

**Case study**

Atypical postcesarean epithelioid trophoblastic lesion with cyst formation: a case report and literature review



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Summary We report an extremely rare case of atypical postcesarean epithelioid trophoblastic lesion with cyst formation. A 41-year-old Chinese woman presented with lower abdominal pain and menstrual disorder. Her serum human chorionic gonadotropin (hCG) was low (0.373 IU/L), and her urine hCG was negative. Ultrasound images showed a $3.7 \times 2.8 \times 2.5 \text{ cm}^3$ mass on the surface of the lower uterine segment, and a laparoscopy indicated a cystic mass in the serosal surface of the lower uterine segment. Histology indicated a cystic lesion consisting of epithelioid trophoblastic cells with an intermediate pattern between a classical placental site nodule and an epithelioid trophoblastic tumor; thus, the term *atypical postcesarean epithelioid trophoblastic lesion with cyst formation* was appropriate. As in atypical placental site nodule, serum hCG monitoring after treatment is necessary.

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1. Introduction

Intermediate trophoblastic lesions are clinicopathologically categorized into exaggerated placental site (EPS), placental site trophoblastic tumor (PSTT), placental site nodule (PSN), and epithelioid trophoblastic tumor (ETT). In terms of cell differentiation, EPS and PSTT are derived from implantation site intermediate trophoblastic cells, and PSN and ETT, from chorionic-type intermediate trophoblastic cells. As for biological behavior, PSN and EPS are benign lesions, but ETT and PSTT are malignant. Atypical PSN

(APSN) has only recently been identified as a suggestive borderline entity between PSN and ETT [1]. In this case report, we discuss an unusual atypical epithelioid trophoblastic lesion with cyst formation at the scar of the patient's cesarean section in the lower uterine segment.

2. Case report

A 41-year-old Chinese woman (gravida 7, parity 2) presented with lower abdominal pain and menstrual disorder. Her medical history included a cesarean section 4 years ago and a terminated cesarean scar pregnancy 2 years later. Her urine human chorionic gonadotropin (hCG) was negative,

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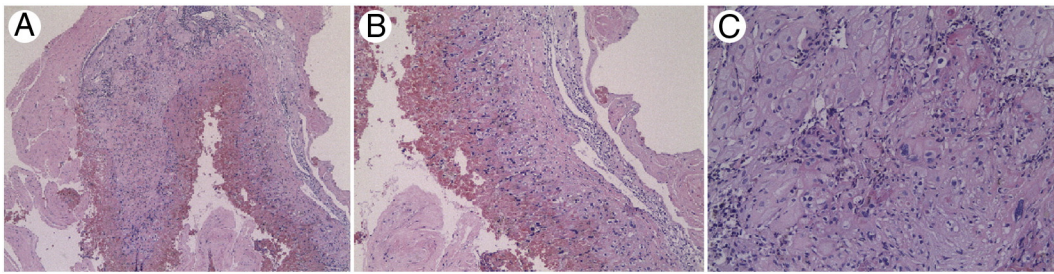


Fig. 1 Microscopic photograph of the uterine lesion. A, The lesion was well circumscribed without invading the surrounding myometrium (original magnification $\times 50$). B, The cyst contained fibrinous material, and the walls of the artery and vein were not invaded by tumor cells ($\times 100$). C, The cyst was lined by epithelioid cells with round nuclei and abundant eosinophilic cytoplasm ($\times 200$).

and her serum hCG was low (0.373 IU/L). An ultrasound examination showed a $3.7 \times 2.8 \times 2.5 \text{ cm}^3$ cystic mass on the surface of the lower uterine segment. The laparoscopy showed a 3-cm cystic mass at the serosal surface of the cesarean scar in the lower uterine segment. This lesion was removed for pathologic examination.

3. Pathologic findings

On macroscopic observation, the cystic mass was 3.5 cm in maximal diameter and surrounded by a thin layer of myometrium. The cyst was filled with brownish fluid with the uneven inner wall attached by clot-like materials. The thickness of the cystic wall was 2 to 3 mm.

Histologically, the lesion was well circumscribed and surrounded by normal muscle fibers without myometrial and vessel invasion (Fig. 1). The cystic wall was composed of several layers (usually 5-10) of mononucleated cells in nests and cords. The cells were of a uniform medium size, irregularly enlarged with hyperchromatic nuclei except for 1 to 2 inconspicuous nucleoli, along with abundant hyalinized or fibrinoid material in the center and a distinct hyaline matrix. These features are similar to those of epithelioid intermediate trophoblastic cells in PSN or APSN. Mitotic figures and necrosis were not found. Both ovaries and fallopian tubes were unremarkable.

Immunohistochemical staining is shown in Fig. 2. The characteristic pattern includes diffuse nuclear p63, cytoplasmic cytokeratin 18 (CK18), focal cytoplasmic inhibin and placental alkaline phosphatase, and negative human placental lactogen (HPL), CD146, and hCG. The Ki-67 index was moderate (14%).

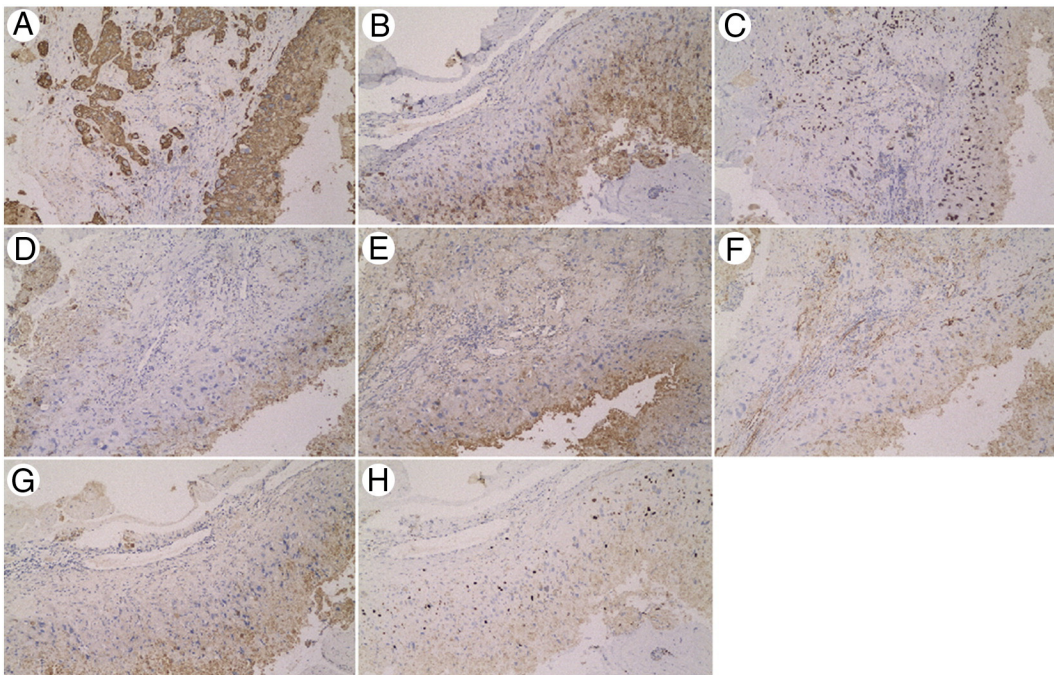


Fig. 2 Immunohistochemical photograph of the uterine lesion. The trophoblastic cells showed positive immunostaining for CK18 (A), inhibin (B), p63 (C), and placental alkaline phosphatase (D); negative staining for HPL (E), CD146 (F), and hCG (G); and a moderate Ki-67 index (H). A-H, Original magnification $\times 100$.

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