TORSION OF LUTEIN CYST ASSOCIATED WITH HYDATIDIFORM MOLE

by

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The association of lutein cysts of the ovaries with hydatidiform mole is fairly common. They may be present in a far higher proportion than reported, because the reported frequency appears to depend upon enlargement of sufficient extent as to permit detection on physical exmination. Stroup reported an incidence of 16%, Eastman 25-60%, Runge & Cottalorda 16% and 59% respectively. Hobson reported lutein cysts in 23% of 176 hydatidiform moles. Chun *et al* gave an incidence of 29% from a large series of cases in Malaya.

However, the appearance of lutein cysts subsequent to evacuation of uterus is a far less common feature of hydatidiform mole, though such cases have been reported by several observers including Joseph, Rabau, Herold, Greenhill and Quigley. The complication of torsion of the pedicle in such a cyst seems to be still rarer. Hence the following case report may be of interest.

Case Report

L, aged 20 years, para 1+0, was admitted in May, 1962 with a history of three months' amenorrhoea, severe nausea, vomiting and headache of two days duration. On examination, the patient's general condition was fair, haemoglobin was 11.5 gm and blood pressure was 130/90 m.m. of Hg. Uterine fundus was felt at the level of the umbilicus. No foetal parts or foetal heart sounds were discernible. Within 24 hours of admission, the patient developed oedema of face, feet and abdominal wall and the height of the uterine fundus rose by another inch. The blood pressure rose to 150/106 m.m. of Hg. and albuminuria also appeared. There was no vaginal bleeding. Plain x-ray of abdomen showed no evidence of foetal skeleton. No biological tests for chorionic gonadotropins could be undertaken because of lack of laboratory facilities. From the history and findings, the diagnosis of hydatidiform mole was clear and the uterus was evacuated per vaginam under a pitocin drip (20 units in 1 pint of 5% dextrose). Bleeding was not excessive and only one pint of blood was transfused. Tne patient's general condition improved soon after evacuation of mole. Nausea and vomiting stopped, the oedema disappeared and urine became clear of albumin within 36 hours. She was discharged home on the 4th day of evacuation at her own request. Vaginal examination before discharge showed the uterus to be about 10 weeks in size. Both ovaries were enlarged to the size of a duck's egg.

One week after discharge, the patient was readmitted in the early hours of the morning as an emergency. She complained of an acute attack of severe pain in the right lower abdomen, which started about 6 hours prior to admission. On examination the patient looked ill and in severe pain. Pulse was 100/minute and the blood pressure was 120/80 m.m. of Hg. Vaginal examination showed that the uterus had involuted normally. There was a mass about the size of a big sweet lime on the right

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side of the abdomen arising from the right fornix. It was partly mobile, tender and dull on percussion. A diagnosis of twisted lutein cyst was made and a laparotomy was performed within two hours of hospitalization. A right ovarian cyst, 5 x 4 x 2 inches, was found with the pedicle undergoing torsion three times in the anti-clockwise direction. The cyst was haemorrhagic and so was removed with the twisted pedicle. The other ovary was the size of a duck's egg still and was conserved. The patient made an uneventful recovery and was discharged home on the 10th day. A curettage done on the 6th postoperative day, produced only fragments of endometrium showing involution of decidual reaction.

Discussion

Ovarian changes associated with hydatidiform mole have focussed the attention on chorionic gonadotropin as its cause. The luteal elements in the ovary are stimulated by this hormone. The fluid in these cysts is said to store chorionic gonadotropin and this may be the explanation for the persistence of positive pregnancy tests long after complete evacuation of the molar tissue. There is marked polycystic enlargement with exaggerated luteinization of theca cells. The enlargement may be slight, moderate or large. The time of appearance of these cysts seems to be variable. Though in most cases they regress after complete evacuation of the mole, at times they appear for the first time after the mole has been evacuated. Several cases are on record where the cysts became much larger 1 to 5 weeks after evacuation of the molar tissues (Klein). According to Israel it is presumed that delay in appearance of lutein cysts may be attributed to the retention of some trophoblastic tissue within the uterus. This residual tissue is sufficiently viable to continue production of chorionic gonadotropin. Apparently there is no correlation between the size of the cysts and that of the mole. Novak cites the instance of Penkert's case in which a small microscopic area of hydatidiform disease in an otherwise normal placenta was associated with lutein cysts as large as a child's head. There are some who believe that it is really the expulsion of the mole which furnishes the impetus to the growth of the lutein cysts. This, however, may not be so because with the uterus abnormally large it is difficult to palpate small lutein cysts, and when the uterus is emptied the cysts become evident to the palpating hand. The cyst may, however, start increasing in size after evacuation of the mole. Greenhill has seen at least two bilateral cysts becoming much bigger 1-2 weeks after evacuation of the mole. Stoddard reports a case where the cysts increased from 6 cm. to 19 cm. in size three days after evacuation of the mole. They underwent spontaneous regression only after 4 months. Quigley reported a case of hydatidiform mole with severe toxaemia. Five weeks following evacuation of the mole, a lutein cyst which was not recognizable previously, increased in size to 14 cm.

These ovarian changes represent an exaggerated response of the ovarian tissue to abnormally great amount of gonadotropic hormone produced by the molar tissue. Although the exact mechanism is not clear, it must be comparable to the ovarian enlargement noticed in cases receiving HPG and HCG for induction of ovulation. Girouard, Barclay

and Collins record 17 cases unassociated with mole or choriocarcinoma, where the enlargement occurred presumably as a response to gonadotropic stimulation. Experimentally it has been shown that administration of human menopausal gonadotropin will cause formation of theca lutein cysts late in normal pregnancy. Lutein cysts can also occur with other conditions like congenital anomalies, foetal erythroblastosis and even in normal pregnancy.

Oestrogen excretion values are very low in hydatidiform mole in contrast to those in normal pregnancy. Absence of oestrogen suppression postulated for pregnancy may permit production of pituitary gonadotropin as the accessory factor for the formation of theca lutein cysts. Whereas the HCG titres are always elevated in the presence of hydatidiform mole, lutein cysts occur only in about 25%. The presence of the accessory substance, pituitory gonadotropin, may be the decisive factor in the formation of theca lutein cysts (White & Bradbury).

Another possible aetiological factor may be the age of the patient. Chun *et al* noticed that lutein cysts occur in patients below the age of 29 years. This agrees with the findings of Brews that development of large cysts is more common in the younger age groups.

Whatever the aetiology of these cysts, in the vast majority of cases they disappear spontaneously within a few weeks of evacuation of the mole and no particular treatment is indicated for their regression. Their removal is indicated only when there is exceedingly rapid growth with pressure symptoms or when accidents like torsion of the pedicle or rupture occur.

Torsion of the pedicle of a lutein cyst which occurred in the present case, seems to be very rare. It is so rare that, while presenting such a case at the Royal Society of Medicine in 1963, Clift stated that he searched the literature for 25 years and could find only one paper on the subject by Weill (1937).

Clift reported the case of a 28 year old primigravida, 13 weeks pregnant with hydatidiform mole. Patient presented herself with severe abdominal pain and distension of abdomen. On laparotomy, both ovaries were enlarged to $10 \times 7 \times 5$ cm and cystic and had undergone torsion of the pedicle. The right cystic ovary had sloughed away from its necrotic attachment. Most of the left ovary also required resection, but some ovarian tissue was conserved together with the left tube. On the 3rd day of laparotomy, the patient aborted a vesicular mole spontaneously. The uterus was only 12 weeks in size.

A case of acute torsion of an ovarian cyst after abortion of a hydatidiform mole is reported by Robert, H. Boury—Heyler and Robert, C. in the French literature. A 28 year old para 1 was evacuated of 12 weeks mole (uterine size was 18 weeks). Ten days after the evacuation, the patient was readmitted with severe abdominal pain. On laparotomy, there was a left lutein cyst as big as a rugby ball and it had undergone torsion 4 times. The right cyst was smaller and though haemorrhagic there was no torsion of the pedicle. Since both cysts were haemorrhagic,

a hysterectomy with bilateral oophorectomy was performed. There was no molar tissue left in the uterus which was about 8 weeks in size. Hervet from Tarnier clinic reported a similar case in 1957. Removal of the twisted cyst and conservation of the other ovary though very big was done with good results. Prof. Tow in his lectures on hydatidiform moles mentioned torsion of a lutein cyst as a possible accident. He had personal experience of a few such cases.

Histopathology of these cysts is that of cystadenomas in which all inner surfaces are infiltrated or replaced by cells of the luteal type. Varying degrees of thickness of the luteal layers suggestive of colloid ball formation are to be seen. Many of the cysts have a distinct and discrete yellow or grey peripheral rind.

Another feature of interest in the case reported may be mentioned in passing. In a matter of hours the patient developed all the signs of severe pre-eclampsia. Cromwell reported pre-eclampsia in 26.5% of hydatidiform moles. Chesley, Cosgrove and Preece collected 35 cases of probable eclampsia with hydatidiform mole. Dieckman found toxaemia in 3 of 30 cases, page 10 out of 30 and Accosta Sison in 31 out of 85 cases. Several observers have noted that toxaemia was severe only in cases where the uterus had reached the height of the umbilicus or beyond or after an amenorrhoea of 4 to 7 months.

Conclusions

1. Palpable lutein cysts of the ovary occur in about 25% of cases of hydatidiform mole.

2. The cysts may appear after the evacuation of the mole or they may start increasing in size after evacuation of the mole. They may reach enormous size in a matter of few days.

3. Almost all these cysts regress spontaneously after evacuation of the mole so that they do not need any particular treatment. The time taken for regression varies very much. They regress even if they have appeared after evacuation of the mole.

4. An occasional cyst, however, may cause complications by undergoing torsion or rupture or by causing pressure symptoms. Of these complications, torsion of the pedicle is the most common.

5. In dealing with these complications, it is not necessary to remove the whole ovary. Any viable part may be conserved.

6. There is no evidence of malignancy in these cysts even when they are very big and regress slowly.

7. Pregnanediol excretion may be raised in association with lutein cysts.

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