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선천성 회음부 지방종의 산전초음파 진단 1예

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Prenatal sonographic detection of congenital perineal lipoma in a newborn girl

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We report on a newborn girl presenting with a 2.0×1.5 cm sized pinkish, doughy lump arising between right labia majora and anus. We performed antenatal sonogram at 33 and 36th weeks gestation. A polypoid mass of 0.8×1.0 cm sized in size was noted on ultrasonography. After birth, we observed a skin-covered protruding mass not to detect other anatomic anomalies-spinal anomalies, anorectal malformations, etc. After 3 months, excision of the perineal mass was done at the Department of Pediatric Surgery of our hospital. Mature fat cells were noted on histopathological exam. To our knowledge, there are few studies in the English literatures about congenital perineal lipoma without combined structural anomalies.

Key Words: Lipoma, Perineum, Ultrasonography

Lipomas are common, non-cancerous, soft, fatty tissue growths in adults. They can occur anywhere in the body, and one or more lipomas may be present at the same time. However, in neonates, lipomas are very rare. Among them, few cases are found in the perineum. In Korea, 2 case report including 6 male neonates and children have been reported on department of urology.[1,2] All cases were associated with scrotal anomalies. There was no mention to prenatal diagnosis. Our case is the first report in female neonate to perform prenatal work-up without anomalies. In this case, we describe in a serial course which is focused on prenatal diagnosis. Because we experienced one example, we report it with considering documents.

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Case Report

A 30-year-old healthy mother, gravida 2, abortus 1, was referred to our hospital for evaluation of an echoic mass of about 0.7 cm in sized by ultrasonography at 32 weeks gestation. At 33 weeks gestation, sonographic examination using a 5-1-MHz transabdominal transducer of ultrasound (Philips iU22, Bothell,WA,USA) showed an encapsulated, homogeneous, echogenic mass containing lobular structures consistent with a polypoid mass of fat measuring

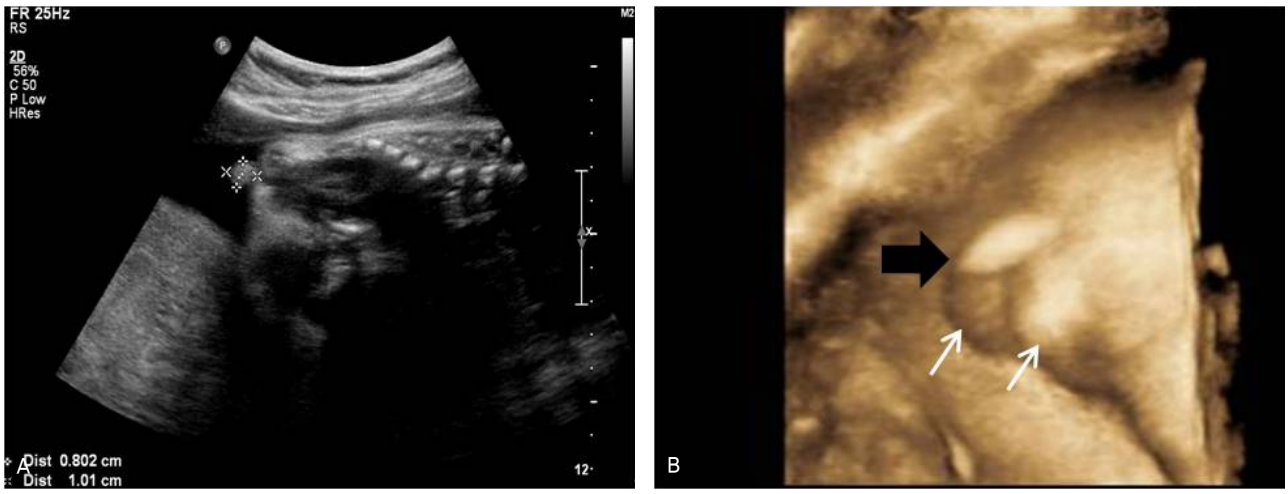

Fig 1. At 33 weeks gestation, a 0.8×1.0 cm sized, an encapsulated, homogeneous, echogenic mass was shown by ultrasound. (asterisk) (A) A 3D image of the same lesion (black arrow) and labia majora (white arrows) (B).

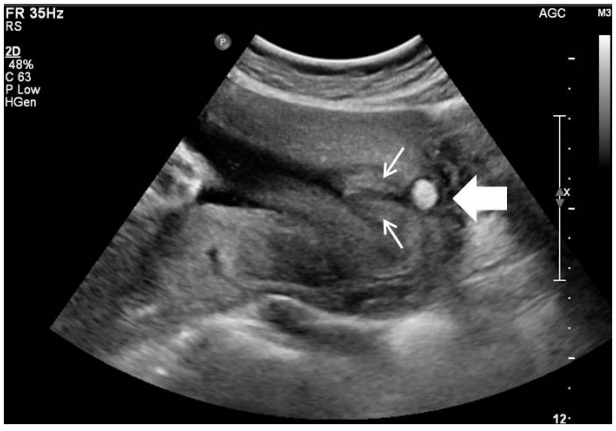

Fig 2. At 36 weeks gestation, it was shown an encapsulated, homogeneous, echogenic mass containing lobular structures consistent with a polypoid mass of fat (thick white arrow) and labia majora (thin white arrows).

0.8×1.0 cm (Fig. 1A). There was no vascularization within the mass by color and power Doppler imaging, and the scans did not disclose abnormalities on whole skelectal structures. A 3D image of the same lesion using a 6-2-MHz curved-array transabdominal transducer of ultrasound (Philips iU22, Bothell, WA, U.S.A.) revealed a clearly protruding mass between the anus and vulva (Fig. 1B). We diagnosed the perineal mass as skin tag or lipoma by ultrasound. Three weeks later, follow-up ultrasound demonstrated no interval change. (Fig. 2) On January 1, 2009, she was admitted to the hospital to deliver her baby after her water broke and she was experiencing regular labor pain at 2-minutes intervals. Two hours later, a female baby weighing 2,960 g was born at 39 weeks gestation by normal vaginal delivery. 1- and 5-minute Apgar scores were 7 and 8, respectively, after delivery. The baby was moved to the nursery room. Grossly a 2×1.5 cm sized well-defined, oval-shaped and exophytic tumor was noted in the perineum between a right labia majora and anus (Fig. 3). There were no gastrointestinal or genitourinary abnormalities. Assessment other systemic dysfunctions was not necessary. When the baby was 2-months-old, the lesion was totally removed in the prone position on a surgical bed under general anesthesia. The tumor measured a 2.3×1.5×1 cm in size, which histopathologically demonstrated a lipoma with mature fat tissue. There were no postoperative complications in passing urine and stool. The wound site was clear and healed well. On postoperative day 4, she was discharged.

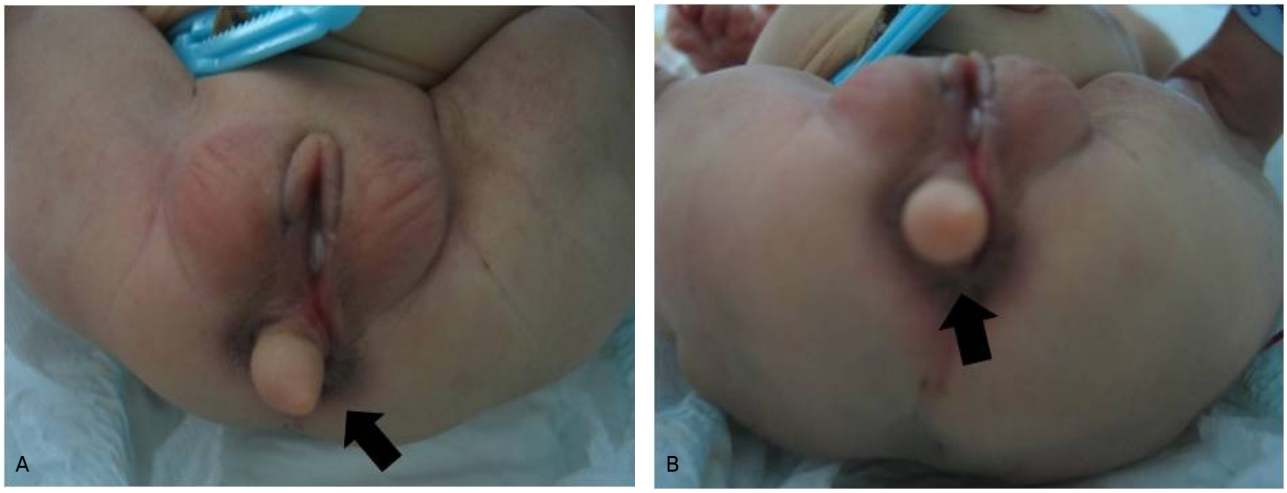

Fig 3. Afterbirth, we pictured a 2×1.5- cm sized well-defined, oval-shaped, exophytic mass in the perineum between the right labia majora and anus (black arrows) (A), (B).

Discussion

Fetal congenital perineal lipoma have rare prevalence.[3] Lipoma presenting in the perineum is particularly a rare. In the past few decades, not many reports of perineal lipoma including 28 cases have been published.[3,11] Among the reported cases, its incidence is higher in males is more than in females. Additionally, these cases of perineal lipoma are commonly combined with anorectal anomalies. Some of these in male were present accessory scrotum.[2,4-6,8,9] In females, we found thirteen cases of perineal lipomas in PubMed.[7-11] Except for 1 case, 10 simultaneously occurred with an accessory labioscrotal fold or anorecrtal malformation simultaneously. To date, the etiologic relationship between perineal lipomas and anorectal malformations is still unknown.[4,6,8,12]

When perineal mass is detected in perinatal periods, we need to consider the several entities[3] including sacroccygeal teratoma, lipoblastoma[13] liposarcoma, polyp, prolapse, enterogenous cyst, (myelo-) meningococele, hamartoma,[14] ependymoma,[15] an ectopic or an accessory scrotum,[3,16] and, rarely, inflammatory lesions etc.[17]

In our case, a non-specific, homogeneous, echogenic, well-contoured and encapsulated mass was shown by a curved array transabdominal transducer. Additionally, we performed 3D ultrasound to provide a realistic image of the mass. This enabled both precise diagnosis of the perineal lipoma subsequently confirmed by histopathology and patient counseling.

Conclusion

As we know, our case is the first literature of sonographic finding about congenital perineal lipoma without other combined systemic anomalies in Korean obstetrics and gynecology. We hope this case to be an additional report for reference when look on a congenital perineal mass on ultrasonography.

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= 국문초록 =

태아의 선천성 지방종은 매우 드문 질환이며, 다른 증례 보고들에 의하면 항문 요로계를 포함한 다른 구조적 동반 기형을 가지고 있는 경우가 대부분이며, 여아보다 남아에서 많았다. 본 저자들은 30세 초산부에서 자궁내임신 33주에 산전 초음파상 태아의 선천성 회음부 지방종 소견을 보였던 1예를 경험하였다. 자궁내임신 33주와 36주에 2D 및 3D를 이용해 산전 초음파를 시행하였으며, 출생 후 육안 및 기능적으로 별다른 이상 소견 보이지 않았고, 2달 후 단순 지방종절제술 시행 후 양호한 경과를 보였다. 여아에서 동반되는 다른 기관의 기형 없는 선천성 회음부 지방종의 산전 초음파 소견은 국내에서 보고된 적이 없었기에 이를 문헌고찰과 함께 보고하는 바이다.

중심단어: 지방종, 회음부, 초음파