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Case Report

Nonreducible Inguinal Hernia Containing the Uterus and Bilateral Adnexa in a Rhesus Macaque (*Macaca mulatta*)

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Inguinal herniation of abdominal viscera is a relatively common condition in both humans and domestic animal species. In captive rhesus macaques (*Macaca mulatta*), the highest incidence occurs in overweight, aged males. However, inguinal herniation of the uterus with bilateral adnexa is extremely rare in both human and veterinary medicine. Here we report a previously undescribed uterine inguinal herniation with bilateral adnexa in a 3-y-old female rhesus macaque. Although uterine herniation remains a rare condition in rhesus macaques, it should be considered as a differential diagnosis in animals with unilateral subcutaneous enlargements in the inguinal region.

Abdominal hernias are a common condition in both human and veterinary medicine with a reported 20 million human hernias repaired globally each year.[7] The most common abdominal hernia types are inguinal and femoral,[7] and an estimated 75% of abdominal hernias in humans are of the inguinal type.[19] Inguinal herniation has been documented in a variety of species, with pigs (*Sus scrofa domesticus*), cats (*Felis cattus*), and dogs (*Canis lupus familiaris*) serving as animal models of disease progression and repair techniques.[5] Although there is limited data on the incidence of inguinal herniation in rhesus macaques (*Macaca mulatta*), this condition is typically an incidental finding in overweight, aged males[1,15,20] but also may occur secondary to trauma,[15] congenital abnormalities,[15] or in utero lead exposure.[14] Five types of inguinal herniation have been described in the human literature[10] and involve a wide variety of incarcerated organs including omentum, bowel, uterus, ovaries, and fallopian tubes.[4,20] Inguinal hernias are particularly prevalent in infants, representing one of the most frequently observed congenital defects; in females, these hernias often contain a single ovary along with the ipsilateral fallopian tube, at a reported incidence of 0.16% to 0.80%.[6] Inguinal hernias containing the uterus and bilateral adnexa are extremely rare in humans, with fewer than 10 cases reported.[16] To our knowledge, this report is the first description of an indirect inguinal hernia containing the uterus and bilateral adnexa in a rhesus macaque.

Case Report

A 3-y-old, captive-reared, female, Indian-origin rhesus macaque was born at Oregon National Primate Research Center and was maintained on a breeding colony protocol. The Center is an AAALAC-accredited facility, and all animals are housed and cared for in accordance with the *Guide for the Care and Use of Laboratory Animals*.[12] Routine husbandry parameters include 12:12-h light:dark cycles; monitored temperature, humidity, and ventilation; commercial NHP chow (Purina 5000, Purina Mills, St Louis, MO); daily dietary supplementation with fresh fruits and vegetables, and free-choice water. At the time of initial evaluation, the patient was not assigned to a research protocol and was presented for pre-assignment physical examination. The patient had a history of mild splenomegaly and lymphadenomegaly, with peripheral lymph nodes measuring 2 to 3 times the normal size. Given that juvenile macaques frequently have mild enlargement of the spleen and peripheral lymph nodes and because her CBC and serum chemistry parameters were within normal limits, no further diagnostics were pursued at that time. During physical examination under ketamine anesthesia (10 mg/kg IM; Ketasthesia, Butler Shein Animal Health, Dublin, OH), a firm, immobile subcutaneous mass (length, 4 cm; width, 2 cm) was palpated in the left inguinal region (Figure 1). Bimanual uterine palpation revealed a 90° leftward turn between the cervix and uterine body. Despite persistent mild splenomegaly, all other physical exam findings were within normal limits. There was no evidence of peripheral lymph node enlargement, and the patient's CBC and serum chemistry parameters were unremarkable. According to palpation the mass was most likely uterus.

In light of the physical examination findings, an ultrasound exam of the abdomen and inguinal region was performed. The uterine body was normal in size and echogenicity, with a clearly defined hyperechoic endometrial stripe. However, the organ was displaced from its normal location: the uterus was herniated through the inguinal ring and corresponded to the externally palpable mass (Figure 2). Ultrasonography did not reveal either ovary within the hernial sac, and the remainder of the ultrasound exam was unremarkable. Given the presumptive diagnosis of inguinal herniation of the uterus, the patient was scheduled for a herniorrhaphy.

Surgery. The macaque was sedated with ketamine hydrochloride (12 mg/kg IM; Ketathesia, Butler Schein Animal Health) combined with glycopyrrolate (0.01 mg/kg IM; West-Ward, Eatontown, NJ). A local block consisting of 0.5% bupivacaine

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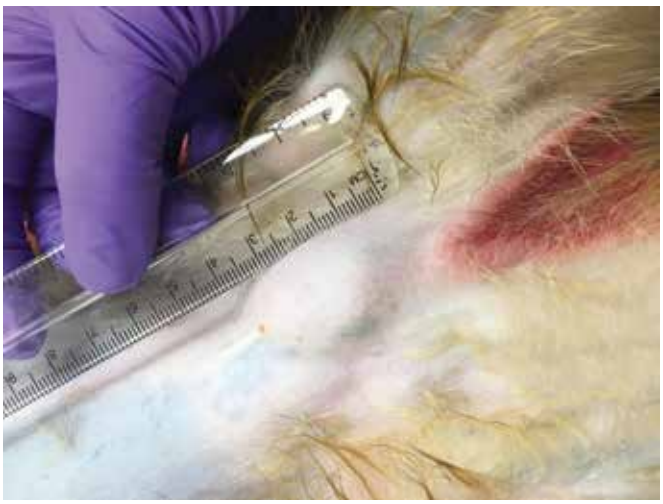

Figure 1. Palpable subcutaneous mass in the left inguinal region of a 3-y-old female rhesus macaque.

Figure 2. Subcutaneous mass with clearly defined hyperechoic endometrial stripe suggestive of herniated uterus.

(0.8 mL ID; Hospira, Lake Forest, IL) combined with 1% lidocaine with epinephrine (0.2 mL ID; APP Fresenius Kabi USA, Lake Zurich, IL) was administered at the proposed incision sites. The animal was intubated, and general anesthesia was maintained by using 1% to 3% isoflurane (Piramal Healthcare, Andhra, India) combined with 100% oxygen. Hydromorphone hydrochloride (0.12 mg/kg IV; Hospira) was administered for analgesia, and intravenous fluid support (10 mL/kg/h IV; Lactated Ringer Solution, Baxter Healthcare, Deerfield, IL) was provided throughout the procedure.

With the macaque in dorsal recumbency after sterile prepping and draping, a linear 4-cm incision, parallel to the long axis of the leg, was made over the herniated tissue in the left inguinal region, with subsequent dissection to and penetration of the vaginalis. All reproductive organs including the uterus, ovaries, oviducts, and fimbria along with a small portion of the omentum were present in the hernia sac (Figure 3). Cephalad enlargement of the inguinal canal was performed, with digital elevation of the apex. The reproductive organs and omentum were manually reduced into the pelvic cavity, followed by confirmation of hemostasis. The left round ligament appeared thickened and contracted, possibly resulting in retraction of reproductive tract through the inguinal ring. This ligament was partially transected by using electrocautery to allow replacement of the tract into the abdomen. A single nonabsorbable cruciate suture was used to partially

Figure 3. Opened left inguinal hernia sac containing uterus, ovaries, oviducts, fimbria, and omentum.

reduce the internal inguinal ring. Interrupted nonabsorbable sutures were used to partially close the lateral aspect of the external inguinal ring, resulting in an approximately 80% reduction of the aperture. The subcutis and skin were closed with continuous and intradermal absorbable sutures, respectively.

During the surgical procedure, to confirm normal reproductive anatomy and return of the reproductive structures to their appropriate anatomic locations after hernia repair, exploratory laparoscopy was performed to visualize the caudal abdominal cavity. The macaque was transitioned to the Trendelenburg position, and a Verres needle was inserted through a 1-cm supraumbilical skin incision followed by insufflation to 15 mm Hg pressure with CO_2 gas. The Verres needle was replaced by a 6-mm trocar and sheath and 5-mm telescope. A right paralumbar accessory port (size, 5 mm) was placed, through which grasping forceps were inserted for manipulation of viscera. The uterus had become slightly twisted and retroflexed caudally after reduction of the hernia; it was repositioned to a normal orientation by using the graspers. All tissues appeared well vascularized and healthy, and no other anatomic abnormalities were observed. A portion of the transected round ligament still extended through the left internal inguinal ring. The laparoscopic instruments were removed, and the incisions were closed with interrupted absorbable sutures in the rectus fascia and skin. The macaque recovered on the operating table until extubation and then was returned to its homecage.

Standard postoperative analgesia was provided (hydromorphone hydrochloride, 0.25 mg/kg IM every 4 to 6 h during the day; buprenorphine, 0.06 mg/kg IM once daily in the evening; Hospira) for 72 h after surgery. Daily postoperative monitoring and assessment of pain and distress were provided by surgical veterinary staff. The macaque recovered well from the procedure.

Discussion

During normal embryologic development, the gubernaculum, the immature basis of the suspensory ligaments of the ovaries and round ligaments of the uterus, develops in the mesonephric fold and provides partial stabilization to the reproductive organs.[2] These ligamentous structures prevent the descent of the ovaries through the open vaginal process. The round ligament of the uterus passes through the inguinal canal and attaches to the labia majora. The vaginal process normally is

reduced by approximately 8 mo of gestation in humans, which correlates to 5 mo of gestation in rhesus macaques. However, if the vaginal process remains patent, it is renamed the canal of Nuck and becomes the site for potential indirect inguinal herniation in females. The suspensory ligaments of the ovaries extend from the lateral pelvic walls and provide additional stabilization to prevent herniation.[2]

Several theories regarding the etiology of uterine and ovarian inguinal herniation are available, including ovarian ligament laxity, mullerian duct abnormality, and ovarian ligament attachment defects. A defect or laxity within the ovarian ligaments has been proposed to allow reproductive organs to herniate through the canal of Nuck.[6,9,17] Elongated ovarian suspensory ligaments were cited as the primary cause of herniation in the case of a 1-y-old infant with an indirect sliding hernia that contained both ovaries and the left fundus of the uterus.[9] Another author postulated that the broad ligament of the uterus or ovarian suspensory ligaments might have been weak in the case of a 1-mo-old with a uterine inguinal hernia.[17] This laxity, in conjunction with increased intraabdominal pressure associated with crying, might ultimately allow herniation.[17] Indirect inguinal hernias that are diagnosed after puberty and occur secondary to a developmental abnormality have been proposed to arise after failure of fusion of the mullerian ducts, the embryologic precursor to the fallopian tubes, uterus, cervix, and cranial aspect of the vagina.[8] Nonfusion leads to separation of the uterine cornua and increased mobility of the uterus and ovaries.[8] This defect occurred in several adult women (age, 16 to 37 y), who had inguinal hernias containing fallopian tubes, ovaries, and a unicornuate uterus.[3,8] The final theory builds on that involving ovarian ligament laxity but proposes a defect in the attachment points of the stabilizing ligaments. In cases of ovarian herniation, the suspensory ligament has been proposed to attach to the vaginal process instead of its normal attachment on the fallopian tube,[2] consequently pulling the ovary and potentially the uterus into the hernial sac.[2] Although the ligament found in the hernial sac is widely accepted to be the round ligament of the uterus, in reality, it might be the inappropriately attached suspensory ligament of the ovary.[2]

Regardless of the etiology of the hernia, there is a classic clinical presentation in humans. Most patients present around 30 d of age with a non-painful, irreducible large swelling in the left inguinal region.[4,6,11,16,17] Although our patient did not present in infancy, the uterine hernia might have been masked during her juvenile exams by peripheral lymphadenopathy that is commonly seen in this age group. Because pregnancy examinations are not routinely performed on animals this young, the herniated uterus was not noted on physical exam. As the size of her inguinal lymph nodes decreased, and her incarcerated uterus continued to grow, the herniation became more apparent. In adult humans, inguinal herniation of female reproductive organs often presents with pain in addition to a bulge in the left inguinal region.[3,8,13] The presentation of our patient follows the more classic infant presentation of left inguinal herniation of the uterus and adnexa without associated clinical signs. The likely etiology for our patient's herniation follows the first theory of ovarian ligament laxity: ligament laxity during embryonic development allowed for distal migration of the uterus and adnexa and entrapment as the vaginal process closed. The herniation became more apparent as the entrapped uterus enlarged with age.

Although this case of herniation appears to be an isolated incident in rhesus macaques, there is some evidence of inheritance in the human literature. An 11-wk-old infant with ovarian herniation through the left inguinal ring had 4 siblings, 3 of whom also presented with the same condition.[11] Another author described a 2-mo-old infant with a left inguinal hernia and a twin brother with bilateral inguinal herniation.[4] Human infants born prematurely, at less than 32 wk of gestation, have an increased risk of inguinal hernias.[18] These infants might experience decreased intrauterine nutrition and delayed contraction of the vaginal process.[18] This time period coincides with the point at which the vaginal process is normally eliminated during development, perhaps explaining why premature infants are born with an intact canal of Nuck and predisposition to herniation.[18] To investigate the role of inheritance in macaques, we performed a 4-generation pedigree analysis that revealed no evidence of inheritance. Intrauterine infection with human cytomegalovirus has also been associated with developmental growth retardation and an increased chance of an inguinal hernia.[18] Because rhesus cytomegalovirus is ubiquitous in NHP breeding colonies, we cannot exclude it as a possible etiology in this case.

In summary, this case represents the first report of indirect inguinal herniation of the uterus and adnexa in a rhesus macaque. Although uterine inguinal herniation is a rare condition that occurs most often in infant humans, it can present in subadult macaques without any associated clinical signs and with few observable abnormalities. As such, uterine inguinal herniation should be considered as a differential diagnosis in macaques with unilateral subcutaneous enlargements in the inguinal region, with ultrasonography as the 'gold standard' for diagnosis and herniorrhaphy as the treatment of choice.

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