

Case Report

Amorphus globosus

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Abstract

Amorphus globosus is a rare malformation that is defined as an acardiac asymmetrical rough spherical mass of tissue covered in skin. To authors' knowledge, amorphus globosus has not been previously reported in canids. Herein, we describe amorphus globosus in an American Bulldog pup with accompanying radiograph, gross images, and histopathology. Additionally, a systematic review (preferred reporting items for systematic reviews and meta-analyses protocol) was performed for veterinary species; amorphus globosus has been reported in 22 bovids, 3 caprids, 1 ovid, and 1 equid.

Keywords: Canine, dystocia, amorphus globosus, acardiac monster

Background

Fetal monsters are a rare occurrence in all species that may result in complications during pregnancy and parturition (e.g. dystocia). Amorphus globosus¹⁻⁵ is a type of acardiac fetal monster that is an asymmetrical rough spherical mass of tissue covered in skin. These monsters are associated with a cotwin and are considered a parasite twin, as the amorphus globosus individual's blood supply relies solely on normal cotwin circulation.⁶

Case presentation

A 2.5-year, female intact American Bulldog, was presented at the university emergency service with the primary complaint of dystocia. The dog had already delivered 4 healthy pups during a 9-hour period; no active labor was observed for 3 hours prior to presentation. Duration of pregnancy was unknown; however, pups appeared normal and were nursing well. Seven months before, the dog had dystocia and delivered live pups.

Appropriate mammary gland development with normal milk production was observed and abdominal distention with a brown vaginal discharge was noted. Fetuses could be palpated within the abdomen. An abdominal and thoracic focused assessment revealed 2 fetuses with heart rates of 184 and 160 beats/minute, respectively. Abdominal radiographs revealed 5 fetuses. Bloodwork revealed normal ionized calcium and glucose concentrations.

Treatment

Cesarean surgery was performed, and 5 additional viable male pups and 1 nonviable pup of an unknown sex were removed. An uncomplicated ovariohysterectomy was performed before closure and recovery. All 5 viable pups were successfully resuscitated. The nonviable pup was within a grossly normal amniotic sac and placenta; the pup was examined and there were no appendages, head, or obvious body orifices noted. This pup was placed in 10% neutral-buffered formalin and submitted for postmortem examination and histopathology.

Outcome

The dog recovered uneventfully from anesthesia and surgery, and was discharged from the hospital with 5 healthy, nursing male pups along with 4 healthy pups (2 male, 2 female) that were delivered before presentation.

The nonviable pup weighed 67 grams and measured 6 x 4.5 x 3 cm. It was composed of a portion of brown to tan haired skin that measured 4.6 x 3.2 x 2 cm and a cystic portion that measured 5.5 x 3.7 x 2.1 cm. There was a remnant ear with an adjacent indentation interpreted as a remnant orbit. There was also a nearby indented puckered area that resembled a malformed nose and a 0.5 cm long tail-like structure with a nearby indentation that resembled an imperforate anus. There were no discernible limbs (Figure 1). Sectioning revealed no discernible consistent anatomy, much of the tissue was

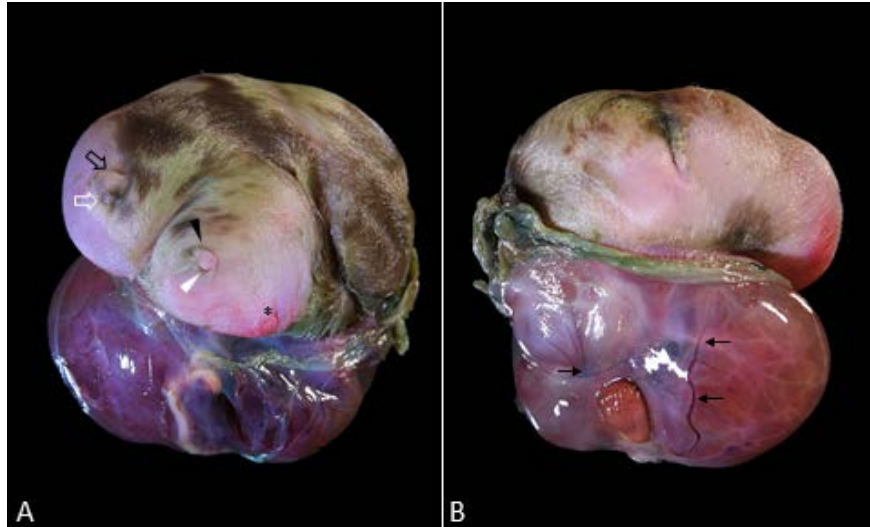


Figure 1. Gross images of the nonviable pup; A. note structures reminiscent of an ear (black arrowhead), closed orbit (white arrowhead), nose (asterisk), tail (open black arrow), and imperforate anus (open white arrow); B. note prominent irregular vasculature in the cystic portion (arrows).

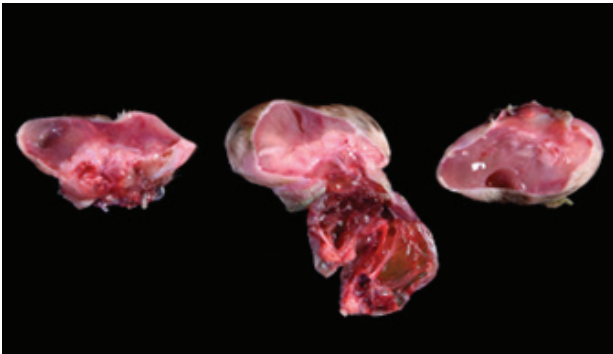


Figure 2. Cut sections of nonviable pup; note the amorphous cystic fluid-filled structures and edematous tissue without heart.

gelatinous, and there was no heart (Figure 2). Sections were taken from what grossly appeared as remnants/portions of the gastrointestinal tract, a large vessel, and fetal bone, and these were submitted for routine histopathology. The remaining tissue was then sutured back together for preservation as a teaching specimen. A radiograph identified a mass of tissue containing irregular radiopaque fragments of partially formed bony structures and multifocal variably sized radiolucent fluid-filled spaces (Figure 3).

On histopathological examination, the tissue had multifocal luminal structures lined by short villi with an organized layer of epithelial cells that multifocally contained finely stippled large clear cytoplasmic vacuoles resembling goblet cells. Submucosa of these luminal structures was filled with mixed inflammatory cells with a preponderance of lymphocytes and plasma cells. There were also scattered variably sized vessels within the submucosa and muscularis. Muscularis layers had luminal structure and were variably sized (Figure 4A). Surrounding tissue contained edema and variably sized vascular structures that occasionally coalesced to form irregular amorphous channels with multifocal aggregates of reticular fibers reminiscent of

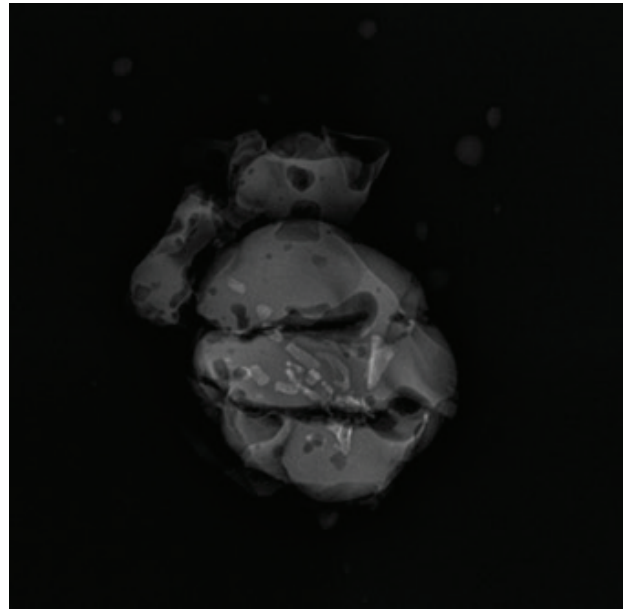


Figure 3. Radiograph of nonviable pup after sectioned for histopathology; note multifocal irregular bony fragments and multifocal fluid-filled spaces. Horizontal defects are artifacts from sectioning for histopathology.

splenic parenchyma. Tissue also contained scattered mixed inflammatory cells, variably sized nerve bundles, and multifocal islands of variably vacuolated polygonal cells organized into cords reminiscent of hepatic cords. There was also a much larger section of these polygonal cells that formed a rudimentary liver including haphazardly arranged portal and central regions and sinusoid-like structures (Figure 4B). Focally, cartilaginous fragments surrounded a luminal space lined by cuboidal to columnar ciliated epithelium reminiscent of a trachea. In another area, bone with marrow elements and nearby large bundles of nerve cell bodies were observed (Figure 4C). Adjacent to this was

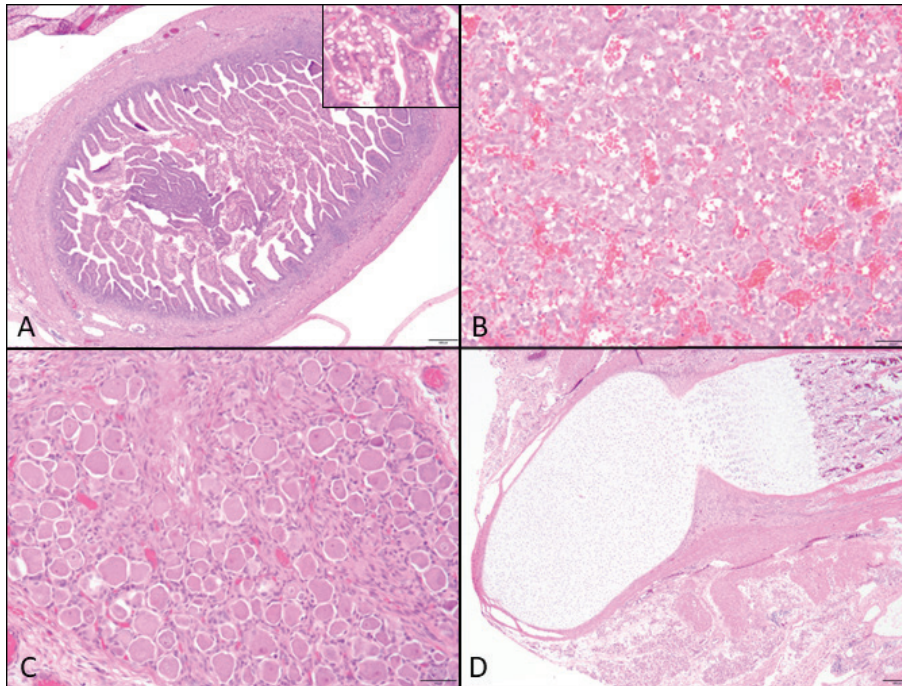


Figure 4. Histopathology (H&E): A. luminal structure resembling intestines with villous mucosa, submucosa, and distinct muscularis layers including mixed inflammatory cells and vessels, 4 x. Inset: prominent goblet cells in villi, 40 x. B. large expanse of tissue resembles liver with hepatocytes forming irregular cords with interspersing sinusoid-like blood channels, 20 x. C. organized bundle of nerve cell bodies, 20 x. D. remnant of organized pale basophilic cartilage with appropriate maturation to bone and marrow resembles a neonatal growth plate; note structure surrounded by amorphous edematous tissue with admixed blood cells, 4 x.

striated skeletal muscle and a section of bone undergoing bony maturation (Figure 4D).

Discussion

To assess the occurrence of this malformation in veterinary literature, a systematic review of case reports published prior to September 30, 2023 was performed. The framework as highlighted in 'preferred reporting items for systematic review and meta-analysis protocols (PRISMA-P)' was used.⁷ PubMed and the Mississippi State University Library system (<http://library.msstate.edu/>) were searched using terms 'amorphus globosus' and 'amorphous globosus' that yielded 102 results. With duplications and non-English papers removed, this left a total of 20 publications. Publications were assessed and compiled (Table). A systematic review identified reports in the following species: 22 bovines, 3 caprines, 1 ovine, and 1 equine (Table). To author's knowledge based on this review, there are no current reports of amorphus globosus in canids.

Amorphus globosus most commonly occurs in bovines, with the most common occurrence in Holsteins (~ 1 in every 3,500 pregnancies).²³ In humans, it occurs in 1 out of every 35,000 births.⁹ The most commonly accepted theory for the cause of this malformation is twin reverse arterial perfusion sequence (TRAP). In TRAP, there is a countercurrent flow of deoxygenated blood from the umbilical cord arteries of the cotwin and the amorphus globosus individual with return along the umbilical cord vein back to the placenta. This increases the cardiac demand on the cotwin.¹⁰ However, most cotwins in veterinary species die due to dystocia or dam-related disease

from multiple fetal burden rather than heart failure.^{10,17} Anastomosis of arteries or veins was lacking in one report of malformed fetus;²² it was speculated in that instance that insufficient blood supply led to complete destruction of a past existing heart structure.²²

Amorphus globosus has reportedly never been observed as a single birth.¹⁴ However, there is a single case report in a buffalo that presented in dystocia with an amorphus globosus structure and no cotwin.²⁰ Whether a cotwin was born out of sight or this entity can truly occur without a cotwin, is uncertain. There are reports of both monozygotic and dizygotic twins.⁶ Interestingly, 2 freemartin cotwins have been reported in cattle.^{11,17} In these cases, the amorphus globosus individuals contained identifiable testicular tissue on histopathology. In another case, a normal heifer cotwin was identified with a male amorphus globosus.¹³

Affected dams of all species are usually multiparous, with only 2 reported cases of uniparity in a Japanese black heifer and a Buffalo heifer.^{16,20} There is no sex predilection for amorphus globosus individuals or the associated cotwins, although there may be a trend towards male cotwins according to this review. There is no evidence to support culling or spaying of affected dams, as there are no reports of this condition occurring more than once in the same dam.

Future research can focus on pregnancy management of identified amorphus globosus dogs including early identification and treatment. Another avenue to explore is the long-term effects on surviving cotwins.

Table. Summary of published amorphus globosus cases in animal species

| Species | Breeds reported (no. animals) | Sex ratio of ag (F:M) | Twin birth status (no. animals) | Sex ratio of twins(F:M) | Complications (no. animals) | Refs. |
|------------------------------------|--|---------------------------|-----------------------------------|---------------------------|---|----------------------------------|
| Bovine (<i>Bovidae</i> family) | Holstein (10); unspecified (5); buffalo (2); Ayrshire (1); Brown Swiss (1); Japanese Black (1); Jersey-Dutch (1); White Galloway (1) | 2:3 (plus 18 unspecified) | Live (13); stillborn (12); NR (3) | 3:4 (plus 13 unspecified) | Dystocia (5); none (6); NR (4); retained placenta (3); death (2); peri-parturient paralysis (1) | 2,3,5,6,9, 10-14, 16-18,20,21-23 |
| Caprine (<i>Capra</i> sp.) | Unspecified (2); Kani-adu (1) | 3 unspecified | Stillborn (4); live (2) | 6 unspecified | Dystocia (3) | 1,15,19 |
| Equine (<i>Equus caballus</i>) | Unspecified (1) | 1 unspecified | Live (1) | 1 unspecified | None (1) | 4 |
| Ovine (<i>Ovis</i> sp.) | Corriedale (1) | 1 unspecified | Live (2) | 0:2 | NR (1) | 8 |
| Canine (<i>Canis familiaris</i>) | American Bulldog (1) | 1 unspecified | Live (9) | 2:7 | Dystocia (1) | |

F = female; M = male; NR = not reported; ag = amorphus globosus.

Learning points

- Amorphus globosus is defined as an acardiac asymmetrical roughly spherical mass of tissue covered in skin
- This disorder may increase the incidence of birth complications, most commonly dystocia and retention of fetal membranes
- This malformation has the potential to cause cotwin mortality due to dystocia and potentially cardiac compromise
- Early identification and treatment of amorphus globosus individuals may decrease cotwin mortality and birth complications
- There is no evidence to support culling or spaying of affected dams, as there are no reports of this condition occurring more than once in the same dam

Conflict of interest

None to report.

Acknowledgement

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