Ectopic Pregnancy Developing in a Cyst of the Canal of Nuck

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BACKGROUND: A cyst of the canal of Nuck is a rare cause of inguinal swelling in adult women. Ectopic

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pregnancies that implant to rare regions are sometimes challenging to diagnose.

CASE: A 45-year-old woman presented with painful swelling in her right groin with a positive urine pregnancy test. Ultrasonography, magnetic resonance imaging, and the histopathologic result of dilation and curettage suggested ectopic pregnancy in the right inguinal canal. Exploratory laparotomy, extraction of the right inguinal mass lesion, and closure of the deep inguinal ring were performed. The mass lesion measured 4×3 cm, and the histopathologic finding revealed an unusual ectopic pregnancy in the cyst of the canal of Nuck.

CONCLUSION: In women of reproductive age presenting with painful swelling of the groin, ectopic pregnancy in a cyst of the canal of Nuck should be ruled out.

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Acyst of the canal of Nuck is one of the causes of inguinal swelling in women; however, it is rare in adult women. The canal of Nuck is an evagination of the parietal peritoneum that accompanies the uterine

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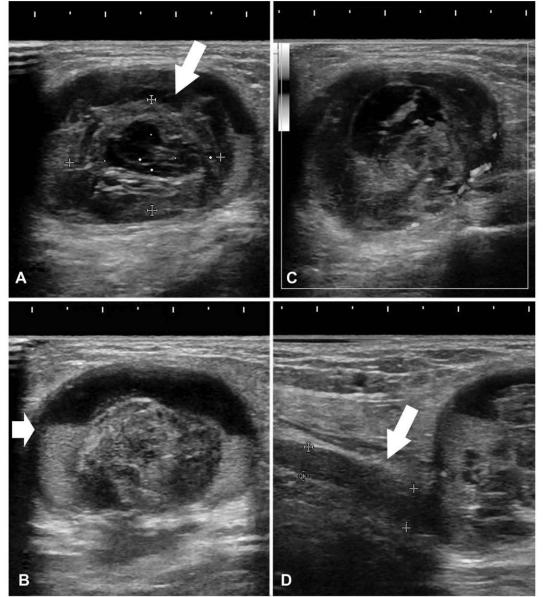


Fig. 1. Transabdominal ultrasonography of the right groin. **A.** A subcutaneous mass lesion measuring $34 \times 28 \times 24$ mm with a 21-mm \times 16-mm solid component in the interior of the mass (*arrow*) containing a 6-mm gestational sac-like cyst at its center with no fetus. **B.** The fluid-fluid level (*arrow*) suggests blood accumulation in the mass. **C.** Color Doppler image shows some vascularity around the mass and no vascularity in the mass. **D.** The mass lesion connects to the 4-mm-wide ligament (*arrow*) in the inguinal canal; however, no connection to the peritoneal cavity is detected.

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round ligament through the deep inguinal ring into the inguinal canal. Incomplete obliteration of the canal leads to a cyst of the canal of Nuck. Ectopic pregnancy is a common condition encountered by obstetrician—gynecologists. However, ectopic pregnancies in which villi implant to a rare region are sometimes challenging to diagnose, even for obstetrician—gynecologists. We present our experience with an unusual ectopic

pregnancy that developed in a cyst of the canal of Nuck.

CASE

The patient was a 45-year-old Japanese woman (gravida 5 para 4) with no relevant medical history. In June 2012, she first noticed swelling in her right groin, which

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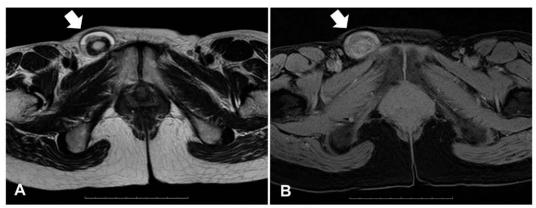


Fig. 2. Magnetic resonance imaging. T2-weighted **(A)** and fat-suppressed T1-weighted **(B)** images. The subcutaneous mass lesion (*arrows*) was observed in her right groin, which was revealed by transabdominal ultrasonography (Fig. 1). Magnetic resonance imaging revealed blood accumulation in the interior of the mass lesion.

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expanded gradually. Her last menstrual period had begun on May 8, 2013, and it lasted 5 days. Thereafter, she experienced irregular genital bleeding; however, she was unaware that she was pregnant. On July 6, 2013 (gestational week 8 calculated from the first day of the last menstrual period), she was referred to a neighborhood clinic with the chief symptoms of rapid expansion and pain in her right groin. The physician diagnosed an approximately 6-cm round swelling in the right groin, and the urine pregnancy test yielded a positive result. The physician referred her to the Department of Obstetrics and Gynecology of Fukaya Red Cross Hospital, Saitama, Japan.

Her serum human chorionic gonadotropin (hCG) level was 3,090 milli-international units/mL. Transvaginal ultrasonography and magnetic resonance imaging revealed no gestational sac in the uterus, no abnormal condition of the pelvic cavity, bilateral adnexa, and few ascites. Transabdominal ultrasonography revealed a subcutaneous mass lesion in her right groin measuring 34×28×24 mm (Fig. 1). Ultrasonography revealed a 21-mm×16-mm solid component in the interior of the mass, which contained a 6-mm gestational sac-like cyst at its center with no fetus. The mass lesion connected to the 4-mm-wide ligament in the right inguinal canal; however, no connection with the peritoneal cavity was detected. Magnetic resonance imaging revealed blood accumulation in the interior of the mass lesion (Fig. 2). To rule out the slight possibility of a normal intrauterine pregnancy, her serum hCG level and transvaginal ultrasonography were reevaluated 1 week later as follow-up.2 The reevaluated serum hCG level of 3,150 milli-international units/mL and transvaginal ultrasonography confirmed no normal intrauterine pregnancy. Dilation and curettage were performed according to protocol for the diagnosis of ectopic pregnancy used by our department. The histopathologic result of the dilation and curettage revealed decidual endometrium without chorionic villi. Moreover, the serum hCG level did not decrease after the dilation and curettage. These clinical results suggested that ectopic pregnancy developed in the right inguinal canal. However, this condition is extremely unusual; therefore, we were not exactly confident regarding the preoperative diagnosis at the time.

On July 17, 2013 (gestational week 10), surgical procedures including exploratory laparotomy, extraction of the right inguinal mass lesion, and closure of the deep inguinal ring were performed by general surgeons in cooperation with gynecologists. The right groin was opened by the anterior approach, subsequently revealing a 4×3-cm mass lesion connected to a ligament that appeared to be the round ligament. Therefore, the mass lesion was extracted with 3 cm of the ligament by high ligation after sufficient dissection (Fig. 3A and B). No hernia was observed in the right groin. No abnormal conditions were observed in the peritoneal and pelvic cavities. The histopathologic finding of the extracted mass revealed the diagnosis of ectopic pregnancy and endometriosis in the cyst of the canal of Nuck (Fig. 3C-G). After surgery, the serum hCG level decreased promptly, and no postoperative complications were observed.

COMMENT

We present the case of an ectopic pregnancy that developed in a cyst of the canal of Nuck, which is considered to be extremely rare. Moreover, the histopathologic finding of the lesion showed evidence of intralesional endometriosis in the cyst of the canal of Nuck, which is also a rare condition.

Ectopic pregnancy that developed in an inguinal canal is considered to be extremely rare. Using

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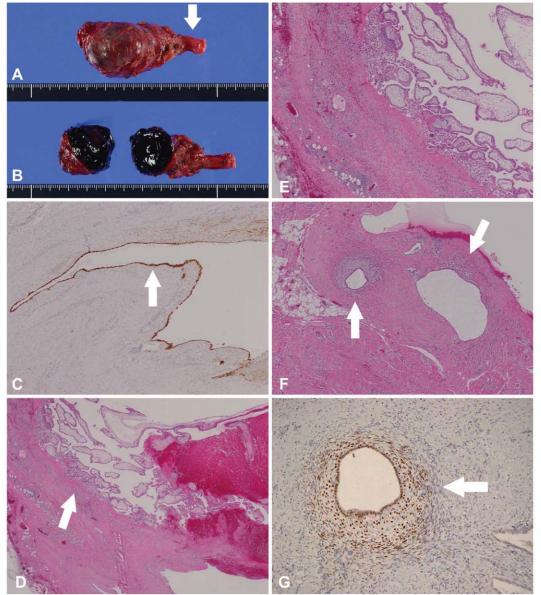


Fig. 3. Macroscopic and microscopic images of the extracted mass lesion. **A.** The mass lesion measuring 4×3 cm connects to the round ligament (*arrow*). **B.** Macroscopic image of the mass filled completely with coagulum. **C.** Microscopic image of calretinin immunohistochemical staining shows that the inner wall of the mass is covered by mesothelial cells (*arrow*). This suggests that the mass lesion originated from the canal of Nuck. Hematoxylin and eosin staining demonstrates villi implanting within the wall of the mass lesion at **D.** low magnification (the *arrow* indicates the villi) and **E.** high magnification. **F.** Hematoxylin and eosin staining under low magnification shows endometrial glandular and stromal cells within the mass lesion (the *arrows* indicate endometriosis). These cells are stained by immunohistochemical staining of estrogen (**G**, *arrow*) and progesterone receptors, confirming the diagnosis of endometriosis.

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PubMed, we performed a systematic search of the literature in October 2013 with the following key words: "pregnancy," "gestation," "abortion," "ectopic," "extrauterine," "fallopian tube," "tubal," "groin," "inguinal," and "Nuck." We did not identify exactly the same case through this search that included the references of the literature searched. However, we found three case reports in which ectopic pregnancy developed in the inguinal area. Levi³ reported ectopic pregnancy that occurred in

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the right inguinal canal. The patient had a surgical history of round ligament uterine suspension. The author determined that the midportion of the right fallopian tube was attached to the peritoneum at the right deep inguinal ring. The author concluded that the diagnosis was rupture of ectopic pregnancy, which was presumable as tubal in origin, with possible reimplantation between the rectus muscle and the fascia in the right inguinal canal. D'Souza et al4 reported a case involving operation for a left painful incarcerated inguinal hernia, in which a sliding hernia containing the left tubal pregnancy was found intraoperatively. Kay and Liang⁵ reported a case of ectopic pregnancy developing in an inguinal herniorrhaphy scar. A small connection between the canal of Nuck and the peritoneal cavity can exist.⁶ We were unable to find a visible connection between the cyst of the canal of Nuck and the peritoneal cavity in this case. However, the development of an ectopic pregnancy in the cyst of the canal of Nuck of the present patient suggested the existence of such a connection, although very minimal, in this case.

Endometriosis in the cyst of the canal of Nuck was observed in this case, suggesting that the patient had endometriosis in the cyst of the canal of Nuck before the pregnancy. Endometriosis in the canal of Nuck is a rare occurrence, with an incidence of 0.5%⁷ among all sites with endometriosis implants. Three-quarters of cases of endometriosis in the canal of Nuck are predicted to also involve pelvic endometriosis.7 In cases of endometriosis in the inguinal canal, incomplete excision of the round ligament in the groin can lead to local recurrence of the endometriosis if the round ligament is affected by endometriosis.8 In this patient, no endometriosis was observed in the pelvic cavity, and the round ligament in the groin was extracted with the mass lesion. The patient had no residual endometriosis.

This is an extremely rare case of an ectopic pregnancy that developed in a cyst of the canal of Nuck. While examining a woman with painful swelling in her groin, the possibility of ectopic pregnancy should be explored to rule out a condition similar to the present case.

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Case Report

Ectopic Pregnancy Developing in a Cyst of the Canal of Nuck

Nuck 管嚢胞内に発生した異所性妊娠

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日本語要約

【背景】Nuck 管嚢胞は女性の鼠径部腫脹を起こす原因の一つであるが、成人女性で見つかることは稀である。また稀な部位に着床した異所性妊娠では診断に苦慮することもある。

【症例】症例は45歳女性。5回経妊、4回経産。主訴は右鼠径部の疼痛を伴う腫脹。尿妊娠反応は陽性を示し、血清 hCG は3090 mIU/mL。超音波断層法で子宮内に胎嚢は見られず、右鼠径部には腫瘤が認められた。その内部は、胎嚢様の嚢胞を伴う充実性部分とそれを取り囲む液体貯留で構成されていた。MRIでその腫瘤内の液体貯留は血液であると示唆された。子宮内容除去術の結果、絨毛組織は認められなかったので、右鼠径部の異所性妊娠が強く疑われた。試験開腹術、鼠径部腫瘤摘出、内鼠径輪縫縮術が行われ4×3 cmの腫瘤が摘出された。病理組織診断の結果、Nuck 管嚢胞内に発生した異所性妊娠と診断された。

【結論】妊娠可能年齢の女性において鼠径部の腫脹を認めた場合には、同部位の異所性妊娠を鑑別診断として考慮すべきである。