Ex utero intrapartum correction of omphalocele in an English bulldog fetus

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Abstract

A fetal abdominal wall defect was diagnosed by ultrasonography during routine Cesarean section staging of a full-term English bulldog bitch. The omphalocele was reduced and repaired prior to delivery at the time of Cesarean section. The neonate recovered without incident. Throughout his first month of life, there was no significant difference in weight gain or clinical status compared to his littermates. However, the body wall defect recurred at ~4 weeks of age in the form of an umbilical hernia. At ~6 weeks of age, the patient was presented for perforation of omentum through the skin overlying the hernia and an umbilical herniorrhaphy was performed.

Keywords: Abdominal wall defect, omphalocele, prenatal ultrasonography, neonatal surgery

Introduction

Prenatal diagnosis of abdominal wall defects is common in human medicine, with potential to identify defects as early as 12 weeks of gestation.[1] A recent prospective study suggests that the rate of detection in women is ~95%, with most diagnoses before 20 weeks of gestation.[2] The practice of screening for congenital defects has not been widely described in veterinary literature, but represents an opportunity to prepare for immediate surgical correction at delivery. A similar case of prenatal diagnosis of omphalocele by ultrasonography has been described, which resulted in euthanasia of the neonate at delivery, due to the severity of presentation.[3] To the authors' knowledge, the current case represented the first reported prenatal diagnosis of omphalocele in a dog subsequently corrected at the time of Cesarean section (C-section). Surgical correction at delivery reduces the risk of abdominal sepsis and compromise of herniated organs, while maintaining fetal oxygenation through the umbilical vasculature. Since the fetus had a continued vascular connection to the dam, we expected the fetus to remain anesthetized, eliminating the need for a separate anesthetic event in the neonatal patient.

Case presentation

A 4-year-old, 26-kg multiparous English bulldog bitch was presented to the Veterinary Teaching Hospital at North Carolina State University for timed C-section staging at 60 days after LH surge. Ovulation timing and insemination had been performed at the same institution, with LH surge estimated from serial serum progesterone concentrations and used as Day 0 of gestation. On routine ultrasonographic evaluation of fetuses to determine maturity, a bulge of mixed echodense contents was visualized in the region anterior and superior to the umbilicus in a single fetus (Figure 1, A-C). All examinations were done with a 4 to 8 MHz micro-curved array transducer (Fujifilm SonoSite S9, Universal Medical, Bothell, WA). On the basis of the ultrasonographic findings, a presumptive diagnosis of congenital abdominal wall defect was made. Color flow Doppler was used to characterize the vascular supply to the herniated tissue, which was determined to be separate from the umbilical vasculature and urachus (Figure 1, D).

Treatment

The bitch was staged for C-section through routine diagnostics, including serial serum progesterone concentrations and ultrasound examinations of fetuses to gauge fetal maturity and monitor for indications of fetal stress. The dam was examined daily from Days 60 to 65 (day of surgery). The bitch's serum progesterone concentration at Day 64 was 4.8 ng/ml, with fair fetal maturation as evidenced by ultrasonographic appearance of kidneys and gastrointestinal tracts. At Day 65, serum progesterone concentration was not measured due to lack of laboratory availability. The decision to proceed with C-

section was made based on ultrasonographic maturity of the fetuses (significantly improved gastrointestinal peristalsis from the previous day). The presence of milk expressible from all teats in the bitch provided additional support for this decision. The bitch was routinely prepared for surgery, including cephalic intravenous catheter placement and administration of methylprednisolone (250 mg IV; Solu-Medrol®; Pfizer, New York, NY) with the goal of improving fetal lung maturity.

Following pre-oxygenation by facemask, anesthesia of the bitch was induced with propofol (140 mg IV). The bitch was intubated and anesthesia maintained with desflurane in oxygen. Ventilation was continually assisted following intubation until all puppies (six) were delivered. Anesthetic monitoring included heart rate, respiratory rate, peripheral oxygen saturation, blood pressure and capnography, all of which were maintained within normal ranges. The bitch received a single dose of intraoperative cefazolin (580 mg IV) before delivery, as well as buprenorphine (0.5 mg IV) immediately after all puppies were delivered.

A standard bilateral hysterotomy was performed; one surgeon removed the unaffected neonates from the right uterine horn and uterine body while a second surgeon removed neonates from the cranial aspect of the left uterine horn until the affected neonate was identified in the caudal aspect of the left uterine horn. The amnion was removed from the affected neonate and the umbilical vasculature left intact during the procedure. Dynamic herniation of small intestine through the defect was held in reduction with a cotton tip applicator while herniorrhaphy was performed. The hernia sac was excised and the external rectus fascia freshened using tenotomy scissors. The external rectus sheath was closed in a simple continuous pattern using 3-0 polydioxanone suture (PDS®II, Ethicon, Bridgewater, NJ). Subcutaneous and skin layers were closed in a single layer using 4-0 nylon (Ethilon®, Ethicon) in a simple continuous pattern. Both layers were closed leaving an ~5 mm defect at the caudal aspect of the original defect to prevent premature occlusion of umbilical vasculature (Figure 2). After closure of the defect, the umbilical cord was clamped and sharply transected ~2 cm distal to the umbilicus. The neonate had spontaneous respiration by completion of the abdominal wall repair and resuscitation was successful using standard techniques, including vigorous manual stimulation with a dry towel and external heat (lamp). Following resuscitation, buprenorphine (0.005 mg SC) was administered to the male, 445 g neonate. The goal of the surgery was to enclose and protect abdominal contents until the patient was mature enough to have a permanent umbilical hernia repair.

Following delivery of the last neonate, the bitch underwent an elective ovariohysterectomy. The external rectus fascia was infiltrated with bupivacaine (45 mg) at closure. The interval from induction of anesthesia to start of ventral celiotomy was ~9 minutes, interval from induction of anesthesia to start of omphalocele correction was ~15 minutes, total surgical time was 58 minutes, and total anesthetic time from induction to extubation was 83 minutes.

Neonates were united with their dam following her recovery from anesthesia. The affected neonate was observed to nurse without incident, with no appreciable deficits compared to littermates. The litter was evaluated for congenital defects: no further defects were identified. Each puppy received 3 mL frozen-thawed canine plasma delivered by orogastric intubation (standard practice at performing hospital). The bitch and puppies were discharged on the day of surgery following confirmation that the bitch had fully recovered from anesthesia and all puppies were observed to nurse.

Outcome

Over the following days, communication was maintained with the owner regarding the neonate's clinical status. According to the owner, he had increased vocalization intermittently during the first 24 hours after discharge, but maintained an appetite and activity level similar to littermates. On Day 12 following discharge, the puppy was presented for a re-check examination and suture removal. The puppy was bright, alert, and responsive with vital parameters within normal limits. The incision was clean, dry, and intact with a thin eschar present overlying the surgical site (Figure 3). The sutures had migrated from the wound edges and were removed.

On Day 30, the puppy was presented for recheck evaluation after the owner noted a soft protrusion in the region of the umbilicus. The puppy was otherwise doing well at home. On examination,

a reducible umbilical hernia ~3 cm in width and 4 cm in length was diagnosed (Figure 4). Given the age of the puppy and the risk that immediate surgical correction could result in an increased likelihood of herniation as the puppy continued to grow, it was decided to delay surgical correction of the abdominal wall defect until the puppy was more mature. The patient was discharged with instructions to monitor carefully for any change in the character of the hernia or the patient's clinical status.

Sixteen days after the initial recurrence was evaluated (at 46 days old, weight 4.4 kg), the patient was presented on emergency for perforation of omentum through the abdominal skin over the site of the umbilical hernia (Figure 5). Emergency surgery to correct the hernia was elected. The puppy was sedated with fentanyl (13 µg IV) and anesthesia was induced with midazolam (1 mg IV), propofol (4 mg IV), and ketamine (8 mg IV). The puppy was intubated with a 4.5 mm endotracheal tube and anesthesia was maintained with isoflurane in oxygen and fentanyl administered as a constant rate infusion (CRI). Anesthesia monitoring included heart rate, respiratory rate, blood pressure, capnography, and temperature. Blood glucose concentrations were serially evaluated and remained > 150 g/dL. Cefazolin (100 mg IV) was administered immediately before surgery. A ventral midline celiotomy was performed and a 2 cm portion of devitalized omentum (observed externally) was excised using ligation and electrocautery. The edges of the abdominal wall defect (5 x 3 cm) were debrided and closed using 2-0 PDS in a simple continuous pattern. The subcutaneous tissue was closed in a simple continuous pattern using 3-0 PDS and the subcuticular layers closed in a continuous horizontal mattress pattern using 4-0 poliglecaprone suture (Monocryl®, Ethicon), with skin edges reinforced using tissue glue.

Recovery from anesthesia was uneventful. The fentanyl CRI was continued in the postoperative period for ~36 hours (3 µg/kg/hour, decreased to 1.5 µg/kg/hour prior to discontinuation). Antibiotic therapy was provided in the form of ampicillin-sulbactam (97 mg IV every 8 hours) while hospitalized. All medications were discontinued at time of discharge after 2 days of postoperative hospitalized monitoring.

Communication was maintained with the owner and at time of writing this case report, the patient is nearly 10 months old and thriving, with no recurrence of the umbilical hernia (Figure 6).

Discussion

Abdominal wall defects are commonly encountered in veterinary practice, with varying incidence across species. In mammalian embryology, a physiologic herniation of the bowel through the umbilicus occurs in early gestation. In dogs, this resolves by 40 days of gestation.[5] Failure of resolution results in omphalocele, as occurred in the present case. The incidence of congenital abdominal wall defects in canines was 0.6% in > 1600 puppies examined from a cohort of pet store puppies.[4] Given the hereditary nature of many umbilical hernias, this incidence is likely to vary by breed and mate selection and may actually be higher due to euthanasia of affected fetuses soon after delivery. Due to the prenatal diagnosis of an abdominal wall defect, this litter was destined for homes as pets, with no intention for puppies to be used as breeding animals. In a litter-bearing species such as the dog, pre-partum transabdominal ultrasound is limited in its ability to singularly scrutinize the entirety of every fetus; therefore, the risk of false negative findings of canine fetal defects exists when using this modality.

Surgical correction of umbilical herniation in dogs has been described, with favorable prognosis and excellent long-term outcome.[6] Umbilical hernias are covered by a layer of subcutaneous tissue and skin which provide protection from both evisceration and abdominal sepsis. In contrast, omphaloceles are a subset of umbilical hernias that lack skin coverage and are often significantly larger than umbilical hernias. A transparent membrane of amniotic tissue initially covers the herniated organs but is easily ruptured; thus, euthanasia of neonates with omphaloceles is often recommended at delivery. Surgical repair of omphaloceles in a litter of 5-day-old kittens was attempted but failed due to suture pull-out from the fascia of the abdominal wall.[7] In humans, the optimal timing of omphalocele repair remains undetermined, with current practice involving immediate correction at birth, stepwise correction throughout the neonatal period, or delayed closure after epithelialization overlying the defect has occurred.[8] Omphalocele repair at the time of C-section in dogs may allow for survival of canine neonates that would otherwise perish due to abdominal sepsis and/or trauma to abdominal organs. Maintenance of

oxygenation and anesthesia via the umbilical vessels during herniorrhaphy at the time of C-section obviates both the need for establishing an airway in the neonate and a separate high-risk anesthetic episode. Consequently, this case has further implications for salvage of endangered species, with correctable congenital defects, that would otherwise be euthanized at the time of delivery.

Traditional repair of ventral midline hernias requires tension-free closure of the defect using non-absorbable or slowly absorbable suture in the external rectus fascia, followed by separate closure of subcutaneous and cutaneous tissues. In the present case, separate closure of the subcutaneous tissue was not feasible, due to the small size of the patient. Tension on the repair and subsequent suture pull-through from the fascial layer is a common cause of herniorrhaphy failure. Tension can be mitigated by using synthetic materials or tension-relieving sutures. The use of synthetic materials in this patient was not feasible for several reasons, including the patient's size and growth potential, surgical time and precision required to apply surgical mesh and potential for contamination of the synthetic material, predisposing to infection. A simple continuous pattern was chosen for both layers in an effort to minimize surgical and anesthetic time for both neonate and dam and because precision of suture placement was limited due to movement of the neonate during suture placement. No tension on the herniorrhaphy was noted at the initial surgical repair.

During omphalocele correction, the affected neonate displayed spontaneous movement, which made surgical repair challenging. Concurrently, the dam was assessed as appropriately anesthetized. Since omphalocele correction occurred ~15 minutes after induction, it would be expected that plasma propofol concentrations would be low or nonexistent. Therefore, the dam and the neonate should have been anesthetized primarily by desflurane. The most likely cause of neonate movement was pain/surgical stimulation as the neonate moved, primarily in response to surgical stimulation. Why the dam appeared properly anesthetized while the neonate appeared not to be was unknown and unexpected. Possible explanations for differences in either pain response or pain threshold include: differing inhalant concentrations in the dam versus the neonate (gas anesthetics move via pressure gradient from dam to neonate), an immature pain pathway in the neonate, and/or progesterone in the dam acting as an analgesic. Fetal activity as monitored through electrocorticogram and muscle activity involves interruptions in the fetal sleeplike state secondary to a decrease in external (or cutaneous) temperature, but not core body temperature, which would have occurred at the time of exteriorization from the uterus.[9] This physiologic response could provide additional explanation for movement during omphalocele repair. Furthermore, development of descending inhibition in the spinal cord is not fully developed until after 1 week of age in the neonatal rat, leading to an exaggerated neural response secondary to skin stimulation.[10] In the current case, the response of the neonate could be attributed to this lack of inhibition and would explain the difference in reaction between the two patients (dam and neonate).

Although many dams undergoing C-section receive opioids for pain from the celiotomy, these drugs are often withheld until all puppies have been severed from the maternal blood supply to prevent neonates from having unnecessary adverse side effects (e.g. sedation, hypoventilation and decreased appetite). Standard of care at our teaching hospital is to administer opioids after all puppies are delivered. In retrospect, it may have been beneficial to administer a short-acting opioid, e.g. fentanyl, to the dam immediately before surgery on the neonate, or increase the desflurane concentration (or both), to assure adequate anesthesia to the neonate.[11] In addition to humane implications, movement made surgical correction more difficult and therefore should prevented. Administration of buprenorphine after resuscitation did not alter ability to nurse or ventilate as compared to littermates. The owner's description of increased vocalization during the first 24 hours of life was likely due to pain from the surgical repair. Consideration of persistent pain should be included following this type of repair.

Although the herniorrhaphy supported the neonate for approximately the first month of life, long-term success of omphalocele repair may be improved with increased analgesia and placement of interrupted, wide-biting mattress sutures in the external rectus fascia. Due to the rapid rate of neonatal growth, however, owners should be prepared for the potential need for revision surgery. This was apparently the first report of repair of omphalocele in a dog at the time of C-section. Ultrasonographic prenatal diagnosis of a large ventral midline defect allowed for necessary surgical and anesthetic planning

to maximize likelihood of successful repair. Herniorrhaphy of an omphalocele at the time of C-section may improve survival of affected neonates.

Learning points

- Prenatal diagnosis of abdominal wall defects is possible with ultrasonography.
- Knowledge of a congenital defect may allow for surgical repair of body wall defects at the time of C-section.
- Analgesia of neonate during surgery and following delivery may facilitate herniorrhaphy.

Conflict of interest

The authors have no conflicts of interest or sources of funding to disclose.

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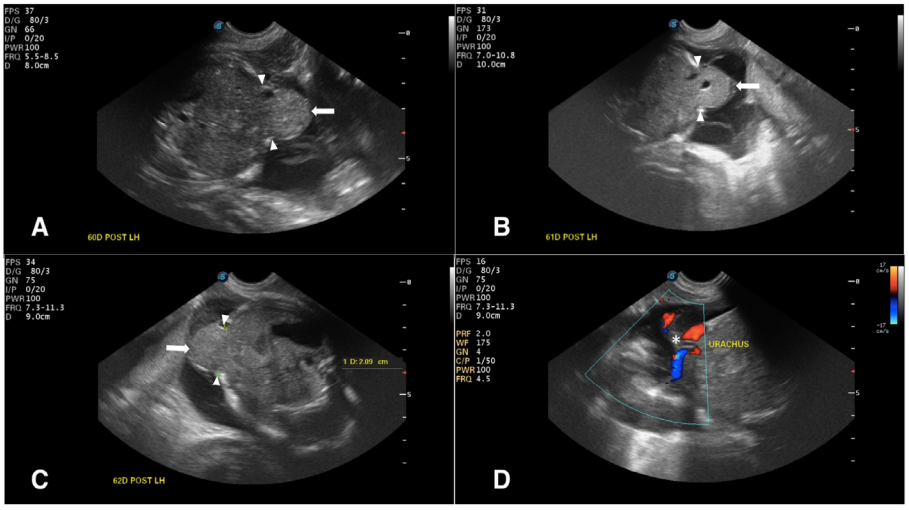

Figure 1 (A-D): Transverse ultrasound images of fetal abdominal wall defect obtained during C-section staging. Panels A-C demonstrate appearance of protruding contents (arrows). Arrow heads indicate body wall margins; distance between arrow heads approximately 2 cm. Panel D illustrates color flow Doppler to identify umbilical cord vasculature and urachus (asterisk), which were separate from the defect.

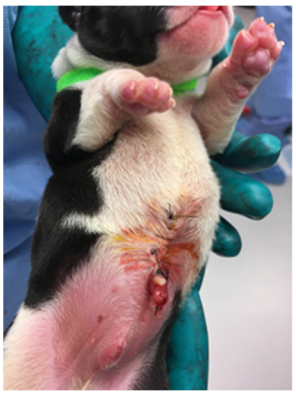

Figure 2. Closure of omphalocele at time of delivery. An approximately 5 mm opening was left at the caudal aspect of the original omphalocele to allow continued patency of the umbilical vasculature while repair was performed.

Figure 3. Healing of incision at age 12 days.

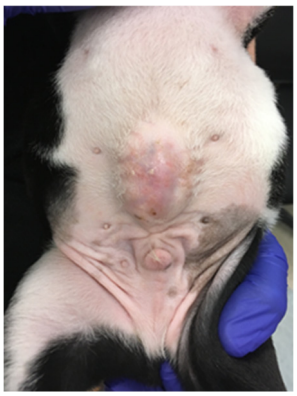

Figure 4. Herniation at site of surgical correction, age 30 days. Hernia contents were non-painful and reducible on examination.

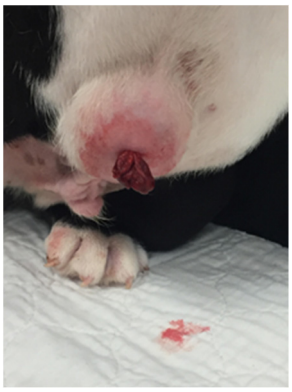

Figure 5. Protrusion of omentum through umbilical hernia, age 46 days.

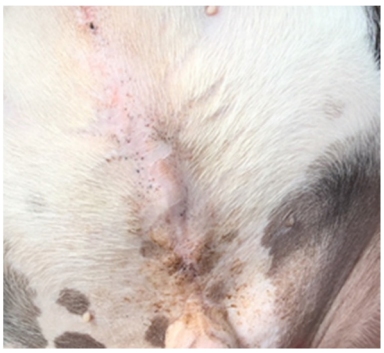

Figure 6. Healed incision at age 114 days.

(Editor's note: Online edition of the manuscript has color photographs)