fetal complications or loss were reported. Round ligament varicosity can be misdiagnosed as a groin hernia, which may lead to unnecessary surgery during pregnancy. Therefore, increased awareness of this condition among clinicians is important.

Key words: Groin lump; Inguinal hernia; Pregnancy; Round ligament; Round ligament varicosity

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Introduction

The round ligament or ligamentum teres uteri is a rope-like structure that develops from the gubernaculum. Both round ligaments seem to play a role in keeping the uterus in position later in life (García-Paredes et al, 2017). The round ligament is about 10–12 cm long and starts from the lateral part of the uterus on each side, then passes laterally across the external iliac vessels before it enters the inguinal canal through the deep inguinal ring, and finally terminates at the labia majora. It mainly consists of fibro-muscular tissue, as well as lymphatics, nerves, arteries and veins that drain into the inferior epigastric vein within the inguinal canal (Ijpma et al, 2009; García-Paredes et al, 2017; Naik and Balasubramanian, 2019). The round ligaments are prone to develop pathologies such as endometriosis, fibroids, mesenchymal tumours and varicosity (Tokue et al, 2008; Huang et al, 2013; Ryu and Yoon, 2014; Ng and Wong, 2019).

During pregnancy, the round ligaments stretch and get longer and wider, and can lead to the sensation of a common physiological symptom that is often described by patients as groin pain, pulling or pressure. This can sometimes be associated with stretching of the deep inguinal ring and also groin swelling in the form of round ligament varicosity (Lechner et al, 2020; Yeo and Tashi, 2021).

Round ligament varicosity is rare and seems to occur almost exclusively in pregnancy. It can sometimes extend into the parametrium and can be simultaneously present with pelvic vein congestions and varicosities in the lower limbs (Jung et al, 2009).

The exact pathophysiology of round ligament varicosity is not well understood. The most common explanation is based on the anatomical location of the round ligaments, where pelvic venous drainage can be delayed because of the gradual increase in the size of the gravid uterus that puts pressure on the pelvic structures throughout pregnancy. This can lead to development of round ligament varicosity (Cheng et al, 1997; Ijpma et al, 2009). However, other theories have been proposed, including the relaxation of venous smooth muscles resulting in loss of vascular resistance during pregnancy through the effect of hormones on oestrogen and progesterone receptors. Furthermore, the high cardiac output and increased blood volume increases venous return, resulting

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in congestion of the pelvic veins by the gravid uterus, which can be worsened by the presence of incompetent valves in most pelvic veins. All these factors could play a role in this condition (Lee et al, 2011; Ryu and Yoon, 2014; Depboylu, 2016; Ng and Wong, 2019).

Patients with round ligament varicosity usually present later in pregnancy with a painless unilateral or bilateral groin swelling that often disappears (spontaneously reduced) when the patient lies flat. Some patients might also have associated signs suggestive of varicosity of the labia and/or lower limbs. Patients do not usually show the classical clinical picture of gastrointestinal obstruction that is usually associated with an incarcerated or strangulated hernia. However, the swelling can sometimes be painful and irreducible, which can lead to it being misdiagnosed as incarcerated or strangulated groin hernia on standard clinical assessment (Al-Qudah, 1993; Ng and Wong, 2019). In practice, a diagnosis of incarcerated or strangulated hernia in the general population is mostly based on clinical assessment, and normally warrants emergency surgery under general anaesthesia without preoperative radiological confirmation of the diagnosis. A misdiagnosis of round ligament varicosity as incarcerated or strangulated hernia may lead to a pregnant patient undergoing acute surgery under general anaesthesia; as both surgery and anaesthesia can be associated with risks to mother and fetus (Al-Qudah, 1993; Ng and Wong, 2019), and round ligament varicosity normally resolves after giving birth and only requires conservative management with reassurance of the patient (Lechner et al, 2020), this surgery is unnecessary.

Despite its rarity, round ligament varicosity is a clinically important condition during pregnancy that is relevant to clinicians from different specialties. This article presents a systematic review of the published evidence regarding the clinical presentation, diagnosis and management of this condition.

Methods

This systematic review was performed according to the Preferred Reporting Items for Systematic Reviews and Meta-Analyses (PRISMA) protocol. The search strategy was based on a combination of the medical subject heading (MeSH) vocabulary and key words (round ligament, round ligament varicosities/varicosity, pregnancy, varicose veins, groin lump, inguinal hernia and groin hernia). No timeframe or specific language was applied to the search strategy. Electronic search was performed of Medline/Pubmed, EMBASE and Google Scholar. Searches were screened and the full-text versions of studies thought to be relevant were retrieved. The reference lists of all retrieved texts were searched for further relevant studies. Inclusion criteria included published studies with a confirmed diagnosis of round ligament varicosity. The search strategy is summarised in Figure 1.

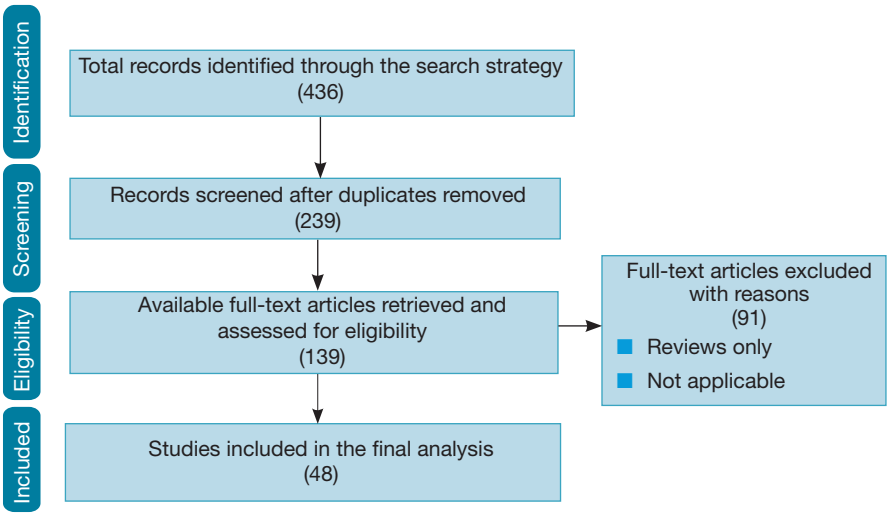

Figure 1. Flowchart of the search strategy.

Table 1. Levels of evidence	
Level I	Multiple randomised controlled trials or meta-analysis
Level II	Adequately powered single randomised controlled trial
Level III	Non-randomised designs
Level IV	Case reports or case series
Level V	Expert opinion

The final analysis included 48 studies: 45 case reports or series and three non-randomised studies, that reported a total of 159 cases of round ligament varicosity. Figure 1 shows the reasons for excluding other studies.

Tables were formulated to summarise the data from the included articles (Appendices 1 and 2). The reviewed evidence was graded as outlined in Table 1. Performing meta-analysis was not possible because of the nature of the available limited related evidence (addressed later on). This article does not contain any information that might allow patient identification. Ethical approval is not required for this type of article according to the authors' local policies.

Results

A total of 159 female cases were included in this review, with a reported mean age of 30.65 years. Round ligament varicosity was associated with pregnancy in all patients but one. Ethnic group was not mentioned in about half of reported cases; 60.2% of the remaining cases were Asian, 39.5% were Caucasian, and <0.5% were reported in other races.

The vast majority of cases presented during the early stages of the third trimester, with a mean of 25.74 weeks of gestation. Six patients (<5% of the cases) first presented with round ligament varicosity during the early postpartum period, specifically within 72 hours of giving birth.

A lump in the groin was the main presenting symptom; this was associated with pain or ache in about 60% of patients. The reducibility of the lump was not highlighted by the authors in more than two thirds of cases, while the remaining authors reported the round ligament varicosity as reducible, partially reducible or a non-reducible groin lump. Therefore, it was not possible to comment further on this clinical sign. Many cases reported spontaneous reducibility of the round ligament varicosity in the supine position. Associated clinical signs of lower limb and/or labial varicosity were also reported in <10% of patients. Around 25% of patients had bilateral round ligament varicosity, while the rest had the condition unilaterally with almost equal distribution between the right and left groins.

More than 90% of cases had the diagnosis established by a combination of clinical examination and radiological assessments through performing standard ultrasound scan with Doppler ultrasound scan of the affected groin. The classic findings are typically described as a hypoechoic mass formed by tubular structures (vessels) that usually have positive signal for venous blood flow (Figure 2). It has also been described as having the appearance of a 'bag of worms' along the inguinal canal (McKenna et al, 2008; Lee et al, 2011; Naik and Balasubramanian, 2019).

Magnetic resonance imaging or computed tomography scans were used in four cases because of the uncertainty of the diagnosis. The diagnosis of round ligament varicosity was established through clinical assessment only in eight patients (<5%), while the mode of diagnosis was not clearly reported in six cases.

More than 90% of cases had successful conservative management where the condition completely resolved soon after pregnancy. Around 5.6% of patients had surgical intervention through groin exploration because incarcerated or strangulated hernia was suspected. Interestingly, the patients who underwent surgery were preoperatively misdiagnosed as having groin hernia through standard clinical assessment only, while their intraoperative findings were of round ligament varicosity. Associated complications seemed to be rare (only reported in around 3% of patients early during the post-gestational period). No maternal deaths, fetal complications or losses were reported.

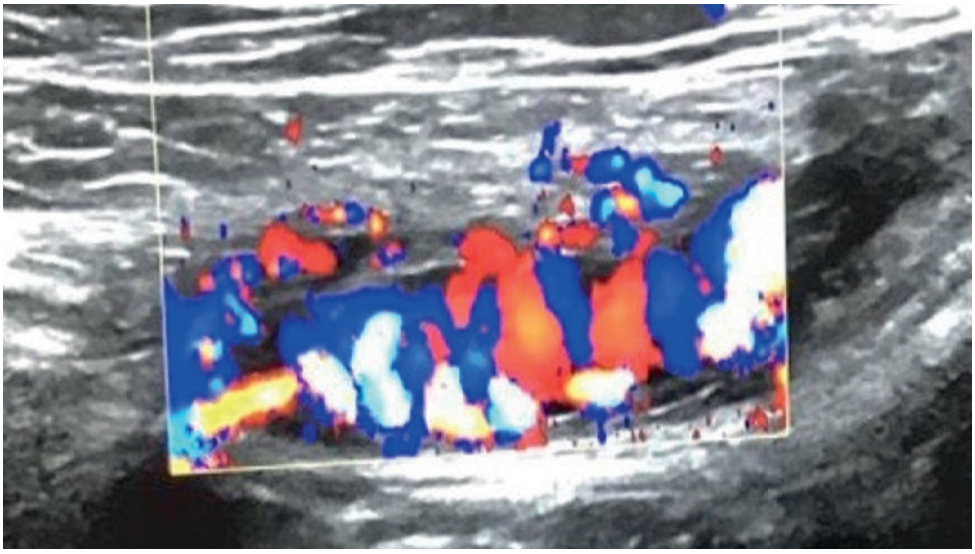

Figure 2. Doppler ultrasound scan longitudinal plane showing vascular structures in the right inguinal region with mainly venous blood flow (blue).

Discussion

Round ligament varicosity is an uncommon cause of unilateral or bilateral groin lumps that is mainly reported during pregnancy. It typically presents in the third trimester as a painless or painful groin swelling that can spontaneously reduce in the supine position. Round ligament varicosity has been suggested to occur more frequently on the left side, since pregnant women are usually advised to sleep on the left side to reduce pressure on the inferior vena cava and increase blood flow to the fetus (Cheng et al, 1997; Yonggang et al, 2017). However, the findings of this systematic review do not support this, as unilateral round ligament varicosity seemed to be equally distributed in both groins. Patients do not usually have an associated clinical picture of gastrointestinal obstruction, but pregnancy in general can be associated with nausea and vomiting. Associated labial and/or lower limb varicosity can also raise the suspicion of a diagnosis of round ligament varicosity, but this is not pathognomonic for the condition and was found in <10% of cases.

As with inguinal hernia, round ligament varicosity can present as a reducible or irreducible painless or painful groin lump, can transmit cough impulse and increase in size with the Valsalva manoeuvre via the associated increase in intra-abdominal pressure and distension of veins (Cheng et al, 1997; Chi et al 2005; Kahriman et al, 2010; Erok et al, 2016). Therefore, it is challenging to differentiate this condition from inguinal hernia by standard clinical assessment alone, particularly when it is associated with pain with or without irreducibility (Erok et al, 2016; Yonggang et al, 2017; Naik and Balasubramanian, 2019).

Although it is reported to be the most common cause of groin swelling in humans, inguinal hernia is not common during pregnancy, with an incidence of 1 in 1000–3000 pregnant women (Ijpma et al, 2009; Yaşar and Genez, 2021). One explanation for the rarity of inguinal hernia during pregnancy is that the gravid uterus displaces the abdominal contents upwards, which could theoretically lead to regression of pre-existing inguinal hernia (Tomkinson and Winterton, 1955; Cicilet, 2017; Mine et al, 2017). The incidence of round ligament varicosity was reported as around 0.13% in a study that involved more than 5000 pregnant patients (McKenna et al, 2008). These figures may indicate that round ligament varicosity is slightly more common than inguinal hernia in pregnant women, although there is no strong evidence to support this.

Doppler ultrasound scan of the affected groin or groins is the imaging modality of choice in establishing the diagnosis, particularly if it is combined with a Valsalva manoeuvre that leads to prominent venous flow through the round ligament. There is no routine indication for computed tomography or magnetic resonance imaging. The use of computed tomography in pregnancy is limited to cases where the diagnosis is uncertain and there is associated risk to the mother's life, such as multiple trauma or sepsis, where the potential pathology outweighs the very low risk of radiation and contrast to the fetus.

Once a diagnosis of round ligament varicosity is established, conservative management is indicated via patient reassurance with simple analgesia if there is associated pain. Complications such as thrombosis and rupture or bleeding seem to be rare and are mostly reported in the postpartum period. Thrombosis of round ligament varicosity is not usually associated with life-threatening outcomes, but the use of a short course of anticoagulation has been advised by a few authors in these cases. Venous rupture and haemorrhage are usually self-limiting, and compression of the affected inguinal area should be sufficient to stop the bleeding (Yonggang et al, 2017; Lechner et al, 2020).

There is no routine role for surgery in the management of round ligament varicosity, unless the diagnosis is uncertain despite thorough clinical and radiological assessment, for example when round ligament varicosity presents as an acute painful mass in the groin and strangulation or bowel obstruction cannot be ruled out.

Limitations

This systematic review has been based on analysis of low grade level III and IV published evidence because of the rarity of the condition. There is inconsistency and heterogeneity of the reported data. This is expected given that the studies were conducted worldwide over a number of decades, so achieving significant statistical outcomes through performing meta-analysis was not possible. Nevertheless, low grade evidence can directly reflect real-life clinical practice, and can provide valuable information and highlight the practical challenges of applying evidence to real-life practice.

Conclusions

Round ligament varicosity is uncommon and is almost exclusively reported during the third trimester of pregnancy. It is difficult to differentiate this from groin hernia through clinical assessment only, because of the similarity of their clinical presentations. A misdiagnosis of round ligament varicosity as a groin hernia may lead to unnecessary surgical intervention under general anaesthetic during pregnancy, with risks to mother and fetus. A high index of clinical suspicion combined with radiological assessment by Doppler ultrasound scan is essential to establish the diagnosis. Increasing awareness of this rare but important differential diagnosis of groin lumps during pregnancy is important in daily practice.

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Conflicts of interest

The authors declare that there are no conflicts of interest.

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Key points

- Round ligament varicosity is rare and is almost exclusively reported during the third trimester of pregnancy.
- It is difficult to differentiate this condition from groin hernia through clinical assessment only because of the similarity of their clinical presentations.
- A high index of clinical suspicion combined with radiological assessment by Doppler ultrasound scan is essential to establish the diagnosis and avoid unnecessary surgical intervention during pregnancy.
- Despite its rarity, round ligament varicosity should be a differential diagnosis of painless or painful groin lumps during pregnancy

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Appendix 1. Summary of the analysed case report and case series

Reference, country, level of evidence	No. of patients	Age (years)	Ethnicity	Gestation age at the time of presentation	Para	Gravida	Previous pregnancy similar episode	Side	Clinical history duration	Painful	Examination	Past history	Associated with lower limb/ labia varicosity	Investigation	Prominent on Valsalva clinically / USS	Intra-operative or other findings	Thrombosis present	Management	Follow up, duration, exam type, outcome
Yaşar and Genez (2021), Turkey, level IV	2	29	?	?	?	?	Yes	B/L	?	?	Enlarging on standing	?	Yes B/L	USS-anechoic tubular structures, DUSS- venous flow	Yes	N/A	?	Conservative	6 months, clinical and radiological, resolved
		36	?	First trimester	?	3	Yes	B/L	?	Yes	?	?	Yes B/L	USS-serpentine VV, DUSS- VV with reflux flow	Yes	N/A	?	Conservative	6 months, clinical and radiological, resolved
Patel et al (2021), USA, level IV	1	31	?	16 weeks	?	?	?	R	8 weeks	No	Painless, reducible mass	Varicose veins in previous pregnancy	Yes, on MRI	USS- prominent vessels + MRI-prominent vessels	?	N/A	No	Conservative	no F/U, resolved
Yeo and Tashi, (2021), Singapore, level IV	1	35	Asian	26 weeks	0	1	?	R	2 weeks	No	Soft, non-tender, non-mobile and non-reducible	Right inguinal hernia-conservatively managed	No	USS-echo-free tubular structures, DUSS- vascularity with venous flow	Yes	N/A	Nov	Conservative	8 weeks, clinical, resolved
Allorsey (2021), Ghana, level IV	1	27	?	?	0	1	?	B/L	?	Yes	Warm, immobile, disappear on supine	?	?	USS- hypoechoic tubular structures, DUSS- vascular	Yes	N/A	No	Conservative	?
Naik and Balasubramanian, (2019), India, level IV	1	28	?	30 weeks	?	2	none	L	2 weeks	Yes	Painful, non-reducible, soft	None	?	USS-anechoic serpentine tubular structures, DUSS- venous flow	Yes	N/A	No	Conservative	Resolved
Ng and Wong (2019), Hong Kong, level IV	1	35	Chinese	7 days post-partum	1	2	none	R	1 day	Yes	Irreducible, cough impulse absent, erythema present	?	?	USS- hypoechoic serpentine structures, DUSS- No flow signal, CT scan	No	CT scan- hyperdense, serpentine tubular structure extending from the right adnexa right labia majora	Yes	Conservative, low molecular weight heparin 40 mg for 6 weeks	6 weeks, radiological, resolved
Dazzi and Schiavon (2019), Italy, level IV	1	36	?	30 weeks	?	?	?	L	?	Yes	?	?	?	DUSS	?	N/A	?	?	?

Appendix 1. Summary of the analysed case report and case series (continued)

Reference, country, level of evidence	No. of patients	Age (years)	Ethnicity	Gestation age at the time of presentation	Para	Gravida	Previous pregnancy similar episode	Side	Clinical history duration	Painful	Examination	Past history	Associated with lower limb/ labia varicosity	Investigation	Prominent on Valsalva clinically / USS	Intra-operative or other findings	Thrombosis present	Management	Follow up, duration, exam type, outcome
Walchhofer et al (2019), Germany, level IV	2	35	?	35 weeks	?	?	?	R	?	Yes	Reducible	?	?	USS- anechoic, tubular structure, DUSS- slow flow signals	Yes	N/A	?	Conservative	Resolved
		39	?	30 weeks	?	?	?	L	?	Yes	?	?	?	USS- anechoic tubular structures, DUSS- venous flow signals	Yes	N/A	?	Conservative	Resolved
Estudillo et al (2018), Mexico, level IV	1	26	?	26 weeks	?	?	?	L	6 weeks	Yes	Painful, reducible,	None	?	USS- tubular structures, DUSS- vascularity present	?	N/A	No	Conservative	2 weeks, clinical, resolved
Cicilet (2017), India, level IV	1	22	?	30 weeks	0	1	?	R	6 weeks	Yes	Soft, non-reducible	?	?	USS- anechoic tortuous tubular, DUSS- venous flow	Yes	N/A	No	Conservative	3 weeks, radiological, resolved
Mine et al (2017), Japan, level IV	10	37	?	28 weeks	0	1	?	L	2 weeks	No	Soft painless	Toxoplasma infection	Yes	USS- tubular structures, DUSS- venous flow	?	N/A	?	Conservative	2 weeks, resolved
		22	?	30 weeks	1	?	?	R	?	Yes	?	?	?	USS+DUSS	?	N/A	?	Conservative	2 weeks, resolved
		27	?	32 weeks	1	?	?	L	?	No	?	?	?	USS+DUSS	?	N/A	?	Conservative	2 weeks, resolved
		29	?	26 weeks	1	?	?	L	?	Yes	?	?	?	USS+DUSS	?	N/A	?	Conservative	2 weeks, resolved
		27	?	19 weeks	3	?	?	R	?	No	?	?	?	USS+DUSS	?	N/A	?	Conservative	2 weeks, resolved
		31	?	26 weeks	2	?	?	L	?	Yes	?	?	?	USS+DUSS	?	N/A	?	Conservative	2 weeks, resolved
		33	?	20 weeks	2	?	?	L	?	Yes	?	?	?	USS+DUSS	?	N/A	?	Conservative	2 weeks, resolved
		25	?	20 weeks	2	?	?	L	?	Yes	?	?	?	USS+DUSS	?	N/A	?	Conservative	2 weeks, resolved
		31	?	13 weeks	2	?	?	R	?	No	?	?	?	USS+DUSS	?	N/A	?	Conservative	2 weeks, resolved
		33	?	17 weeks	1	?	?	L	?	Yes	?	?	?	USS+DUSS	?	N/A	?	Conservative	2 weeks, resolved

Appendix 1. Summary of the analysed case report and case series (continued)

Reference, country, level of evidence	No. of patients	Age (years)	Ethnicity	Gestation age at the time of presentation	Para	Gravida	Previous pregnancy similar episode	Side	Clinical history duration	Painful	Examination	Past history	Associated with lower limb/ labia varicosity	Investigation	Prominent on Valsalva clinically / USS	Intra-operative or other findings	Thrombosis present	Management	Follow up, duration, exam type, outcome
Çiçek and Yardimicioglu (2017), Turkey, level IV	1	31	?	29 weeks	?	3	No	R	2 weeks	Yes	Groin mass	?	?	USS- anechoic tortuous tubular, DUSS- venous flow	?	N/A	No	Conservative	8 weeks, radiological, resolved
García-Paredes et al (2017), Puerto Rico, level IV	1	34	Hispanic	34 weeks	0	1	No	R	2 weeks	No	Small painless reducible	None	No	USS- serpentine tubular structures, DUSS- hyper vascular venous flow	Yes	N/A	No	Conservative	2 weeks, resolved
Depboylu (2016), Turkey, level IV	1	28	?	32 weeks	?	2	No	R	3 weeks	Yes	Reducible	none	No	USS- multiseptated, anechoic tubular, DUSS- filled with colour-blood	Yes	N/A	No	Conservative	4 weeks, resolved
Bulbul et al (2015), Turkey, level IV	1	32	?	32 weeks	1	2	No	B/L	16 weeks	No	Reducible soft mass	None	Up to labium major	USS- anechoic tortuous tubular, DUSS- venous flow	Yes	N/A	No	Conservative	4 weeks, resolved
Erok et al (2016), Turkey, level IV	1	31	?	28 weeks	?	?	Yes	L	2 weeks	Yes	Tender, reducible soft mass	?	Lower extremity varices	USS- anechoic, tubular channels, DUSS- abundant venous flow	Yes	N/A	No	Conservative	Radiological, resolved
Ryu and Yoon (2014), Korea, level IV	2	33	?	Not pregnant	2	?	No	L	N/A	Yes	Palpable mass	?	?	USS- dilated echo-free, tubular channels, DUSS- hypervascularity	Yes	N/A	?	Conservative	?
		28	?	21 weeks	0	1	N/A	L	1 week	Yes	Tender soft mass	?	?	USS- echo-free serpentine tubular, DUSS- hypervascularity and venous flow	Yes	N/A	No	Conservative	3 weeks, resolved
Polat et al (2013), Turkey, level IV	5	27	?	28 weeks	2	?	No	B/L	15 weeks	Yes	Partially reducible	?	?	USS- echo-free, serpentine, tubular channels, DUSS- hypervascularity and venous flow	Yes	N/A	No	Conservative	5 weeks, resolved
		30	?	30 weeks	3	?	Yes	B/L	9 weeks	Yes	Partially reducible	?	?		Yes	N/A	No	Conservative	4 weeks, resolved
		28	?	24 weeks	2	?	Yes	R	10 weeks	Yes	Partially reducible	?	?		Yes	N/A	No	Conservative	6 weeks, resolved
		29	?	21 weeks	2	?	Yes	R	5 weeks	Yes	Partially reducible	?	?		Yes	N/A	No	Conservative	8 weeks, resolved
		25	?	31 weeks	2	?	Yes	R	13 weeks	Yes	Partially reducible	?	?		Yes	N/A	No	Conservative	6 weeks, resolved

Appendix 1. Summary of the analysed case report and case series (continued)

Reference, country, level of evidence	No. of patients	Age (years)	Ethnicity	Gestation age at the time of presentation	Para	Gravida	Previous pregnancy similar episode	Side	Clinical history duration	Painful	Examination	Past history	Associated with lower limb/ labia varicosity	Investigation	Prominent on Valsalva clinically / USS	Intra-operative or other findings	Thrombosis present	Management	Follow up, duration, exam type, outcome
Huang et al (2013), China, level IV	1	28	?	32 weeks	?	?	?	L	20 weeks	Yes	Partially reducible	?	?	USS- echo-free serpentine tubular channels, DUSS- multiple dilated veins	Yes	USS- the varicosities extended from the left labia majora to the abdominal cavity	No	Conservative	2 weeks, resolved
Etsuko et al (2012), Japan, level IV	2	29	?	30 weeks	?	?	?	R	?	No	Soft, reducible	?	?	USS- hypoechoic oval mass, DUSS- blood flow	Yes	N/A	?	Conservative	2 weeks, radiological, resolved
		36	?	28 weeks	?	?	?	L	?	Yes	Soft, reducible	?	?	USS- well-defined hypoechoic spindle-shaped mass, DUSS- blood flow	Yes	N/A	?	Conservative	0 weeks, resolved
Leung (2012), Hong Kong, level IV	1	30	?	24 weeks	0	1	No	L	4 weeks	No	Partially reducible	None	No	USS- multiple anechoic serpentine tubular channels, DUSS- hypervascular venous flow	Yes	USS- extended to the left parauterine space	No	?	?
Heymans et al (2011), Belgium, level IV	1	24	?	25 weeks	?	?	?	L	1 week	No	Reducible	?	?	USS- anechoic structure, DUSS- venous flow	Yes	?	?	Conservative	Resolved
Baek et al (2011), Korea, level IV	1	32	?	23 weeks	?	?	?	L	3 weeks	Yes	Soft mass, partially reducible	?	No	USS- echo free serpentine tubular channels, DUSS- good flow signals MRI	Yes	MRI showed a multicystic tubular structure coursing from the left inguinal canal into the labia major	No	Conservative	5 months, clinically, resolved
Athwal and Hoar (2011), UK, level IV	3	26	Asian	35 weeks	?	?	?	R	12 weeks	Discomfort	Soft, palpable and non-tender. reducible	?	?	USS+DUSS- multiple large varicosities	?	?	?	Conservative	8 weeks, clinically, resolved
		27	Caucasian	34 weeks	?	?	?	L	8 weeks	No	Non-tender soft reducible	?	?	USS+DUSS- multiple large varicosities	?	?	?	Conservative	6 weeks, clinically, resolved
		28	Caucasian	24 weeks	?	?	?	R	8 weeks	No	Non-tender soft reducible	Opposite side inguinal herniorrhaphy	?	USS+DUSS- multiple large varicosities	?	?	?	Conservative	4 weeks, clinically, resolved

Appendix 1. Summary of the analysed case report and case series (continued)

Reference, country, level of evidence	No. of patients	Age (years)	Ethnicity	Gestation age at the time of presentation	Para	Gravida	Previous pregnancy similar episode	Side	Clinical history duration	Painful	Examination	Past history	Associated with lower limb/ labia varicosity	Investigation	Prominent on Valsalva clinically / USS	Intra-operative or other findings	Thrombosis present	Management	Follow up, duration, exam type, outcome
Lee et al (2011), Korea, level IV	1	29	?	36 weeks	?	2	No	R	4 weeks	Yes	Reddish skin, oedematous tender, reducible	?	?	USS- multiple, echo-free, tubular Channels, DUSS- hypervascular and abundant venous flow	?	None	No	Conservative	2 weeks, resolved
Kahriman et al (2010), Turkey, level IV	1	22	?	28 weeks	0	1	N/A	B/L	1 week	Yes	Soft painful, reducible	?	?	USS- bilateral inguinal cystic mass, DUSS- VV	Yes	?	?	Conservative	4 weeks, clinically, resolved
Dent et al (2010), UK, level IV	1	28	?	20 weeks	0	1	N/A	L	Several days	Yes	Tender palpable lump, reducible	Laparotomy for perforated appendix	?	USS- leash of veins extending into the inguinal canal	?	None	?	Conservative	Following delivery, clinically, resolved
Jung et al (2009), Korea, level IV	1	32	?	29 weeks	?	?	?	R	1 week	Yes	Tender mass	Appendictomy	?	USS- cystic and tubular lesion, DUSS- flow augmentation	Yes	?	?	Conservative	4 weeks, clinically, resolved
Copete et al (2010), Spain, level IV	1	32	?	20 weeks	?	?	?	R	?	No	Soft consistency	?	?	USS- multiple tubular structures, DUSS- flow present	?	?	?	Conservative	8 weeks, clinically, resolved
Uzun et al (2009), Turkey, level IV	1	24	?	26 weeks	?	?	?	R	?	Yes	Small tender soft mass in the right groin	?	?	USS- echo-free serpentine tubular channels, DUSS- hypervascularity and venous flow	Yes	None	No	Conservative	2 weeks, resolved
Videhult and Jansson (2009), Sweden, level IV	2	32	?	Third trimester	?	?	?	B/L	?	Yes	?	?	?	?	?	?	?	Conservative	Couple weeks, resolved
		38	?	14 weeks		3	Yes	R	?	Yes	?	Appendictomy	?	?	?	Intraop- greatly dilated veins in the ligamentum rotundum	?	Surgery- 21 weeks pregnancy, no resection done	Ongoing pain throughout pregnancy, Tx- dalteparin sodium
Andriessen et al (2009), The Netherlands, level IV	3	34	?	29 weeks	?	?	?	B/L	Several	Yes	Reducible	Nil	Varix of labia	USS+DUSS- extensive varices	?	?	?	Conservative	Resolved
		28	?	27 weeks	?	?	?	R	Several	No	Reducible	Nil	?	USS+DUSS- multiple varices	?	?	?	Conservative	Resolved
		39	?	25 weeks	?	3	?	R	?	No	Painless	Nil	?	USS+DUSS- superficial venous collaterals	Yes	?	?	Conservative	Resolved

Appendix 1. Summary of the analysed case report and case series (continued)

Reference, country, level of evidence	No. of patients	Age (years)	Ethnicity	Gestation age at the time of presentation	Para	Gravida	Previous pregnancy similar episode	Side	Clinical history duration	Painful	Examination	Past history	Associated with lower limb/ labia varicosity	Investigation	Prominent on Valsalva clinically / USS	Intraoperative or other findings	Thrombosis present	Management	Follow up, duration, exam type, outcome
Tokue et al (2008), Japan, level IV	1	37	?	6 days post NVD	?	?	None	R	?	Yes	Serpentine and tender mass	Hysteromyomectomy	No	USS- hypoechoic moniliform mass, CT- well-demarcated serpentine mass, MRI	?	MRI- signal intensity of the mass was relatively higher than that of the muscle, suggestive of an acute or subacute haematoma	Yes	Conservative	Slowly afterwards, resolved
Copeland (2008), USA, level IV	1	38	African	21 weeks	?	?	?	B/L	?	No	Soft and non-tender to palpation	?	?	USS- multiple, large varicosities, DUSS- varicosities	?	?	?	Conservative	16 days, resolved
Ijpma et al (2009), Netherlands, level IV	1	29	?	19 weeks	?	3	Yes	B/L	11 weeks	Yes	Soft painful, reducible	?	No	Clinical	Yes	Blue varicose veins arising from the round ligament, 2 weeks later developed opposite side RLV	No	Surgery at 22 weeks on right then 26 weeks at left	6 weeks, clinically, resolved
McKenna et al (2008), USA, level IV	5	35	?	23 weeks	2	3	No	L	?	No	Reducible	?	?	USS- ultrasound diagnosis made	?	N/A	?	Conservative	8 weeks, resolved
		35	?	30 weeks	1	4	?	R	?	No	Reducible	?	?	USS- multiple prominent vessels with venous flow, DUSS- compatible with varices	Yes	N/A	?	Conservative	8 weeks, resolved
		37	?	31 weeks	1	6	?	B/L	?	Yes	Painful groin bulges	Mitral prolapse	Yes - labia	USS- prominent engorged vessels	?	N/A	?	Conservative	8 weeks, resolved
		38	?	19 weeks	1	3	No	B/L	?	Yes	Groin swellings	Nil	?	USS- abnormal venous structures, DUSS- compatible with varices	?	N/A	?	Conservative	12 weeks, resolved
		40	?	Seconnd trimester	0	2	?	R	?	No	Palpable right groin mass	Nil	?	USS+DUSS- engorged blood vessels	?	N/A	No	Conservative	4 weeks, resolved
Murphy et al (2007), Ireland, level IV	1	37	?	28 weeks		2	No	B/L	3 weeks	Discomfort	Reducible with cough impulse		No	USS- multiple echo-free serpentine tubular channels, DUSS- flow detected	?	N/A	?	?	?

Appendix 1. Summary of the analysed case report and case series (continued)

Reference, country, level of evidence	No. of patients	Age (years)	Ethnicity	Gestation age at the time of presentation	Para	Gravida	Previous pregnancy similar episode	Side	Clinical history duration	Painful	Examination	Past history	Associated with lower limb/ labia varicosity	Investigation	Prominent on Valsalva clinically / USS	Intra-operative or other findings	Thrombosis present	Management	Follow up, duration, exam type, outcome
Nguyen and Gruenewald (2008), Australia, level IV	1	18	?	33 weeks	0	1	N/A	R	1 week	Yes	Small tender partially reducible	Iron deficiency anaemia	?	USS- circumscribed hypoechoic soft tissue mass, DUSS- venous flow	Yes	N/A	No	Conservative	1 month, persisted, underwent mesh repair
Chi et al (2005), UK, level IV	1	30	?	15 weeks	?	2	Yes	L	?	No	Reducible, soft, nontender	?	Yes	USS- multiple dilated varicosities	?	N/A	No	Conservative	6 weeks, clinically, resolved
Guillem et al (2001), France, level IV	1	27	?	24 weeks	?	?	?	R	2 weeks	Yes	Soft mildly painful, reducible	?	?	Clinical	?	Surgical exploration revealed varicosities – not resected	?	Surgery	3 months, resolved
Cheng et al (1997), Hong Kong, level IV	1	22	?	28 weeks	?	?	?	L	1 week	Yes	Soft mildly tender, partially reducible	?	No	USS- thickened left round ligament with varicosities, DUSS- good flow signals	Yes	N/A	?	Conservative	9 months, clinically, resolved
Buxton et al (1994), UK, level IV	1	37	?	1 day post NVD	3	3	?	R	1 day post NVD	Yes	Firm tender swelling	?	Yes	Clinical	?	Inflamed collection of thrombosed veins – excised	Yes	Surgery	?
Al-Qudah (1993), Jordan, level IV	2	27	?	1 day post NVD	?	?	?	L	1 day post NVD	Yes	Firm, tender, irreducible	?	?	Clinical	?	Surgery – oedematous, thrombosed varicose veins	Yes	Surgery	1 month, clinical, recovered
		29	?	3 days post-partum	?	?	?	L	3 days post-partum	Yes	Tender and firm mass	?	Extending to labia	Clinical	?	x	?	Conservative	1 week, clinical, resolved
Hodgkinson and Kroll (1957), USA, level IV*	5	?	?	28 weeks	0	1	?	?	?	?	Ovoid mass	?	?	?	?	?	One patient developed retroperitoneal haematoma after NVD	Conservative	Resolved
		?	?	14 weeks		Multi	Yes	?	?	?	Ovoid mass	?	?	?	?	?		Conservative	Resolved
Tomkinson and Winterton (1955), UK, level IV	2	36	?	10 weeks	?	3	Yes	B/L	?	?	No mass	?	Yes	Clinical	?	?	?	Conservative	2 months, clinical, resolved
		27	?	Right 1 day post NVD	0	1	N/A	L- second trimester, R- post NVD	R- 1 day NVD	?	R- irreducible	?	Yes	Clinical	?	Intraop- collection of thrombosed vessels in substance of round ligament	R- yes; L- no	R- surgery, L- conservative	2 months, clinical, R- recovered, L-resolved

*B/L = bilateral; CT = computed tomography; DUSS = Doppler ultrasound scan; F/U = follow up; L = left; MRI = magnetic resonance imaging; N/A = not applicable; No. = number; NVD = normal vaginal delivery; Op = operative; Pt = patient; R= right; RLV = round ligament varicosity; Tx = treatment; USS = ultrasound scan; VV = varicose veins; ? = unknown. * three further cases were resolved but no clinical details were given*

Appendix 2. Summary of the analysed non-randomised studies

Reference, study details	No of patients	Age	Ethnicity	Gestation age at the time of pregnancy	Para	Gravida	Side	Painful	Investigation	Thrombosis present	Management	Follow up, examination type, outcome
Orban et al (2020), Egypt, level III, prospective	13	Unknown	Unknown	Unknown	Unknown	Multigravida: five patients	Bilateral: four; right: five; left: four	Unknown	Doppler ultrasound scan	Unknown	Conservative	2–3 weeks, resolved
Lechner et al (2020), Austria, level III, prospective	28	Median 31.1 years	Caucasian	Unknown	Unknown	Unknown	Bilateral: six, left: six, right: 16	Unknown	Doppler ultrasound scan	Unknown	Conservative	68 months, clinical and radiological, resolved (one patient developed inguinal hernia)
Yonggang et al (2017), China, level III, retrospective	41	Median 29.7 years	Chinese	Median 25.8 weeks	Unknown	Unknown	Right: nine; left: 19; bilateral: 13	Yes: 25; no pain: five	Doppler ultrasound scan	None	Conservative	12–24 weeks, resolved