Articulos Originales

Adnexal cyst pedicle torsion during pregnancy: clinical experience of conservative management

Torsión de pedículo de quiste anexial durante el embarazo: experiencia clínica de manejo conservador

Maita Freddy; Niño de Guzmán, O.; Antezana Eufronio; Céspedes, Gaby; Hochstatter, Edwin

Maita Freddy *
freddymaita@hotmail.com
Hospital Obrero No2 of the CNS, Bolivia
O. Niño de Guzmán
Hospital Obrero No2, CNS, Bolivia
Antezana Eufronio
Hospital Obrero No2, CNS, Bolivia
Gaby Céspedes
Hospital Obrero No 2, Bolivia
Edwin Hochstatter
Hospital Obrero No 2, Bolivia

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gacetamedicaboliviana@gmail.com

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Abstract: Introduction: the adnexal masses in pregnancy range from 2% to 10%, the adnexal torsion is a rare cause of acute abdomen during pregnancy. Surgical management traditionally involved adnexectomy without detorsion of the compromised structure; recently conservative surgery was proposed which consists in detorsion and conservation of the affected annex.
Objective: to describe the value of ultrasound guided fine needle puncture and aspiration as the initially treatment in pedicle torsion of adnexal cyst during pregnancy.
Methods: prospective observational study of 9 patients that had the diagnosis of pedicle torsion of adnexal cyst during pregnancy who underwent ultrasound guided fine needle aspiration as a treatment of the adnexal cyst torsion between 2014 and 2019.
Results: in all cases it was possible to save the compromised structure; in two cases we observed recurrence of the cyst, that was subsequently removed during a caesarean section in both cases; in the rest of the patient's resolution was complete. Spontaneous abortion was seen in one of the patients, this event was not related to the invasive procedure.
Conclusion: ultrasound guided fine needle aspiration of an adnexal who has suffered of pedicle torsion seems to be a good alternative for the management of this acute condition.

Keywords: adnexal torsion, pregnancy, conservative management.

Resumen: Las masas anexiales en el embarazo oscilan entre el 2% a 10%; su torsión es causa rara de abdomen agudo durante el mismo. La conducta frente al diagnóstico fue, tradicionalmente, anexectomía sin detorsión del lado comprometido; posteriormente, detorsión y conservación del anexo torcido.
Objetivo: describir el valor de la punción y aspiración bajo guía ecográfica como tratamiento, inicialmente temporal, en cuadro de torsión de pedículo de quiste anexial durante el embarazo.
Métodos: estudio observacional prospectivo de 9 casos clínicos con diagnóstico de torsión de pedículo de quiste anexial y embarazo sometidos al tratamiento de punción aspiración del quiste torcido bajo guía ecográfica entre el año 2014 a 2019.
Resultados: en todos los casos fue posible salvar en anexo comprometido; en dos casos hubo persistencia del quiste que se

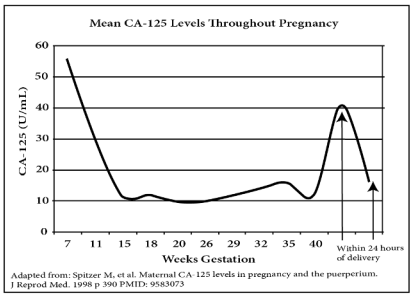
extirpó durante la cesárea; en el resto, la resolución fue completa. Hubo una pérdida de embarazo atribuible al cuadro clínico.
Conclusión: la punción-evacuación del quiste simple torcido parece ser una buena alternativa en la resolución del cuadro agudo.

Palabras clave: torsión de pedículo de quiste anexial, embarazo, manejo conservador.

Introduccion

The incidence of adnexal masses during pregnancy ranges from 2% to 10%; in most cases, they are diagnosed incidentally in the first trimester ultrasound screening, and this is the method of choice for their study due to its ability to differentiate their morphology and categorisation[1,2,14]

During normal pregnancy, CA 125 concentrations are variable (Graph 1); additionally, other tumour markers established as diagnostic aids are physiologically synthesised and secreted during fetal development[21]; in contrast, gestational ultrasonography allows morphological characterisation of the adnexal mass initially as a simple or complex cyst (Table 3), thereby stratifying risk without compromising maternal or fetal safety.



GRAPH 1.
CA 125 concentration in normal pregnancy[21]

Fortunately most adnexal masses in pregnancy are benign and resolve spontaneously[3,5,15-18,20]. However, they can become complicated by torsion, rupture, bleeding and infection. The risk of adnexal mass pedicle torsion increases 5-fold in pregnancy, with a reported incidence of 5 per 10 000 pregnancies[3,4,6].

The most frequently described predisposing factor to adnexal pedicle torsion is the presence of an ovarian cyst or tumour, accounting for up to 3% of surgical emergencies in gynaecology, depending on the published series[7,14,20].

The typical clinical picture of adnexal cyst pedicle torsion is manifested by the triad: pain, peritoneal irritation and impact in the general condition of patients with a previously identified adnexal tumor, although this picture is present in only slightly more than 50% of cases[7]. The presentation of adnexal torsion is similar in pregnant and non-pregnant women[7, 21].

Echography as a method of torsion diagnosis has been first described to identify the adnexal mass and to find non-specific signs such as free fluid. The use of color Doppler, initially promising, is currently shown to be

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* **Correspondence to**: Maita Freddy
E-mail: freddymaita@hotmail.com

non-determinant for diagnosis, with studies showing up to 19% of patients with pedicle torsion with normal preoperative Doppler in the affected adnexa[8]. Likewise, the absence of flow in the wall of the torsioned cyst is not diagnostic either, as one study shows a positive predictive value of only 69%, because some simple cysts, mainly para-ovarian ones, have little flow in their wall and do not capture the colour Doppler signal despite the optimisation of the gains[7].

The identification of the pedicle affected by torsion by real-time two-dimensional ecography, which consists of visualising the pedicle as a visible isoechogenic "spiral or swirling" bar between the cyst and the uterus, has substantially improved the diagnostic possibility, showing a positive predictive value of up to 87% for such a finding7.

Although the vast majority of cysts affected by torsion will be benign[7,9,19,22] pedicle torsion of an adnexal cyst is a surgical emergency that should be resolved immediately, regardless of its histopathological nature, gestational status or age.

There is evidence to support adverse outcomes in pregnant patients undergoing abdominal surgery, most frequently in the first trimester or after 24 weeks[2,19]. Until 2013, at the Hospital Obrero Nº 2 of the Caja Nacional de Salud, the traditional approach for the diagnosis of adnexal cyst with pedicle torsion was adnexectomy without detorsion of the involved side. Following numerous reports recommending management involving detorsion and preservation of the twisted adnexa for the majority of premenopausal patients[11,19,22], this conservative approach was adopted and a management protocol for temporary conservative treatment of pregnant patients with simple adnexal cyst with pedicle torsion was developed using aspiration puncture under ultrasound guidance.

Objective: to describe the value of temporary conservative treatment of adnexal cyst pedicle torsion during pregnancy by puncture aspiration under ultrasound guidance.

Material and methods

Prospective observational study of clinical cases with a diagnosis of simple adnexal cyst pedicle torsion with ongoing pregnancy, who underwent cyst aspiration puncture under ecography guidance as an initially temporary treatment, at the Hospital Obrero No 2 of the Caja Nacional de Salud, Cochabamba-Bolivia, between 2014 and 2019.

The study was approved by the Teaching and Research Committee of the GOB Service of Hospital Obrero No 2. Informed consent was obtained from all participants and confidentiality of all patients was maintained.

The ultrasound equipment used in the study was a Voluson E8 Expert model, for both transparietal and endocavitary scans.

The inclusion criteria for the study were: pregnant patient, from the initial ultrasound identification until 34 weeks (from which time it was considered low neonatal risk if gestation was terminated) with characteristic acute abdominal symptoms (classic triad) or with non-specific acute abdominal pain or both with ovarian or para-ovarian simple adnexal cyst with the presence, on ultrasound examination, of the direct sign of pedicle torsion defined as a visible "spiral or swirling" twisted pedicle between the pregnant uterus and the adnexal mass.

"Simple cyst" was defined as: unilocular ovarian or paraovarian cyst smaller than 10cm, thin-walled and regular, with an anechogenic liquid content or with low intensity echoes (Table 3) verified during the acute episode.

The presence of indirect signs such as absent vascular flow in the cyst wall, assessed by color Doppler and Power Angio® did not influence the initial decision for intervention or inclusion in the study.

The procedure was performed under strict aseptic antisepsis and sterile surgical drapes, using local anaesthesia with 1% lidocaine without epinephrine in all cases.

The aspiration puncture was performed under ultrasound observation of the tip of the catheter, Spinocan® 20 G x 3", at all times, until total collapse of the cyst involved. The material obtained from the puncture was sent for cytological study.

In patients with first trimester pregnancy, micronized progesterone 200 mg vaginally was indicated as a prophylactic measure to prevent miscarriage up to 14 weeks.

The nine patients in the study were recorded by age, gestational age at the time of the procedure, time elapsed since the onset of symptoms, symptoms on admission, size and characteristics of the adnexal cyst, immediate maternal and fetal post-puncture effect, length of hospitalisation, relevant laboratory tests and subsequent evolution.

Results

Details of the cases undergoing puncture-evacuation of the adnexal cyst compromised by pedicle torsion during pregnancy are shown in Table 1.

N°	Maternal age (years)	Size of twisted adnexal cyst	EG Weeks	Time of sympto-matology onset (hrs)	Macroscopic puncture findings	Maternal Complication	Fetal Complication	Internment (days)	Evolution
1	32	52mm for right ovary	16	3	70cc serohematic liquid	None, immediate relief	No	3	C-section at 40 week normal ovaries 30mm paraovarian cyst
2	29	95mm for right ovary	17	14	420cc serous liquid	None, immediate relief	No	2	Normal delivery at 39 weeks Normal control ultrasound
3	28	60mm left ovary	34	12	80cc serohematic liquid	None, immediate relief	No		C-section at 39.2 weeks Normal ovaries
4	23	65mm right annex	11	8	90cc citrine liquid	For +/-30 minutes • increased nausea and vomiting • FID* and flank pain • Chills • Thirst • Anxiety	No	3	C-section at 39,3 weeks, intraoperative finding: normal ovaries
5	35	60 mm in right annex	7	72	110cc of citrine liquid	None, immediate relief	Abortion on day 5	4	Eterine evacuation, normal posterior ultrasound study
6	39	52 mm right ovary	12	3	80cc of citrine liquid	None, immediate relief	No	2	Abortion at 16 weeks, not attributed to puncture.
7	32	80 mm in right ovary	17	8	200cc of citrine liquid	None, symptomatic relief after 4 hours	No	2	Caesarean section at 40 weeks. Normal ovaries
8	23	57 mm for - right ovary	21	12	80cc of serohematic liquid	None, symptomatic relief after 10 min	No	6	Normal term delivery. Control ultrasound shows normal adnexa.
9	29	62 mm in right annex	30	4	135cc of serohematic liquid	None, immediate relief	No	3	Cesarean section for term pregnancy. Intraoperative finding: simple paraovarian cyst in right adnexa of 20mm.

*FID: right iliac fossa

TABLE 1.
Summary of clinical cases of adnexal cyst pedicle torsion during pregnancy with aspiration puncture as temporary conservative treatment.

Ten pregnant patients presented with adnexal cyst pedicle torsion during the study period, nine of whom met the inclusion criteria. The case not reported in the study was a case of 110 mm adnexal cyst pedicle torsion during pregnancy resolved by laparotomy with resulting histopathology as serous cystoadenofibroma.

All nine cases in the study (9/9) presented mild discomfort at the time of puncture for local anaesthesia and aspiration of the torsioned cyst.

The mean age at presentation was 30 years (23 to 39 years); the mean pre-puncture-evacuation cyst size was 64 mm (52 to 94 mm) measured in three dimensions; the mean gestational age at presentation was 18 weeks (7 to 34 weeks); the time between symptom onset and the interventional procedure ranged from 3 to 72 hours. Eight (8/9) patients had relief of symptoms within 10 minutes; one patient (1/9) had exacerbation of symptoms immediately after the procedure with increased pain, nausea, vomiting, chills, thirst and distress for about 30 minutes and then spontaneously relieved all symptoms. In one case (1 of 9) the pregnancy ended in miscarriage on the fifth day of a 7-week pregnancy; in another case another miscarriage occurred four weeks after the procedure at 12 weeks. Seven pregnancies were carried to term, of which five were terminated by caesarean section and two by normal delivery. In seven patients (7/9) remission of the adnexal cyst was observed, verified by ultrasound studies and during caesarean section in patients who terminated the pregnancy by this route; in two patients (2/9) two remaining para-ovarian cysts of 30 and 20 mm were observed, the two cases of remaining cyst were removed during caesarean section, the final histopathological study was papillary cystadenofibroma in both cases (Table 2). In all cases (9/9) cytology was negative for malignancy. The average length of hospital stay was 2.7 days (2 to 6 days).

Cytological study		Histopathologic*	
Benign cystic lesion and/or acellular amorphous material	100%	Papillary serous cystoadenofibroma	2 cases

**by excision of the remaining cysts, during cesarean section*

TABLE 2

cytologic finding of fluid obtained from twisted adnexal cyst by aspiration puncture during the acute condition and histopathology of excision of the remaining cyst during cesarean section. N: 9

Discussion

Adnexal cyst pedicle torsion during pregnancy is a rare condition, constituting a rare cause of acute abdomen, with an incidence of 5 per 10 000 pregnancies[13].

The characteristic clinical picture is a triad of pain, peritoneal irritation, and impact in the general state; if localised pain is present, it is most intense in the corresponding iliac fossa and may range from diffuse discomfort to disabling pain and shock, which is present in slightly more than 50% of cases and may be accompanied by general condition compromise, hypotension, tachycardia[2,6,7,10,11]. In other cases, symptoms may be non-specific and can be confused with acute abdominal conditions such as acute appendicitis, ureteral colic, cholecystitis and intestinal obstruction[7,12,13].

The visualisation of the direct sign of adnexal cyst pedicle torsion by ultrasound has improved the diagnosis of acute symptoms[7]; in small studies this sign was observed in all cases of pedicle torsion in pregnancy13. The application of colour Doppler has not added much to the diagnosis made by direct ultrasound findings of torsion[7].

Due to the greater experience acquired in the correct identification of a twisted pedicle in an adnexal cyst by ultrasound and reports recommending management involving detorsion and preservation of the twisted adnexa for most premenopausal patients[11,19,22], the initially temporary, conservative treatment protocol was developed for pregnant patients with adnexal cyst pedicle torsion by puncture-aspiration under ultrasound guidance, avoiding exploratory laparotomy and its possible adverse consequences for the pregnancy.

This protocol was based on: the identification of the direct ultrasound sign of ovarian cyst pedicle torsion; the ultrasound characterization of this pathology as a simple adnexal cyst.(Table 3); the histopathological correlation of benign lesions in cases of simple cysts reported by ultrasound studies, in accordance with

what is reported in the international literature[3,5,15-18,20] and the multiple reports of conservative management involving detorsion of the twisted adnexa[13].

Size:	Less than 10 cm
Number of loculi	Unilocular
Content:	Anechogenic or low intensity fluid echoes
Wall thickness:	Fine (less than 3 mm)
Wall regularity:	Regular
Vascularization:	Absent or scarce high resistance (IR greater than 0.45)

TABLE 3.
Criteria for the classification of simple adnexal cyst. Hospital Obrero Nº 2, Caja Nacional de Salud.

The basis was the correct diagnosis of pedicle torsion and characterisation of the adnexal mass as a simple cyst, as the technical procedure is relatively simple and safe for the mother and the product.

In our experience, eight of the nine patients (89%) showed almost immediate symptomatic improvement within the first ten minutes; one patient (1/9) had an exacerbation reaction of the acute picture with sweating, chills and vomiting, which showed no relationship with the time of evolution of the clinical picture and subsided spontaneously in about 30 minutes. In one case (1/9) a miscarriage occurred on the fifth day of the procedure in a 7-week pregnancy that could be attributable to the condition itself. There was another pregnancy loss distant from the procedure and was therefore not associated with the torsional picture or the intervention.

The hypothesis used in this protocol was that by eliminating the main torsional condition, the size of the cyst, detorsion would occur and initiate vascular irrigation again which would improve the clinical picture and salvage the adnexa; in our study in all cases the compromised adnexa was salvaged.

Compared to a previous study in our department, the main benefit observed was the preservation of the compromised adnexa in all cases, and a significant reduction in hospitalisation time, with an average of 2.7 days compared to 8.2 days when laparotomy was performed[13](Table 4).

Event	Exploratory laparotomy* N: 5	Aspiration puncture N:9
Maternal mortality	0	0
Abortion	1	1
Adnexal loss	5	0
Days of hospitalization	8,2	2,7

**Data from a previous study[19].*

TABLE 4
Summary of the main differences between exploratory laparotomy and traditional treatment versus puncture aspiration under ultrasound guidance.

The results have been considered satisfactory so far, with no negative outcome, in terms of serious or permanent morbidity or mortality, for pregnant women. The treatment, which was initially considered temporary, has so far resolved the acute clinical picture in all cases, resolving the adnexal cyst in 78% of cases, with the remaining cases having smaller remaining cysts which in our cases were excised during caesarean section resulting in benign histological lesions.

Conclusion

Suspicion of the characteristic clinical picture is the first step towards diagnosis. Identification of the triad makes the diagnosis of pedicle torsion in just over 50% of patients.

The diagnostic approach, in addition to clinical examination, should include a thorough ultrasound examination by qualified personnel, seeking to identify the direct sign of torsion (visible twisted pedicle

"spiral" or whirlpool), if this sign is clearly identified, it correlates with almost all cases. Pregnancy would not appear to influence the identification of this direct sign.

Surgical management by laparotomy brings with it some complications such as loss of the involved adnexa, miscarriage, longer hospital stay, so that puncture-evacuation of the simple twisted cyst, which was initially proposed as a temporary treatment, appears to be a good alternative for definitive treatment in certain cases, however, it deserves to be better studied.

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