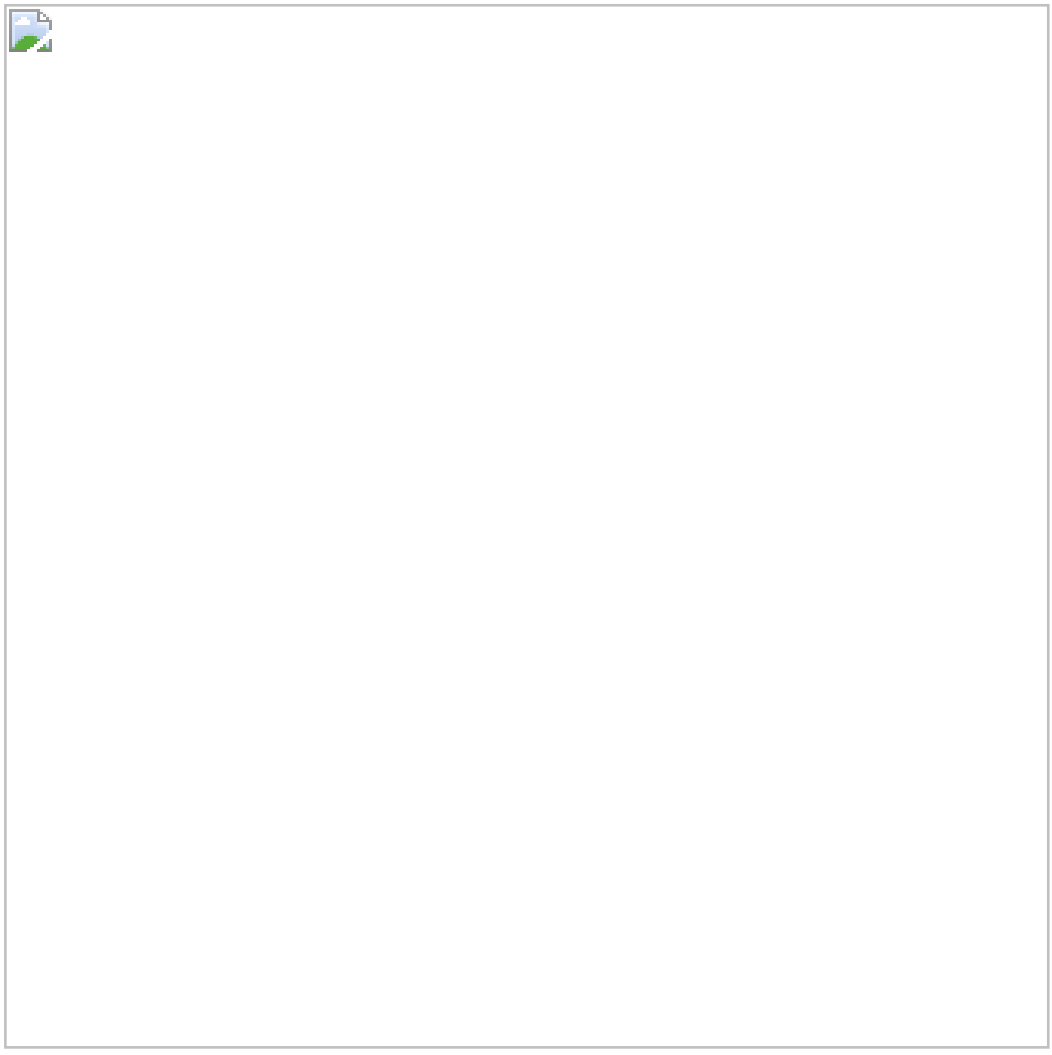

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Source: Journal of Feline Medicine and Surgery Open Reports, 10(2)

Published By: SAGE Publishing

URL: https://doi.org/10.1177/20551169241261582

Case Report

jfms OPEN REPORTS

Journal of Feline Medicine and Surgery Open Reports
1–6

DOI: 10.1177/20551169241261582
journals.sagepub.com/home/jfmsopenreports

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Successful surgical separation of asymmetrical caudal duplication or parasitic twinning in a kitten

Rachel Botsoglou[1], Rebecca Albert[2] and Maximiljan Krauß[1]

Abstract

Case summary A female kitten with two ectopic supernumerary pelvic limbs arising from the umbilicus was born in our clinic. Five weeks after the kitten's birth, she showed an acute deterioration in condition, with clinical signs of an ileus in the intestines entering the supernumerary body part. We then performed a surgical separation of the supernumerary limbs and intestines. Based on clinical findings and radiography, the kitten was classified as a case of parasitic twinning-omphalopagus or a subtype of asymmetrical caudal duplication, rather than a case of polymelia. Parasitic twinning and caudal duplication are rare congenital malformations in many animal species and in humans, which can often have a poor outcome. Almost 1 year after the separation, the cat appeared to be living a normal life without complications.

Relevance and novel information A good outcome is rare in parasitic twinning because many of these animals present with other concurrent health issues, such as congenital heart malformation or cleft palate, or they are stillborn. However, this pathological entity is scarce in small animal medicine, and there is no documented survival rate. To our knowledge, this is the first officially documented case of parasitic twinning-omphalopagus in a cat in the past decade.

Keywords: Parasitic twins; caudal duplication; omphalopagus; heteropagus

Accepted: 22 May 2024

Introduction

Caudal duplication, or dipygus, is a duplication of various organs in the caudal region of the body, including the distal spine, urinary tract, genitalia and distal intestine, presumably due to a division of the spinal cord at its distal part during embryogenesis. This syndrome is rare in people[1] and animals and the embryologic basis is unclear.[2–4] In the present case, there was no duplication of the spine, genitalia or bladder, thus resembling parasitic twinning. Parasitic twinning, or heteropagus, is a subtype of incomplete conjoined twinning. Conjoined twins can be symmetrical or asymmetrical. In the latter case, one fetus is not fully developed, which means that it does not have all its organs, so it shares some of them with its twin and its vital functions depend on it. The twin that is not fully developed is called the parasite, while the fully developed twin is called dominant or the autosite.[5] That is called parasitic twinning. The reason for this attachment is unknown. There are two theories: fission and fusion. According to the fission theory, the embryonic axis is not completely split in the first days of pregnancy, whereas in the fusion theory, two distinct cell masses merge at a later stage of pregnancy.[2,5–8] This second theory is no longer believed to be the basis of conjoined twinning.[7,8] In both cases, the fetus is almost exclusively

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Figure 1 The kitten after the birth in (a) dorsal and (b) lateral recumbency.
Note the two supernumerary hindlimbs arising from the abdomen

monozygotic with a monoamniotic, monochorionic type of placenta. Vascular compromise of the parasite is thought to be the reason for the asymmetrical development between the autosite and the parasite, leading to a relationship of dependency on the collateral circulation between the twins. As a result of this circulatory compromise, there is ischaemic atrophy of the parasite.[2,6,7,9] There are many types of parasitic and conjoined twins, which are typed according to the site of the conjunction.[7,10,11] Thoracoomphalopagus and thoracopagus are thought to be the most common ones.[11] In the present case, according to the attachment point, the parasitic twin was classified as omphalopagus/epigastric heteropagus, from the ancient Greek words *omphalos*, the term for umbilicus, and *pagus*, meaning fixed or joined. Parasitic twinning is a rare condition, with an incidence of <0.1 in 100,000 births in human medicine.[5–7,12] In veterinary medicine, there are a few published case reports and case series.[2,7,8,10,13–31] To our knowledge, there is only one case series[26] and a few case reports [17–22,27,29] on cats with different types of conjoined twins, mostly symmetric, the majority of which had a bad outcome. The aim of this case report was to summarise the current literature and to enrich it with a new and interesting case of an omphalopagus-parasitic twin in a cat, with a favourable outcome.[17–22, 26, 27]

Case description

A kitten with supernumerary limbs was delivered in our clinic via caesarean section because of dystocia in an 11-month-old queen, together with three other healthy kittens. The kitten was born with two supernumerary, non-functional pelvic limbs originating from the umbilical area. The supernumerary limbs were almost the same size as the main pelvic limbs. Behind the ectopic pair of limbs, there was a rudimentary anal opening, and neither urethra nor external genitalia were present. There were no other malformations. The main limbs were well-developed and fully functional, and the main body had

a normally shaped anogenital area with female genitalia (Figure 1). The queen was free from health issues. There were no other similar incidents in the family history, but the parents were siblings. The kitten was stable (heart rate 220/min, respiratory rate 24/min) and discharged from our clinic 4 h after the birth with a recommendation for surgical separation of the supernumerary limbs a few months later once the kitten was adequately mature, to minimise the risks of such a procedure. The kitten showed a normal growth rate, with good food intake and body score at its second checkup in our clinic at the age of 1 month. However, in the fifth week, there was an acute deterioration of its general condition, with apathy, anorexia, swelling in the umbilical area and a smudgy faecal discharge around this anus-like opening on the supernumerary body part. The kitten was brought to our clinic as an emergency case. The physical examination showed a heart rate of 240/min and a respiratory rate of 44/min, without abnormalities on auscultation. The kitten's body temperature was 34.3°C and glucose was 1.3 mmol/l. The buccal mucosa were pale pink and dry, and the capillary refill time was longer than 2 s. The kitten's body weight was 450 g. The kitten was stabilised with an intravenous (IV) fluid (lactated Ringer's; B Braun) bolus of 5 ml in 15 mins and then 3 ml/h via catheterisation of the cephalic vein. A glucose bolus (Glucose 200 mg/ml; B Braun) of 2 ml/kg in 15 mins was given when indicated. The hypothermia was treated with a patient warming system (Bair Hugger; 3M). Methadone hydrochloride (Comfortan; Dechra) at 0.1 mg/kg IV was given for pain relief. As a result of the clinical instability of the patient, further diagnostic investigations were not performed and only plain radiographs were taken (Figure 2) so that the duration of anaesthesia and risks were kept as low as possible. For anaesthesia, methadone (Comfortan; Dechra) 0.2 mg/kg IV was used as premedication and propofol (Narcofol; CP Pharma) 7 mg/kg IV in total for induction. Inhalational anaesthesia with isoflurane (Isospire; Dechra) was given for maintenance. During anaesthesia, the blood glucose was controlled regularly and a 20% glucose (Glucose 200 mg/ml; B Braun) bolus was given when necessary. Antibiotic treatment with amoxicillin–clavulanic acid (AmoxClav; HEXAL) 20 mg/kg IV was used intraoperatively, as a prophylactic because of the poor general condition and the intestinal inflammation. The surgical separation was performed through an exploratory laparotomy (Figure 3). The median laparotomy was performed through an incision of the linea alba and an elliptical incision of the conjunction point around the umbilical area. There was no sign of free fluid in the abdominal cavity. During the exploration of the abdominal cavity, a duplication of the intestines at the transition point of the jejunum to ileum was detected. From the duplication point, there was a second ileum, cecum and colon along with the physiological anatomical intestinal structures. The second colon was leading through the conjunction point into the parasitic twin. The colon and rectum were dilated and filled with compressed stool, with the intestinal wall seeming extremely thin. The engorged rectum was leading to a not fully functional anus. There was no double genital or urinary system. In the abdominal cavity of the autosite, there was only the right kidney. The rest of the abdominal organs had no specific deformities. Distal to the attachment point, two extra hindlimbs were present. Resection of the double ileum, cecum, colon, and rectum at the point of the duplication was performed through enterectomy. The enterectomy site was closed with a simple interrupted suture pattern with polydioxanon (PDS II 4-0; ETHICON). The parasitic twin was separated at the umbilicus of the autosite (Figure 4). The abdominal wall and the subcutaneous tissue were sutured in a simple continuous pattern with poliglecaprone 25 (Monocryl PLUS 3-0, 4-0; ETHICON). A simple interrupted suture with polyamide 6 (Ethilon 4-0;

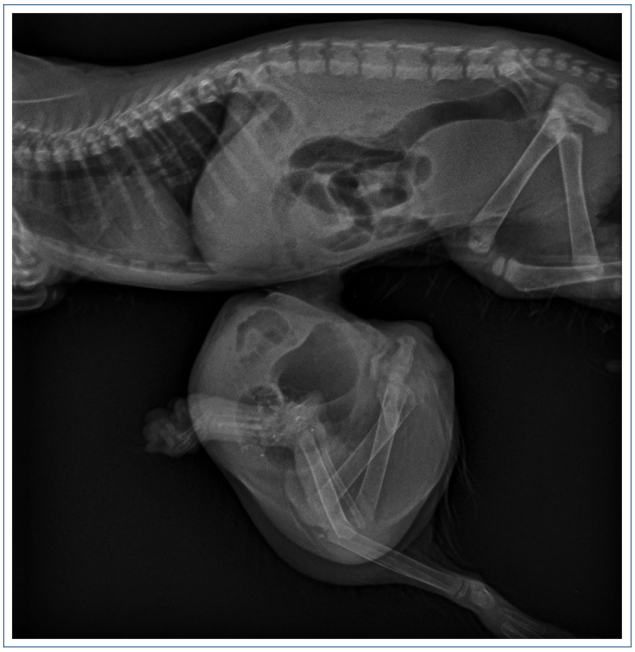

Figure 2 Radiograph of the kitten before the surgery. At the height of the umbilicus and directly associated with the abdominal wall of the main twin, there was a pair of accessory hindlimbs. They were symmetrical and fully formed. The accessory hindlimbs were connected through a fine soft tissue-like bridge to the abdominal wall. Proximally there was a second pelvis, two-thirds as long as the normal one. The long bones of the accessory limbs were almost the same in length, diameter and density as the normal ones. Only the soft tissues around them were slightly less developed. Notice the round tubular gas filled structures, thought to be intestines with sign of an ileus in the parasitic twin

Figure 3 Images of the kitten (a) before and (b) after the surgical separation through median laparotomy

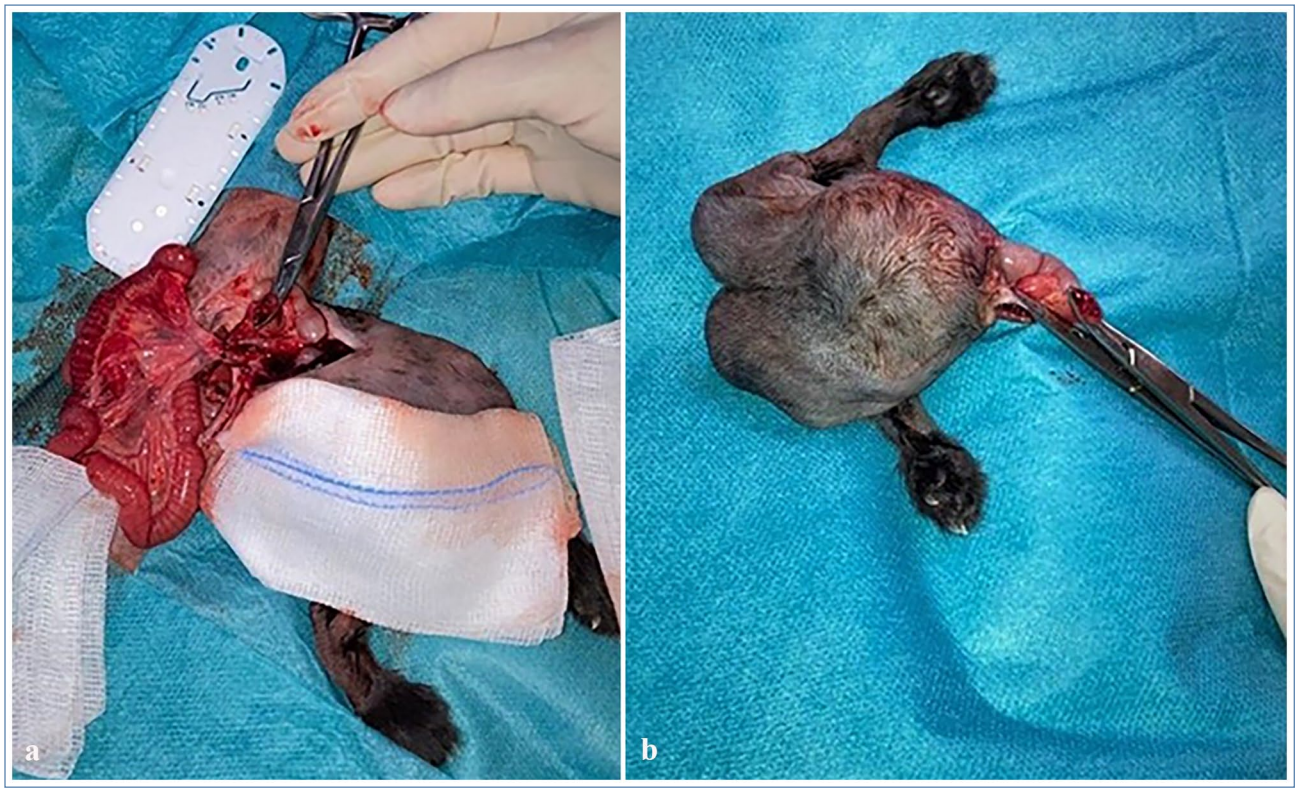

Figure 4 Internal configuration of (a) the supernumerary intestines and (b) hindlimbs. The clamp underlines the junction point

ETHICON) was used for the cutaneous tissue. The patient was stabilised postoperatively with IV fluids (Sterofundin VG-5 Plus; B Braun) at 2 ml/h and hourly blood glucose assessments, temperature maintenance with a patient warming system, pain management with buprenorphine q8h (Buprenovet Multidose; BAYER) for the first day postoperatively, followed by meloxicam q24h (Metacam; Boehringer Ingelheim) for the next 5 days. Amoxicillin–clavulanic acid (AmoxClav; HEXAL) was given at 20 mg/kg IV q8h for 5 days. The kitten was fed with commercial kitten milk and canned kitten wet food. There were no postoperative complications, and the patient showed

normal food consumption and normal urine output and defecation. The patient was discharged after 5 days.

Ten days postoperatively, the patient presented with good wound healing and physical condition at the follow-up in our clinic. Almost 6 months later, the kitten received a second follow-up in another clinic, where everything was within normal limits and no further malformations were found.

Discussion

Parasitic twins are joined to an otherwise relatively normal foetus. Parasites in the case of an omphalopagus usually consist of supernumerary limbs attached to the lower thoracic and upper abdominal wall, mostly not functional. These limbs are seldom not connected to viscera, but are rarely associated with a second functional heart or brain.[9] According to Spencer,[9] a possible explanation for this issue is that when the heart begins to function, a major portion of its output will be directed to the largest and most rapidly growing structure, the brain, which would not survive without a sufficient supply of blood. Consequently, both the heart and the brain are either non-functional or completely absent in all parasitic twins, where blood circulation is limited.[9] There are many factors that could contribute to the occurrence of parasitic or conjoined twinning.[7,17] Physical agents, such as lowered temperature,[17,32] ultraviolet light,[17,33] radiation[17,34] and lack of oxygen in the first stages of pregnancy, have been proposed to induce conjoined twins.[17,32] Chemical factors, such as prochlorpemazine,[17] chlorhydrate of *N*,*N*-dimethylbiguanidine,[17] thalidomide,[35] vitamin A, dimethyl sulfoxide, urethan,[36] vincristine sulfate[37] and parbendazole,[38] may also contribute to the occurrence of conjoined twins.[7,17] Treatment with selamectin in the first month of pregnancy was documented in one case report in a cat, without concrete proof of its relevance.[21] Genetic factors and oocyte ageing before fertilisation can also play a role.[7,17,38,39] In the present case, there was no known exposure to any of these substances.

A thorough diagnostic imaging investigation is recommended to determine the extent of the malformation.[5,12]

Surgical separation is important because there are not only the cosmetic grounds but health issues as well. For example, parasites can cause respiratory distress and may restrict the growth of autosites.[5] The right timing for diagnostic investigation and surgical separation is a topic of concern and controversy. Even in human medicine, there is no gold standard, and in cases where the patient is stable, the surgery is often delayed for a couple of months until the patient is more mature.[5] In case of unstable patients, a more direct intervention is usually applied. This decision depends on the individual patient and as in the present case, the circumstances can change spontaneously, making an emergency intervention crucial.[5]

Conclusions

Parasitic twinning and caudal duplication are rare conditions. If the parasitic twin or the duplication does not contain life-sustaining organs, and the main twin has no other congenital abnormalities, such as cleft palate or heart malformation, a surgical separation should always be considered a reasonable option, as the long-term outcome can be favourable for the autosite. To our knowledge, ours is the first case of parasitic twinning-omphalopagus or partial caudal duplication in a cat to be published in recent decades, and the only one that received surgical management and had a good outcome.

Conflict of interest The authors declared no potential conflicts of interest with respect to the research, authorship, and/or publication of this article.

Funding The authors received no financial support for the research, authorship, and/or publication of this article.

Ethical approval The work described in this manuscript involved the use of non-experimental (owned or unowned) animals. Established internationally recognised high standards ('best practice') of veterinary clinical care for the individual patient were always followed and/or this work involved the use of cadavers. Ethical approval from a committee was therefore not specifically required for publication in *JFMS Open Reports*. Although not required, where ethical approval was still obtained it is stated in the manuscript.

Informed consent Informed consent (verbal or written) was obtained from the owner or legal custodian of all animals(s) described in this work (experimental or non-experimental animals, including cadavers) for all procedure(s) undertaken (prospective or retrospective studies). For any animals or people individually identifiable within this publication, informed consent (verbal or written) for their use in the publication was obtained from the people involved.

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