Case Report

MR"I": An "eye" for the preoperative diagnosis of ectopic molar pregnancy, a case report

Sujith Rajashekar Swamy[1], Meghana Rajashekara Swamy[2], Samanvitha H.[1], Sara Elsayed[2]

[1]Department of Radiodiagnosis, Bangalore Medical College and Research Institute, Bengaluru, Karnataka, India, [2]Department of Family Medicine, McLaren Flint – Michigan State University College of Human Medicine, U.S.A

Abstract

Hydatidiform mole (complete and partial), invasive mole, choriocarcinoma, placental site trophoblastic disease, and epithelioid trophoblastic tumour constitute the spectrum of benign and malignant gestational trophoblastic disease[1] Invasive mole, choriocarcinoma, placental site trophoblastic disease, and epithelioid trophoblastic tumour also classify under gestational trophoblastic neoplasia.[1] The prevalence of molar pregnancy shows great worldwide variation with reported rates of 12 per 1,000 pregnancies in Indonesia, India, and Turkey; one to two per 1,000 pregnancies in Japan and China; and 0.5 to one per 1,000 pregnancies in North America and Europe.[1] Ectopic pregnancy, which is primarily tubal, is the leading cause of first trimester maternal mortality.[2] Diagnosis of ectopic pregnancy is a combinatorial analysis of clinical signs and symptoms; beta-hCG trends; and ultrasonography.[2] Since ectopic gestations cause maternal deaths, the decisive role of the diagnostic test employed measured by its discriminative potential for a reliable preoperative diagnosis is paramount.[2] Although ultrasonography demonstrates high sensitivity and specificity in diagnosing ectopic gestations, inconsistencies in sonographic identification have been known to occur.[2] Particularly, ultrasonography suffers from limitations such as specifying the exact location of infrequent extrauterine presentations and identifying ectopic gestations with atypical features.[2] Molar pregnancies that are largely known to be placental in location have a known but rare potential for extrauterine proliferation.[3] Ectopic molar gestations are rare with only more than a hundred reported cases in scientific literature.[4] Our case delineates this uncommon entity and the superiority of magnetic resonance imaging in terms of diagnostic performance in characterizing the gestational mass over ultrasonography. This is pertinent considering the need to differentiate an ectopic molar pregnancy from an ectopic pregnancy without molar tissue because the potential for malignancy in the former atypical form is akin to that of an intrauterine molar pregnancy.[4]

Keywords: Complete hydatidiform mole, ectopic pregnancy, invasive mole, molar pregnancy

Introduction

Ectopic molar pregnancies are a rare occurrence.[4] Ultrasonography is the first imaging modality employed to evaluate first trimester bleeding in an emergency setting. Fowler *et al.*[5] reported that its detection rate for molar pregnancy is less than 50% which is compounded by the fact that the diagnosis is largely operator dependent. Although histopathology of the postoperative specimen is the gold standard for diagnosis, magnetic resonance imaging (MRI) is a promising tool which aids in the preoperative diagnosis of ectopic molar pregnancy.[6] MRI's superior resolution better delineates tumour margins, hemorrhage, and necrosis in gestational trophoblastic neoplasia.[7] Considering the reported rate of rupture of 67% for ectopic molar pregnancies, MRI in stable patients, by clarifying the ectopic mass preoperatively, supports shared decision-making in choosing medical management versus

Address for correspondence: Dr. Meghana Rajashekara Swamy, McLaren Flint Family Medicine Residency, G-3230 Beecher Road, Suite 1, Flint, MI - 48532, U.S.A.
E-mail: Meghana.rajashekara@gmail.com; swamymeg@msu.edu

Received: 06-09-2022 **Revised:** 21-11-2022
Accepted: 08-12-2022 **Published:** 17-03-2023

Access this article online

Quick Response Code:

Website: www.jfmpc.com

DOI: 10.4103/jfmpc.jfmpc_1770_22

This is an open access journal, and articles are distributed under the terms of the Creative Commons Attribution-NonCommercial-ShareAlike 4.0 License, which allows others to remix, tweak, and build upon the work non-commercially, as long as appropriate credit is given and the new creations are licensed under the identical terms.

For reprints contact: WKHLRPMedknow_reprints@wolterskluwer.com

How to cite this article: Swamy SR, Swamy MR, Samanvitha H, Elsayed S. MR"I": An "eye" for the preoperative diagnosis of ectopic molar pregnancy, a case report. J Family Med Prim Care 2023;12:581-5.

surgery.[6] Primary care physicians attending to the comprehensive healthcare needs of pregnant patients in multiple settings need to be informed of this entity and its management to augment practice potential. This case report expounds the utility of MRI to aid generalists in delivering high value care by supplementing transvaginal ultrasonography (TVUS) findings. Patient's consent for publication has been obtained.

Case History

A 24-year-old East Indian woman with an obstetric history of gravida two and para two presented to the emergency department with worsening hyperemesis and pelvic pain for one month. She also endorsed vaginal spotting for the past two days. Her last menstrual period was four weeks prior to presentation. Palpation elicited left-sided pelvic tenderness. She was hemodynamically stable. Her urine pregnancy test was positive. Serum beta hCG levels were found to be significantly elevated at 150,045 ng/mL. TVUS performed showed no evidence of an intrauterine pregnancy but it localized a left adnexal heterogeneous hyperechoic mass [Figure 1]. Colour Doppler revealed inconsistent uptake. Commonly, nonmolar ectopic pregnancies present as a simple or complex cystic adnexal mass with an echogenic rim and seldom as a heterogenous mass.[8] The peripheral hypervascularity can be recognized on Doppler examination as a "ring of fire".[8]

Considering that the patient was vitally stable, MRI was performed for further characterization of the mass. MRI revealed a seven-centimeter lesion with multicystic areas in the left adnexa which were hypointense on T1-weighted images (T1-WI) and hyperintense on T2-weighted images (T2-WI) [Figures 2 and 3]. Diffusion weighted imaging and gradient echo sequences are shown in Figures 4, 5 and 6. The mass enhanced heterogeneously with gadolinium contrast [Figure 7].

These findings raised concern for an ectopic gestation with atypical molar features. Surgery was consulted for emergency laparotomy. Salpingectomy was performed considering the high rupture risk.[9] There was no evidence of hemoperitoneum. The surgical specimen revealed no fetal parts and cut section was a mix of ruptured and intact grape-like vesicles covered in membranes as shown in Figure 8. Patient tolerated the procedure well.

Histopathology shown in Figure 9 of the salpingectomy specimen favored a complete ectopic hydatidiform mole. Genetic testing was not pursued as it was cost-prohibitive for the patient.

Postpartum, she was followed with scheduled clinic visits till her beta-hCG level was negative.

Discussion

Hydatidiform mole develops from cytotrophoblasts and syncytiotrophoblasts, with two major variants being complete and partial.[2] A complete mole expresses a diploid androgenetic

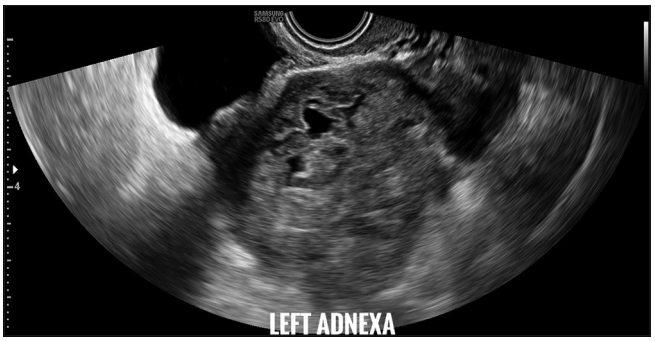

Figure 1: TVUS image of the left adnexa, original, showing an echogenic mass with cystic spaces

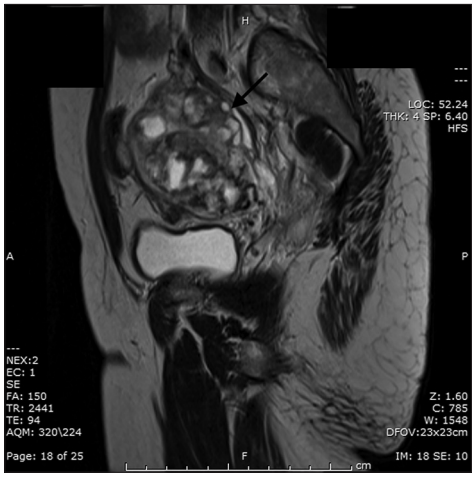

Figure 2: Sagittal T2-WI of the pelvis, original, showing a heterointense lesion in the left adnexa, with T1 hypointense and T2 hyperintense cystic areas. Arrow points to the left ovary superolateral to the lesion

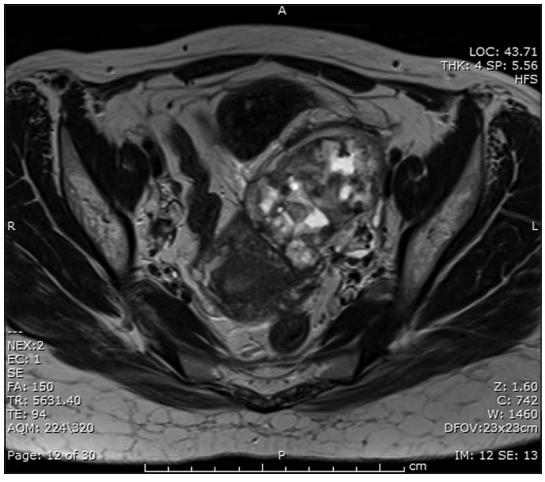

Figure 3: Axial T2-WI of the pelvis, original, showing a heterointense lesion in the left adnexa

karyotype, and hence consists purely of molar tissue while a partial mole demonstrates a triploid karyotype and is, therefore, a mix of molar-fetal tissue and placental villi.[10]

Molar pregnancies are relatively common when compared to only 132 documented cases of ectopic molar pregnancy.[4] Discerning

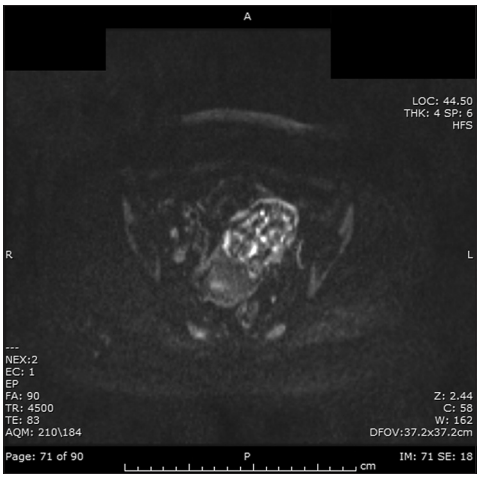

Figure 4: DWI image, original, of the lesion showing patchy areas of diffusion restriction

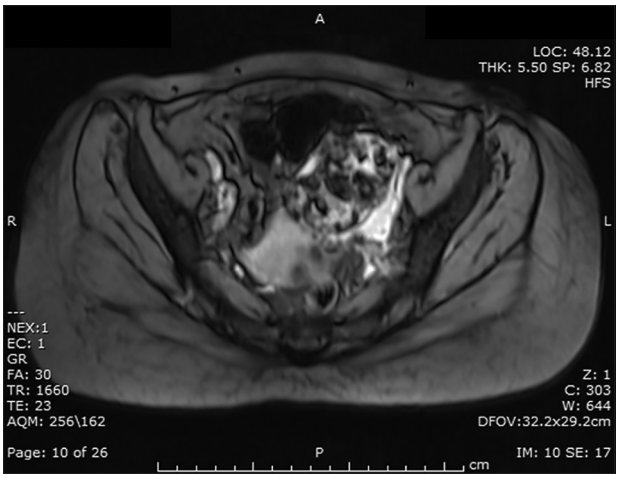

Figure 5: GRE image, original, showing blooming foci within the lesion suggestive of intralesional hemorrhage

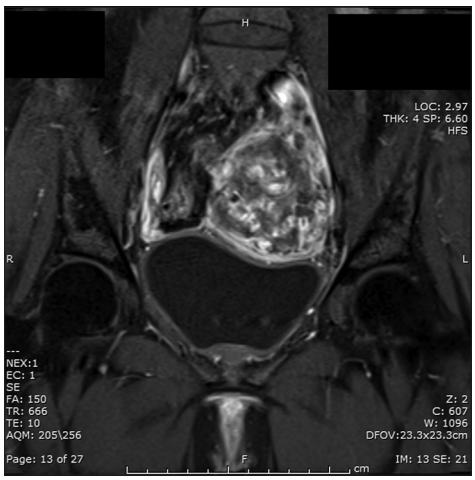

Figure 6: Postcontrast coronal T1-FS image, original, showing heterogenous contrast uptake by the left adnexal lesion

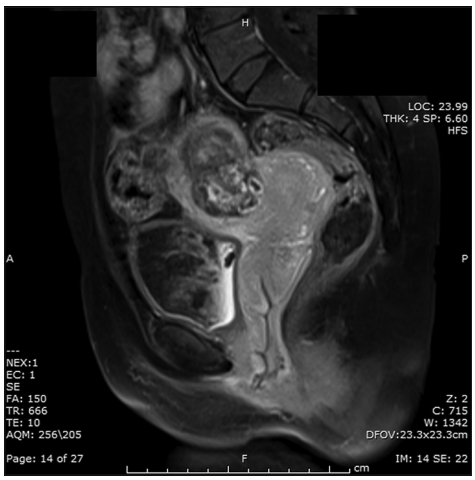

Figure 7: Postcontrast sagittal T1-FS image, original, showing heterogenous contrast uptake by the left adnexal lesion

ectopic molar from nonmolar ectopic gestations is essential due to perilous complications such as invasive mole, persistent trophoblastic illness, or choriocarcinoma that could ensue.[4,6,11] Table 1 summarizes the utility of hormonal assays in both such presentations. As histopathology is the confirmative diagnostic test, surgery is preordained when the condition is suspected.[6,12] However, preoperative diagnosis with reliable characterization of the ectopic mass identified by TVUS is relevant for stable patients with pure ectopic gestations in whom medical management alone offers satisfactory treatment.[6,12]

From our review of literature, we found only two cases where a competent preoperative diagnosis of ectopic molar gestation was made by diagnostic imaging.[12] Yamada Y *et al*.[6] characterized an ectopic molar pregnancy in the right uterine cornu via MRI. Asseryanis *et al*.[9] preoperatively identified a left tubal invasive molar pregnancy using colour Doppler imaging which revealed arteriovenous shunting between the tumour and myometrium. However, the efficacy of Doppler imaging for diagnosing ectopic molar pregnancy remains controversial due to the possibility of inhibited vascularization related to fallopian tube implantation.[6]

MRI by reason of its sterling soft tissue resolution can deliver a reliable high-stakes diagnosis when ultrasonographic findings are equivocal for adnexal masses.[13] A comparison of ultrasound and MRI as modalities for the characterization of adnexal masses is provided in Table 2.

In our patient, heterogeneously enhancing cystic lesions identified by MRI represented molar tissue. In stable patients, MRI can clarify the preoperative diagnosis considering the risks of emergency laparotomy, when the patient could have otherwise benefited from methotrexate therapy.[2,6,18] The invasive nature of trophoblasts in gestational trophoblastic diseases predispose ectopic molar gestations to a higher rupture rate and hemoperitoneum in comparison to ordinary ectopic pregnancies.[6,18]

Family physicians' scope of practice is known to include maternity care, which can range from low-risk gestations to

Table 1: Comparison of the clinical relevance of hormonal analysis between molar and non-molar ectopic pregnancies

Clinical entity	Hormonal analysis	Clinical relevance
Nonmolar ectopic pregnancy	Urine or serum beta-hCG are confirmatory for a pregnant state.[2] Serum beta-hCG helps to quantify hormonal levels with consensus that intrauterine pregnancies are identified by TVUS at 1500 UI/mL.[2]	Drug therapy with methotrexate is favored if serum beta-hCG is <5000 IU and the size of the ectopic mass is <3 cm.[2] The drug is delivered intramuscularly for a targeted decline of serum beta-hCG of 15% in 4-7 days.[2]
Molar ectopic pregnancy	Serum beta-hCG levels help to confirm the diagnosis and organize follow up following molar pregnancy termination.[1] Higher beta-hCG levels have been recorded in molar ectopic gestations when compared to regular ectopic pregnancies, with complete moles being higher than partial moles.[6]	Beta-hCG levels alone are insufficient to differentiate molar and non-molar ectopic pregnancies.[6] Trending beta-hCG levels for decrease post pregnancy termination can allow for early detection of GTNs.[1] This can prevent progression to advanced malignancy with timely chemotherapy.[1]

Table 2: Comparison of ultrasonography and MRI for the characterization of ectopic hydatidiform molar gestation

Diagnostic features	Ultrasonography	MRI
Image characteristics	A heterogeneous echogenic mass in the adnexa with cystic spaces, showing a characteristic "snowstorm" or "cluster of grapes" appearance.[14]	Isointense lesion on T1-WI and hyperintense lesion on T2-WI which are suggestive of hydropic villi.[6,14]
Vascularity	Colour flow imaging shows intense colour uptake with high velocity low resistance waveforms and chaotic vasculature of the tumoral arteriovenous shunt.[14]	Heterogeneous enhancement will be seen in contrast studies.[15] Numerous signal voids can be seen within the adnexa and myometrium, representing dilated vessels of the intratumoral arteriovenous shunts and tumor neovascularity when present.[14,16]
Features of local invasion	Features of local invasion include a large pelvic mass with lobulated contour extending into other pelvic organs.[16] Other features include asymmetrical myometrial thickening or an indistinct endometrial-myometrial junction.[16]	Myometrial invasion is seen as diffuse myometrial signal hyperintensity with obscuration of zonal anatomy.[16] It is superior to ultrasound in characterizing parametrial invasion seen as heterogenous on T2.[16]
Detection of hemoperitoneum	Rupture at the time of presentation results in hemoperitoneum, which can be seen as hypoechoic fluid within the peritoneal cavity.[10] There are frequently multiple areas of increased echogenicity within the hypoechoic fluid.[10]	Signal characteristics of the intraperitoneal blood may vary considerably and depends on the age of the blood products.[10] By the time most patients have an MRI performed, the blood will have a high signal intensity on T1-WI and intermediate signal intensity on T2-WI.[10]
Strengths	Better to evaluate unstable patient.[17] Helps identify hemoperitoneum easily.[17] Short scan time.[17]	Helps characterize the lesion better.[17] Helps characterize lesion extensions better.[17] Better soft tissue resolution.[17] Not operator dependent.[17]
Limitations	Operator-dependent.[17] Less specific.[17]	Longer scan time.[17] Gadolinium contrast can be used provided a viable intrauterine pregnancy has been excluded.[15]

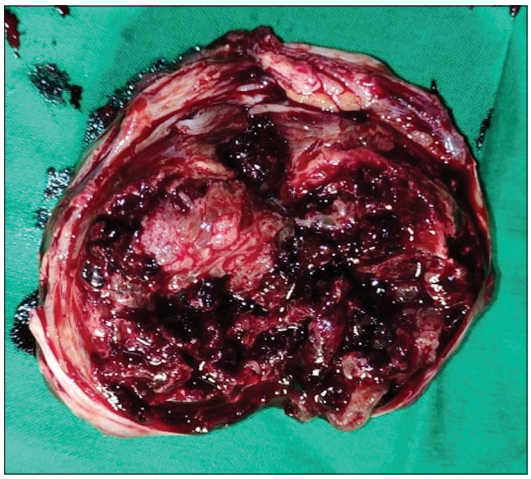

Figure 8: Cut section, original, of the surgical specimen showing edematous hemorrhagic tissue with no evidence of fetal parts

pregnancies with high-risk features.[19] Generalists are usually the first point of contact for pregnant women and often care for patients in emergency settings.[19] Knowledge of the utility of MRI as a powerful diagnostic support tool can assist providers in such settings in clinical decision-making for hemodynamically stable ectopic molar gestations. Bearing in mind that an average MRI is expensive and costs time, we believe that it is best suited for stable patients in whom a dependable preoperative diagnosis can save costs and avoid morbidity related to surgical management.[20] A preoperative diagnosis also allows generalists to educate patients on the possibility of identifying gestational trophoblastic neoplasia in the surgical specimen which warrants further surveillance.[13,21]

Nonetheless, considering the scarcity of literature on diagnosing ectopic molar gestations, we recommend for future directions, more investigation to advance dialogue on optimal diagnostic and management approaches.

Abbreviations

All definitions have been expanded in text.

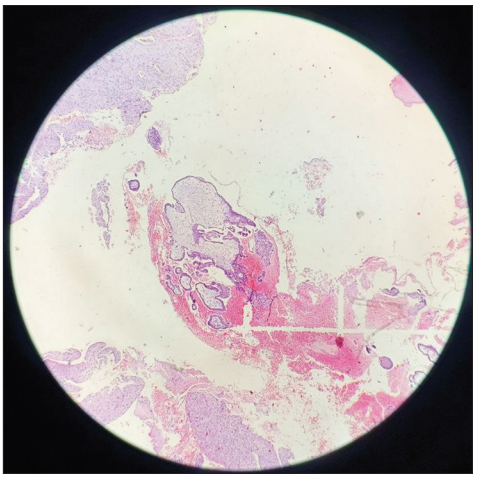

Figure 9: Histopathological image, original: 4 X magnification, H and E–stained section of the salpingectomy specimen showing chorionic villi without fetal blood vessels, proliferating syncytiotrophoblasts and edematous stroma

Patient declaration of consent

Obtained.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form the patient(s) has/have given his/her/their consent for his/her/their images and other clinical information to be reported in the journal. The patients understand that their names and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

Reporting guidelines

The article adheres to the CARE reporting guidelines for case reports.

Financial support and sponsorship

Nil.

Conflicts of interest

There are no conflicts of interest.

References

1. Steigrad SJ. Epidemiology of gestational trophoblastic diseases. Best Pract Res Clin Obstet Gynaecol 2003;17:837-47.
2. Stucki D, Buss J. The ectopic pregnancy, a diagnostic and therapeutic challenge. J Med Life 2008;1:40-8.
3. Ghassemzadeh S, Farci F, Kang M. Hydatidiform Mole. StatPearls. Treasure Island (FL): StatPearls Publishing; 2022.
4. Beena D, Teerthanath S, Jose V, Shetty J. Molar pregnancy presents as tubal ectopic pregnancy: A rare case report. J Clin Diagn Res 2016;10:ED10-1.
5. Fowler DJ, Lindsay I, Seckl MJ, Sebire NJ. Routine pre-evacuation ultrasound diagnosis of hydatidiform mole: Experience of more than 1000 cases from a regional referral center. Ultrasound Obstet Gynecol 2005;27:56-60.
6. Yamada Y, Ohira S, Yamazaki T, Shiozawa T. Ectopic molar pregnancy: Diagnostic efficacy of magnetic resonance imaging and review of the literature. Case Rep Obstet Gynecol 2016;2016:7618631.
7. Ha HK, Jung JK, Jee MK, Kim HH, Lee JM, Nam-Koong SE, *et al.* Gestational trophoblastic tumors of the uterus: MR imaging--pathologic correlation. Gynecol Oncol 1995;57:340-50.
8. Jones J, Gaillard F. Ectopic pregnancy. Radiopaediaorg 2008;rID 1258:. Available from: https://radiopaedia.org/articles/ectopic-pregnancy?lang=us.
9. Asseryanis E, Schurz B, Eppel W, Wenzl R, Vavra N, Husslein P. Detection of an atypical invasive mole in an ectopic pregnancy by transvaginal color-flow Doppler. Am J Obstet Gynecol 1993;169:1656.
10. Lurain JR. Gestational trophoblastic disease I: Epidemiology, pathology, clinical presentation and diagnosis of gestational trophoblastic disease, and management of hydatidiform mole. Am J Obstet Gynecol 2010;203:531-9.
11. Chauhan S, Diamond MP, Johns DA. A case of molar ectopic pregnancy. Fertil Steril 2004;81:1140-1.
12. Mbarki C, Jerbi E, Hsayaoui N, Zouari F, Ben Brahim E, Oueslati H. Molar tubal ectopic pregnancy: Report of two cases. J Obstet Gynaecol Res 2014;41:985-8.
13. Sohaib SA, Mills TD, Sahdev A, Webb JA, Vantrappen PO, Jacobs IJ, *et al.* The role of magnetic resonance imaging and ultrasound in patients with adnexal masses. Clin Radiol 2005;60:340-8.
14. Shaaban AM, Rezvani M, Haroun RR, Kennedy AM, Elsayes KM, Olpin JD, *et al.* Gestational trophoblastic disease: Clinical and imaging features. RadioGraphics 2017;37:681-700.
15. Garcia-Bournissen F, Shrim A, Koren G. Safety of gadolinium during pregnancy. Can Fam Physician 2006;52:309-10.
16. Dhanda S, Ramani S, Thakur M. Gestational trophoblastic disease: A multimodality imaging approach with impact on diagnosis and management. Radiol Res Pract 2014;2014:842751. doi: 10.1155/2014/842751.
17. Varwatte P, Ilangovan G, Balaganesan H, A P. Comparative study of ultrasonography and magnetic resonance imaging in the diagnosis of adnexal lesions. Int J Contemp Med Surg Radiol 2020;5:B38-43.
18. Hwang JH, Lee JK, Lee NW, Lee KW. Molar ectopic pregnancy in the uterine cornus. J Minim Invasive Gynecol 2010;17:239-41.
19. Dera JD, Marso A, Hughes C, Moore KJ, editors. Maternal/Child Care (Obstetrics/Perinatal Care). American Academy of Family Physicians; 2019. Available from: https://www.aafp.org/about/policies/all/maternal-child-care.html. [Last accessed on 2019 Dec 12].
20. van Beek EJR, Kuhl C, Anzai Y, Desmond P, Ehman RL, Gong Q, *et al.* Value of MRI in medicine: More than just another test? J Magn Reson Imaging 2019;49:e14-25.
21. Seckl MJ, Sebire NJ, Berkowitz RS. Gestational trophoblastic disease. Lancet 2010;376:717-29.