

자궁선근증을 가진 초산모의 조산

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Preterm delivery in a primigravida with uterine adenomyosis

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Adenomyosis has been well known to be associated with infertility, spontaneous rupture of the uterus during labor in a primiparous woman, spontaneous preterm labor, preterm premature rupture of membranes (PPROM), and delayed postpartum hemorrhage. We recently experienced a case of preterm delivery at 29 gestational weeks in a primigravida woman with uterine adenomyosis. We report the case of preterm delivery accompanied by various complications such as uncontrolled pain, preterm labor, and oligohydramnios in a woman with uterine adenomyosis.

Key Words: Uterine adenomyosis, Abdominal pain, Preterm birth

Adenomyosis was firstly used by discovered by Frankl, and pathologically defined by Bird et al. in 1972; the benign invasion of endometrium into the myometrium, producing a diffusely enlarged uterus which microscopically exhibits ectopic, non-neoplastic, endometrial glands and stroma surrounded by the hypertrophic and hyperplastic myometrium.^{1,2} Several previous studies were reported that the risk of adenomyosis was increased after spontaneous abortions and dilation and curettage.^{3,4} Uterine adenomyosis most commonly occurs in multiparous wom-

en in their late 30s or 40s, but rare in nulligravida women. Uterine adenomyosis can be classified into two categories: focal adenomyosis with lesions localized in the anterior or posterior wall; and diffuse adenomyosis with lesions in the entire uterus. Regarding obstetric problems related with adenomyosis, recent reports showed that adenomyosis was associated with infertility,^{5,6} spontaneous rupture of the uterus during labor in a primiparous woman,^{7,8} spontaneous preterm labor, preterm premature rupture of membranes (PPROM),⁹ and delayed postpartum hemorrhage.¹⁰ We recently experienced a case of preterm delivery at 29 gestational weeks in a primigravida woman with uterine adenomyosis.

접 수 일 : 2009. 11. 2.
채 택 일 : 2009. 12. 1.
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Case Report

A 35-year-old woman, gravid 1 para 0 was admitted to our hospital with acute lower abdominal pain at 15th gestational week. The patient had tried unsuccessfully to become pregnant for about 2 years. In infertility work up, bilateral tubal obstruction and adenomyosis were noted. She became pregnant after undergoing in vitro fertilization and embryo transfer (IVF-ET) to be pregnant of adenomyosis. After admission, transabdominal ultrasonography (Aloka Prosound alpha-10,



Fig. 1. At 15th gestational week, uterine adenomyosis was seen in the mainly fundal area (thick white arrow). Normal gestational sac and fetus were visible in lower uterine segment area.

Tokyo, Japan) revealed uterine adenomyosis including normal gestational sac in lower uterine segment (Fig. 1). Thickened uterine wall in adenomyosis was extended from fundal area to mid portion of uterus, and in this area, there was a narrow endometrial cavity without gestational sac. Anterior wall thickness of uterus was about 10 cm, and posterior was about 3 cm. Routine chemistry, ultrasonography for exclusion of the acute appendicitis, and tococardiogram for uterine contraction was performed. Laboratory results showed hemoglobin 10.8 g/dL, white blood cell count $20,19 \times 10^3/\mu\text{L}$, neutrophil segmented 81%, C-reactive protein (CRP) 17.96 mg/dL (reference range: 0.01~0.3 mg/dL). With leukocytosis and elevated CRP being noted, and she had mild fever (body temperature: 37.5°C). Acute appendicitis, placental abruption, ureteral stone, and pyelonephritis were ruled out by physical examination, ultrasonography, and laboratory test. Uterine contraction was detected at 15-minutes interval. She was diagnosed with threatened abortion, and adenomyosis. She was prescribed analgesics including acetaminophen and codeine for pain control, and antibiotics with ampicillin and sulbactam were taken. Uterine contraction was subsided after hydration and bed rest. At hospital day 10, uterine cervix length es-

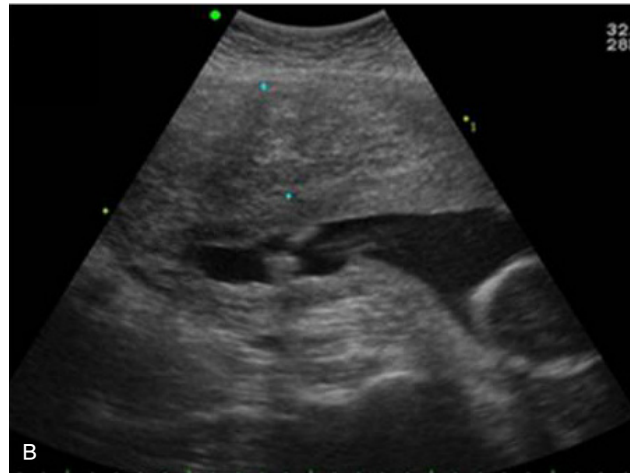
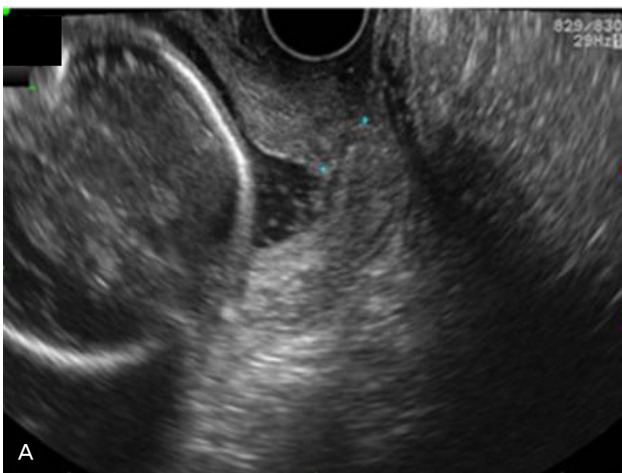


Fig. 2. At 24th gestational week, shortening of uterine cervix with funneling was visible by transvaginal ultrasonography (A), and uterine adenomyosis with normal gestational sac was seen in transabdominal ultrasonography (B).

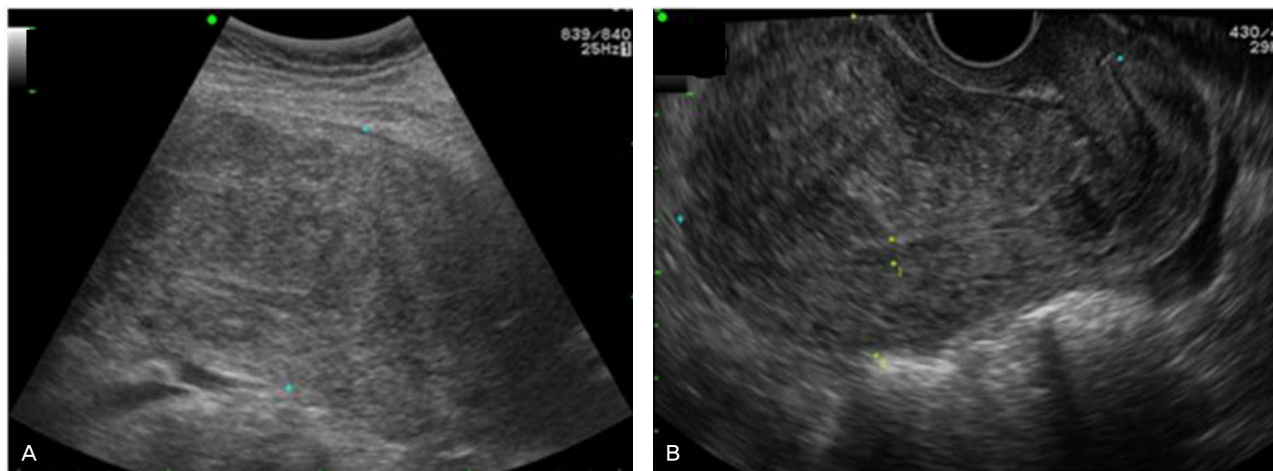


Fig. 3. Uterine adenomyosis was serially decreased in size at 2 weeks (A), and 8 weeks (B) after delivery.

timated by transvaginal ultrasonography was 3.1 cm, abdominal pain was subsided, and she was discharged without uterine contraction.

After 24th gestational week, she was repeatedly admitted to the hospital because of severe lower abdominal pain, and regular uterine contraction. Ultrasonography (Accuvix, Medison, Seoul, Korea) showed that uterine cervix was shortened and funneled (cervix length: about 1.3 cm) (Fig. 2). Tocolytics with ritodrine and analgesics including acetaminophen, codeine, nalbuphine, and pethidine were taken for uterine contraction and pain control. Pain control was daily performed due to extreme and frequent pain on low abdomen. At hospital day 6, uterine contraction was disappeared, but abdominal pain was sustained. At hospital day 9, abdominal pain was decreased, so she was discharged.

At 27th gestational week, she was readmitted with severe abdominal pain, and regular uterine contraction. Uterine cervix length estimated by transvaginal ultrasonography was 1.0 cm. In fundal area, anterior wall thickness was about 9 cm, and posterior was 3 cm. Estimated fetal weight was adequate to gestational age, but amniotic fluid index was 8 cm. There was no evidence of preterm premature rupture of membranes

(PPROM). During 2 weeks, tocolytics with ritodrine and analgesics including acetaminophen, codeine, nalbuphine, and pethidine, and betamethasone were taken. At 29th gestational week, emergency cesarean section was done, because oligohydramnios (amniotic fluid index: 4), breech presentation by ultrasonography, uncontrolled abdominal pain, and fetal distress (moderate to severe variable deceleration during fetal monitoring) were noted. A male baby weighing 1,140 g was born. Apgar scores were 3 at 1 minute and 6 at 5 minute. The baby was moved to the neonatal intensive unit (NICU) with endotracheal intubation and oxygen supply. The estimated blood loss during surgery was about 500 mL. The baby had respiratory distress in the early period of NICU admission but was recovered in NICU and was discharged from the hospital after 72-day stay, and so was the mother 6-day after the operation without any other problem. Ultrasonography at 2 and 8 weeks after delivery was performed. Uterus size was serially decreased (Fig. 3).

Discussion

Because uterine adenomyosis is associated with in-

fertility, spontaneous uterine rupture, preterm labor, and preterm premature rupture of membranes, pregnancy in a primigravid woman with uterine adenomyosis is rare and highly risky.

The effects of myoma on pregnancy or pregnancy on myoma are well known,^{11,12} but adenomyosis not. A recent case report showed that a rapidly growing adenomyosis was identified during the first trimester in a 35-year-old infertile woman achieving pregnancy after controlled ovarian hyperstimulation.¹³ Although the pregnancy was terminated at the 18th gestational week after premature rupture of membranes following genetic amniocentesis, severe abdominal pain by red degeneration was similar to our case. No imaging study for pregnant uterus for prepregnant and 1st trimester period was performed, so we don't know whether the enlarged uterine was rapidly grown during first trimester. But, pregnancy in our case was maintained to 29th gestational week, and the more the pregnancy progresses, the more severe the intensity of pain was. During follow up periods, there was no evidence of rapid growing adenomyosis. From 15th gestational to delivery, one of major problems of pregnancy maintenance was pain in our case. Various analgesics were fully used for pain control, but the pain was not satisfactorily controlled. It is well known that the most complaint associated with uterine myoma during pregnancy is pain. Although initial hypotheses suggested that pain was secondary to fibroid growth during pregnancy, painful episodes appear to be unrelated to absolute fibroid size or growth.¹⁴ Prospective, longitudinal studies have also failed to document sig-

nificant change in fibroid size during pregnancy.¹⁵⁻¹⁷ A case for successful pregnancy outcome in a woman with large uterine intramural myoma was reported by Skrablin S et al. Pregnancy with about 13 cm sized intramural myoma in fundal area was reported. That case showed that abdominal pain did not worsen during pregnancy, and an elective cesarean section was performed at 38 weeks.¹⁸ However, it is little known that pregnancy with adenomyosis is associated with pain. The most common cause of neonatal morbidity reported in any of these studies was preterm delivery. Women with myomas in pregnancy seem to be at increased risk of threatened preterm labor with reported rates of up to 21.5%, and the risk of preterm labor correlated positively with the size of the largest fibroid. And it is generally accepted that tocolytic agents used in pregnancy with myoma and preterm labor is not different from with only preterm labor.¹⁹⁻²²

In this case, the pregnant woman had low abdominal pain, mild fever, leukocytosis, and increased inflammatory markers. These symptoms and signs were similar to that of pregnancy with red degeneration of myoma. Also, it was difficult to differentiate the pregnancy with uterine adenomyosis from other medical-surgical, and obstetrical problems such as appendicitis, placental abruption, preterm labor, ureteral stone, pyelonephritis, and chorioamnionitis.

Here, we report the case of preterm delivery accompanied by various complications such as uncontrolled pain, preterm labor, and oligohydramnios in a woman with uterine adenomyosis.

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= 국문초록 =

임신과 관련하여 자궁선근증은 불임, 초산임신에서의 자연적인 자궁파열, 조기진통, 조기양막파수, 산후출혈 등과 관련이 있는 것으로 알려져 있다. 최근 본원에서는 자궁선근증을 가지고 임신된 초산모를 임신 29주까지 유지한 후 조산하게 된 경험을 하게 되었다. 임신기간 중 산모는 반복적인 조기진통과 다양한 진통제에도 잘 조절되지 않는 극심한 복통을 호소하였고, 원인이 밝혀지지 않는 양수 감소 등의 증상이 나타나서 결국 임신 29주에 조산으로 분만하게 되었다. 수술 후 태아는 27일 만에, 산모는 6일 만에 특별한 합병증 없이 퇴원하였다. 저자들은 초산모에게는 드물게 생기며, 여러 가지 위험요인을 안고 있는 자궁선근증과 동반된 임신을 경험하였고, 임신 29주까지 성공적으로 임신을 유지하였기에 간단한 문헌고찰과 함께 보고하는 바이다.

중심단어: 자궁선근증, 복통, 조산