Molar Pregnancy - Associated Ectopic Decidua : Report of a Case and Review of the Literature

CHURAIRAT KULARBKAEW, M.D.*,
WIBOONCHAI YUTANAWIBOONCHAI, M.D.*,
CHAWALIT PAIROJKUL, M.D.*

Abstract

Ectopic or extrauterine decidual transformation is regularly associated with pregnancy. This phenomenon has been observed most often in the ovary and cervix. Sporadic reports have documented decidua formation of peritoneal surfaces, lymph nodes and other organs in pelvic and abdominal cavity. Molar pregnancy-associated ectopic decidua has never been documented. We report the case of ectopic decidua at posterior surface of uterus, near the cul de sac, in a 45-year-old woman with molar pregnancy. The article review reveals that it is the first report of molar pregnancy-associated ectopic decidua.

The ectopic decidua was first described by Walker in 1887[1] and ten years later by Schmorl[2]. In general, the normal decidua consists of the stromal cells of the endometrium transformed during pregnancy as a result of the influence of ovarian and placental hormones, especially of progesterone[3]. Ectopic or extrauterine decidual transformation is regularly associated with pregnancy and has been induced occasionally in non-pregnant and postmenopausal woman by progesterone or progesterone-like substances from an active corpus luteum, or from adrenal cortex[4]. However, molar pregnancy-associated ectopic decidua has never been reported. Such a situation occurred in one of our patients.

CASE REPORT

A 45-year-old woman, gravida 3, para 1, abortion 1 presented at the antenatal clinic at 10 weeks gestation with a 3 days history of vaginal spotting. Pelvic examination reveled an enlarged uterus about 20 weeks gestation. On investigation by ultrasonogram revealed snow storm appearance and serum level of beta-human chorionic gonadotropin (BhCG) was more than 80,000 mIU/ml. A clinical diagnosis of hydatidiform mole was made.

* Department of Pathology, Khon Kaen University, Khon Kaen 40002, Thailand.

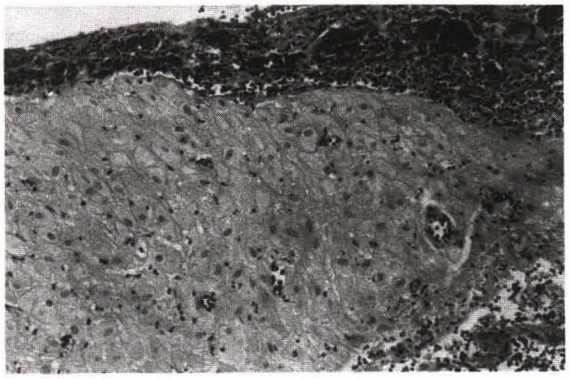

Fig. 1. Nest of ectopic decidual cells.

Total abdominal hysterectomy with bilateral salpingo-oophorectomy were subsequently performed.

Pathologic examination revealed invasive mole at uterine corpus with pregnancy luteoma at right ovary and ectopic decidua at posterior surface of uterus near cul de sac (uterosacral ligament).

The patient's serum level of beta-human chorionic gonadotropin (BhCG) decreased rapidly after surgery and was below 10 mIU/ml after 4 courses of chemotherapy.

Microscopic investigations : The decidual cells were visible on the serosal surface of the uterus. Nests of large, round, ovoid or polyhedral cells with sharply outlined cell borders, slightly basophilic granular or slightly foamy cytoplasm and round, slightly eccentric nuclei with single prominent nucleoli were present (Fig. 1).

Immunohistochemical findings : For immunohistochemical studies, formalin-fixed, paraffin-embedded tissue sections were stained with antibodies against vimentin, desmin, keratin AE-1, AE-3, and epithelial membrane antigen (Dako; Copenhagen, Denmark).

The decidual cells stained strongly positive with vimentin antibodies and also showed strong focal positivity with desmin. Stains for keratin and EMA were negative in the decidual cells.

DISCUSSION

By immunohistochemistry, the decidual cells reacted strongly with vimentin antibodies and were negative for keratin, CEA and EMA as well as for neural and/or neuroendocrine markers[5]. The demonstration of focal strong reactivity of the decidual cells with antibodies to muscle-specific actin and desmin both are cytoskeletal filaments, are generally accepted as markers of muscle differentiation[5-7]. Although the normal decidual cells result from the transformation of endometrial stroma cells or subserosal peritoneal stromal cells in response to hormonal stimuli[8,9], recent evidence would seem to suggest that such cells may also respond to the same hormonal stimuli by undergoing smooth muscle differentiation[5]. In his classic study of decidua formation in the primitive subcoelomic stromal cells, Hofbauer found that the cells, may, under the appropriate stimuli, differentiate toward either decidua or smooth muscle cells[10]. The possibility therefore exists that this capacity for multiple differentiation is retained in certain subsets of decidual cells where it is manifested by the expression within those cells of biochemical markers of smooth muscle differentiation.

In patients with molar pregnancy, it is possible that ectopic decidua occurs as a result of the influence of progesterone from ovarian and placental hormones. Seki studies relaxin, progesterone and hCG levels by using radioimmunoassay in patients with hydatidiform mole before and after evacuation of the mole and found that serum immunoreactive relaxin and progesterone levels in patients with hydatidiform mole were similar to those in normal women at corresponding weeks of pregnancy before evacuation of the mole, though hCG levels were significantly higher[11].

Table 1 shows the review of reported cases of ectopic decidua. Almost all publications are in intrauterine fetal pregnancy-related except 25 cases reported by Bassis[12] and 16 cases reported by Ober[13] showing ectopic decidua at the ovary in the absence of pregnancy. Walker[1] reported ectopic decidua at pelvic peritoneum in association with an extra-uterine pregnancy. Taussig reported ectopic decidua at parovarian cyst, both tubes and ovaries, peritoneum and cervix in association with tubal pregnancy[14]. After reviewing all reported cases of ectopic decidua we find that the most common site is ovary (82 cases)[12-16], followed by cervix (71 cases)[17-23] and omentum (51 cases) [3,8,24,25]. Sporadic reports have documented decidua formation at anterior and posterior uterine surfacee[10], para-aortic and pelvic lymph nodes

Table 1. Review of reported cases of ectopic decidua.

Author	year	Number of cases	Sites of decidua reaction	Associated pregnancy
Walker	1887	1	pelvic peritoneum	extrauterine
Hirschberg	1905	1	appendix	intrauterine
Taussig	1906	1	parovarian cyst, both tubes and ovaries, peritoneum and cervix	extrauterine (tubal pregnancy)
Hofbauer	1929	15	posterior aspect of the uterus	intrauterine
Konwer	1929	1	scattered throughout pelvis with bleeding	intrauterine
Reis	1940	1	appendix	intrauterine
Hennessy	1943	1	cervix	intrauterine
Russel	1945	1	pelvic lymph node	intrauterine
Klein	1946	1	cervix	intrauterine
Bettinger	1947	1	renal pelvis	intrauterine
Lapan	1949	3	cervix and vagina simulating carcinoma	intrauterine
Melody	1950	1	omentum with bleeding	intrauterine
Israel	1954	19	ovary	intrauterine
Bassis	1956	25	ovary and uterine tube	no pregnancy
Doyle	1957	1	lateral pelvic wall with bleeding	intrauterine
Mathie	1957	1	vagina simulating carcinoma	intrauterine
Ober	1957	16	ovary	no pregnancy
O'Sullivan	1960	2	peritoneum	intrauterine
Orr	1961	1	cervix, grossly simulating carcinoma with antepartum hemorrhage	intrauterine
Armenia	1964	2	cervix, simulating reticulum cell sarcoma	intrauterine
Kwan	1964	1	anterior surface of uterus, omentum and broad ligament	intrauterine
Roger	1965	1	left broad ligament with bleeding	intrauterine
Winkelstein	1967	1	endocervix, deep myometrium, parametrium and several obturator lymph nodes with associated squamous cell carcinoma of the cervix	intrauterine
Hulme-Moir	1969	1	omentum with bleeding	intrauterine
Rewell	1971	15	fallopian tubes	intrauterine
Sabautelle	1973	1	posterior surface of uterus with bleeding	intrauterine
Covell	1977	1	pelvic lymph nodes with associated squamous cell carcinoma of the cervix	intrauterine
Herr	1978	21	ovary	intrauterine
Schneider	1981	62	uterine cervix	intrauterine
Yoonesei	1982	1	periurethral and external iliac nodes with associated squamous cell carcinoma of the cervix	intrauterine
Mills	1983	1	with squamous metaplasia in abdominopelvic lymph nodes	intrauterine
Richter	1983	1	posterior surface of uterus and broad ligaments with bleeding	intrauterine
Ashfar	1984	1	para-aortic and pelvic lymph nodes with associated squamous cell carcinoma of the cervix	intrauterine
Burnett	1986	1	single lymph node of the right internal iliac group with associated infiltrating adenocarcinoma of the cervix	intrauterine
Zaytsev	1987	10	peri-aortic lymph node, round ligament, serosa surface of appendix, omentum, right peritubal cyst, cyst of Morgagni	intrauterine
Cobb	1988	1	present with metastatic squamous cell carcinoma of the cervix in a single pelvic lymph node	intrauterine
Suster	1990	6	appendix	intrauterine
Buttner	1993	48	omentum	intrauterine
Kularbkaew	present report	1	posterior surface of uterus near cul de sac	Molar pregnancy

(22,26-32,45), peritoneum(25,33), diaphragm, liver, spleen(34), fallopian tubes(26,35), renal pelvis(36), appendix(5,37,43) and vaginal wall(44).

Although usually present as asymptomatic and incidental microscopic finding in surgically removeal tissue, ectopic decidua may rarely be the cause of abdominal symptoms, in particular, pain and fatal intraperitoneal hemorrhage during the third trimester of pregnancy, labor and postpartum. Seven cases were previously reported that severe ectopic decidual reaction of the peritoneal surface was the etiology of unexpected intraabdominal hemorrhage(8,24,38-42). The patients presented with peripartum shock and/or an acute abdomen. Suster reported four cases of patients who presented with clinical signs and symptoms of acute appendicitis during pregnancy(5). Rare cases have been reported as the cause of symptoms of hydronephrosis and hematuria secondary to renal pelvis involvement(36).

To our knowledge, this is the first case reported of ectopic decidua in molar pregnancy.

(Received for publication on April 28, 1997)

REFERENCES

1. Walker A. Der bau eihaute bei graviditatic abdominalis. Virchows Arch Path Anat 1887;197:72-99.
2. Schmorl G. Uber grofszellige (deziduaahnliche) wucherungen auf dem. Peritoneum und den ovarien bei intrauteriner schwangerschaft. Mschr Geburtsh Gynecol 1897;5:46-50.
3. Buttner A, Bassler R, Theele C. Pregnancy-associated ectopic decidua (deciduosis) of the greater omentum. An analysis of 60 biopsies with cases of fibrosing deciduosis and leiomyomatosis peritonealis disseminata. Path Res Pract 1993;189: 352-9.
4. Pieslor PC, Orenstein JM, Hogan DL. Ultrastructure of myofibroblasts and decidualized cells in leiomyomatosis peritonealis disseminata. Am J Clin Pathol 1979;72:875-82.
5. Suster S, Moran CA. Deciduosis of the appendix. Am J Gastroenterol 1990;85:841-5.
6. Gabbiani G, Kappanci Y, Brazzone P, Franke WW. Immunochemical identification of intermediate sized filaments in human neoplastic cells : A diagnostic aid for the surgical pathologist. Am J Pathol 1981;104:206-14.
7. Skalli O, Gabbiani G, Babai F, Seemayer TA, Pizzolato G, Schurch W. Intermediate filament proteins and actin isoforms as markers of soft tissue tumor differentiation and origin. II. Rhabdomyosarcoma. Am J Pathol 1988;130:515-31.
8. Melody GF. Deciduation and massive hemorrhage of the omentum in the final month of pregnancy. West J Surg Obstet Gynecol 1950;58: 460-2.
9. Weller CV. The ectopic decidual reaction and its significance in endometriosis. Am J Pathol 1935; 11:287-90.
10. Hofbauer J. Decidual formation on the posterior surface of the gravid uterus. Am J Obstet Gynecol 1929;17:603-12.
11. Seki K, Uesato T, Tabei T, Kato K. Serum relaxin in patients with hydatidiform mole. Obstet Gynecol 1986;67:381-3.
12. Bassis ML. Pseudodeciduosis. Am J Obstet Gynecol 1956;72:1029-37.
13. Ober WB, Grady HG, Schoenbucher AK. Ectopic ovarian decidua without pregnancy. Am J Pathol 1957;33:199-217.
14. Taussig FJ. Ectopic decidua formation. Surg Gynec Obstet 1906;2:292-303.
15. Israel L. The ovary at term : decidua-like reaction and surface cell proliferation. Obstet Gynecol 1954;3:399-407.
16. Herr JC, Heideger PM Jr, Scott JR. Decidual cells in the human ovary at term. Incidence, gross anatomy and ultrastructural features of merocrine secretion. Am J Anat 1978;152:7-27.
17. Hennessy JP. Unusual decidual reaction of the cervix. Am J Obstet Gynecol 1943;46:570-2.
18. Klein J, Domeier LH. An unusual decidual reaction in the cervix. Am J Obstet Gynecol 1946;51: 423-6.
19. Lapan B. Decidussis of the cervix and vagina simulating carcinoma. Am J Obstet Gynecol 1949; 58:743-7.
20. Orr CJB, Pedlow PRB. Deciduosis of the cervix manifesting as antepartum hemorrhage and simulating reticulum cell sarcoma. Am J Obster Gynecol 1961;89:808-16.
21. Armenia C. Decidual transformation of the cervical stroma stimulation reticulum cell sarcoma. Am J Obstet Gynecol 1964;89:808-16.
22. Winkelstein CE. Decidua in pelvic lymph nodes. Obstet Gynecol 1967;29:824-7.
23. Schneider V. Ectpic decidual reaction of the

uterine cervix. Acta Cytolog 1981;25:616-21.
24. Hulme-Moir I, Ross MS. A case of early post-partum abdominal pain due to hemorrhagic deciduosis peritonei. J Obstet Gynecol Br Cwlth 1969; 76:746-9.
25. Kwan D, Pang LS. Deciduosis peritonei. J Obstet Gynecol Br Cwlth 1964;71:804-6.
26. Zaytsev P, Taxy JB. Pregnancy-associated ectopic decidua. Am J Surg Pathol 1987;11:526-30.
27. Cobb CJ. Ectopic decidua and metastatic squamous carcinoma presenting in a single pelvic lymph node. J Surg Oncol 1988;38:126-9.
28. Ashfar M. Ectopic decidual cell reaction in para-aortic and pelvic lymphnodes in the presence of cervical squamous cell carcinoma during pregnancy. J Surg Oncol 1984;26:6-8.
29. Burnett RA, Millan D. Decidual change in pelvic. lymph nodes : a source of possible diagnostic error. Histopath 1986;10:1089-92.
30. Covell L. Decidual change in pelvic lymph nodes in the presence of cervical squamous cell carcinoma during pregnancy. Am J Obstet Gynecol 1977;127:674-6.
31. Mills SE. Decidua and squamous metaplasia in abdominopelvic lymph nodes. Int J Gynecol Pathol 1983;2:209-15.
32. Yoonessi M. Benign glandular elements and decidual reaction in retroperitoneal lymph nodes. J Surg Oncol 1982;19:81-6.
33. O' sullivan D, Heflerman HK. Deciduosis peritonei in pregnancy. Report of two cases. Br J Obstet Gynecol 1960;67:1013-6.
34. Geipel P. Weiterer beitrag zur kenntnis des decidualen gewebes. Arch Gynak 1927;131:650-700.
35. Rewell RE. Extrauterine decidua. J Pathol 1971; 105:219-22.
36. Bettinger HF. Ectopic decidua in the renal pelvis. J Pathol Bacteriol 1947;59:686-7.
37. Hirschberg A. Deciduale zellbildungen am wurmfortsatz bei tubenschwangershaft (periappendicitis decidualis). Arch Gynak 1905;74:620-32.
38. Konwer BJ. Hemorragie lethale intra-abdominale a la fin de la grossesse causee par excroissance deciduales sur la paroi posterieure de la matrice. Gynecol Obstet 1929;19:34-7
39. Doyle GE, Phillips DL. Fatal intraperitoneal hemorrhage during pregnancy. J Obstet Gynecol Br Cwlth 1957;64:270-1.
40. Roger WS, Seckinger DL. Decidual tissue as a cause of intraabdominal hemorrhage during labor. J Obstet Gynecol 1965;25:391-7.
41. Sabautelle R, Winger E. Postpartum intraabdominal hemorrhage caused by ectopic deciduosis. Obstet Gynecol 1973;41:873-5.
42. Richter MA, Choudhry A, Barton JJ, Merrick RE. Bleeding ectopic decidua as a cause of intraabdominal hemorrhage : A case report. J Reprod Med 1983;28:430-2.
43. Reis RA, Sinykin MB. Ectopic ducidua in the vermiform appendix. Am J Obstet Gynecol 1940; 39:870-2.
44. Mathie JG. Vaginal deciduosis simulating carcinoma. J Obstet Gynacol Brit Emp 1957;64:720-3.
45. Russel HB. Decidual reaction of endometrium ectopic in an abdominal lymph node. Surg Gynec Obstet 1945;81:218-20.

การเกิดเดซิดวลนอกมดลูกในผู้ป่วยตั้งครรภ์ไข่ปลาอุก : รายงานผู้ป่วย 1 รายและทบทวนวารสาร

จุไรรัตน์ กุหลาบแก้ว, พ.บ.,*
วิบูลย์ชัย ยุทธนวิบูลย์ชัย, พ.บ., ชวลิต ไพโรจน์กุล, พ.บ.**

การเกิดเดซิดวลนอกมดลูกมักเกิดร่วมกับการตั้งครรภ์ จะพบบ่อยที่รังไข่และปากมดลูก นอกจากนั้นมีรายงานว่าพบที่ผนังช่องท้อง ต่อมน้ำเหลืองและอวัยวะต่างๆในช่องท้องและอุ้งเชิงกราน แต่ยังไม่เคยพบร่วมกับการตั้งครรภ์ไข่ปลาอุก ผู้รายงานได้รายงานผู้ป่วยหญิง 1 ราย อายุ 45 ปี ที่ตั้งครรภ์ไข่ปลาอุกและพบเดซิดวลนอกมดลูกที่ผนังด้านหลังของมดลูกบริเวณใกล้ cul de sac

* ภาควิชาพยาธิวิทยา, คณะแพทยศาสตร์ มหาวิทยาลัยขอนแก่น, ขอนแก่น 40002