CASE REPORT



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Duodenoduodenal intussusception in a 16-year-old German Warmblood mare

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Summary

A 16-year-old German Warmblood mare was presented with an acute history of abdominal pain that failed to respond to medical treatment. Diagnostic evaluations including transabdominal and transrectal ultrasound and palpation were suggestive of a smallintestinal intussusception. Exploratory laparotomy revealed a duodenoduodenal intussusception of the proximal and middle third of the duodenum. The horse was subjected to euthanasia due to the poor prognosis. The post-mortem examination showed the intussusception to be associated with a pseudodiverticulum orally to the intussusception and duodenal muscular hypertrophy.

KEYWORDS

horse, colic, diverticulum, duodenum, intussusception

INTRODUCTION

Intussusception is an invagination of a segment of bowel (intussusceptum) into an adjacent segment of bowel (intussuscipiens) and is considered an uncommon cause of colic in the mature horse (Edwards, 1986; Nelson & Brounts, 2012). It can lead to an interruption of the food passage as well as to venous infarction of the affected intestine resulting in colic (Edwards, 1986). Intussusceptions can occur in several segments of the gastrointestinal tract. Jejunojejunal, jejunoileal, ileoileal and ileocecal intussusceptions have been reported in the equine small intestine (Edwards, 1986; Nelson & Brounts, 2012). In large intestines, caecocaecal, caeocolic, colon or colorectal intussusceptions were described (Nelson & Brounts, 2012). Intussusceptions are believed to be caused by abnormal motility patterns because of intestinal neoplasia, foreign bodies, previous resection and anastomosis, parasitism, sudden dietary changes, enteritis, recent general anaesthesia or gastroduodenal ulcers (Haddad et al., 2021; Matsuda et al., 2013). Idiopathic

intussusceptions have also been described in horses (Sasani et al., 2013). Duodenoduodenal intussusceptions are extremely rare in humans (Yalamarthi & Smith, 2005) and, to the authors' best knowledge, have so far not been described in horses.

Case summary

A 16-year-old German Warmblood mare was presented to the Swiss Institute of Equine Medicine (ISME) Equine Clinic of the University of Bern for evaluation of acute onset of abdominal pain. Clinical signs had been present for 6h prior to admission and had not resolved with conservative treatment by the private veterinarian consisting of administration of Flunixin meglumine (1.1 mg/kg bodyweight [bwt] i.v.; Finadyne N, MSD Animal Health GmbH), metamizol (40 mg/kg bwt i.v.; Vetalgin N, MSD Animal Health GmbH) and Butorphanol (0.01 mg/kg btw i.v.; Morphasol -10, Dr E Graeub AG). The owner reported no previous history of colic or weight loss.

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Physical examination findings

Upon initial examination, the mare was depressed, with a heart rate of 40 beats/min and an elevated respiratory rate of 40 breaths/min and sweating. No further clinical signs of shock were noted. Abdominal gut sounds were reduced in all four quadrants. Nasogastric intubation revealed no net reflux but a moderately feed-filled stomach. At transrectal palpation a prominent, obstipated pelvic flexure running horizontally and cranial to the pelvis was detected. In addition, a mass of approximately 10–15 cm diameter was palpated cranial to the pelvis at the right craniodorsal aspect. The mass could not be delineated cranially. The horse passed a small amount of faeces during the clinical examination.

Diagnostic procedures and conservative treatment

Serum biochemical profile and complete blood count showed a reduced packed cell volume of 27% (reference values: 30%–47%) and total protein of 42 g/L (reference values: 52–78 g/L) with a lowered albumin (17 g/L) (reference values: 19–32 g/L). A neutrophilia of 9.0 10e9/L (reference values: 2.5–6.0 10e9/L) and an elevated urea (13.3 mmol/L) (reference values: 3.6–8.9 mmol/L) were present. The measured venous blood lactate was 2.4 mmol/L (reference values <2 mmol/L). The transabdominal ultrasound assessment (Busoni et al., 2011) (Mindray M7Vet with a C5-2s Convex ultrasound transducer, Shenzhen Mindray Bio-Medical Electronics Co.) was unremarkable at the initial examination. Clear, straw-coloured abdominal fluid was obtained by

abdominocentesis, with a lactate of 2.8 mmol/L (reference values: $<2 \, \text{mmol/L}$), a total protein of 1 g/L (reference values: $<25 \, \text{g/L}$) and a total nucleated cell count below the measurable limit (reference values: $<569 \, \text{cells/L}$).

An intravenous catheter (Extended use MILACATH 14 Ga \times 13 cm [5.25 in], MILA International Inc.) was aseptically placed. The horse received one dose of metamizol (40 mg/kg bwt i.v.; Vetalgin N, MSD Animal Health GmbH) and was placed on double maintenance intravenous fluids, a combination of Ringer's lactate solution and isotonic Equibiserol (Laboratorium Dr G Bichsel AG). In addition, two litres of plasma (PlasmaLife, II Ceppo GmH) were administered to address the hypoproteinaemia.

Four hours after starting the initial treatment, the mare began to show signs of colic and became tachycardic (60 beats/min), so transabdominal ultrasound and transrectal palpation were repeated. The pelvic flexure felt less impacted, but the right craniodorsal intra-abdominal mass was still evident. During transabdominal ultrasound examination, a segment of small intestine with a thickened and congested wall and a hyperechoic centre could be visualised in the right proximal third of the flank just caudal to the last rib. In addition, a transrectal ultrasound (Mindray M7Vet with a 6LE5Vs intrarectal veterinary ultrasound transducer, Shenzhen Mindray Bio-Medical Electronics Co.) of the palpated mass was performed, where multiple concentric small-intestinal wall layers were observed, creating the typical ultrasonographic concentric ring sign of an intussusception (Figure 1). Based on these findings, in particular the mass felt by transrectal palpation and the recurring signs of colic despite appropriate pain medication, an exploratory celiotomy was recommended.



FIGURE 1 Transrectal ultrasonographic image of the intussusception obtained at the right craniodorsal aspect of the abdomen. Note the concentric multi-layered appearance of the intestinal walls (ring sign). Hyperechoic mesenterial fat is invaginated with the intussusceptum within the intussuscipiens. The arrowheads show the lumen of the intussusceptum. The arrows point to the intestinal wall of the intussusceptum. The stars indicate separate mucosal layers of the intussuscipiens

EXPLORATORY CELIOTOMY

Within 1h prior to induction of general anaesthesia, the mare received preoperative anti-inflammatory drugs (Flunixin meglumine 1.1 mg/kg bwt i.v.; Finadyne N, MSD Animal Health GmbH) and antibiotics (Benzylpenicilline 30,000 IU/kg bwt i.v.; Penicillin Natrium Streuli, Streuli Tiergesundheit AG), (Gentamicine 6.6 mg/kg bwt i.v.; Pargenta-50, Dr E Graeub AG). General anaesthesia was induced using ketamine (2.5 mg/kg bwt i.v.; Ketasol-100, Dr E Graeub AG) and diazepam (0.05 mg/kg bwt. i.v.; Valium, Roche Pharma [Schweiz] AG). The horse was positioned in dorsal recumbency. Maintenance of anaesthesia was achieved with isoflurane and 100% of oxygen in a closed-circuit vaporiser system. Lidocaine (1.8 mg/kg/h bwt i.v.; Lidocaine 2% Streuli Tiergesundheit AG) and dobutamine (0.03-0.09 mg/kg/h bwt i.v.; Dobutrex, Teva Pharma AG) were added to the anaesthetic protocol as continuous rate infusions (CRI). The surgical site was aseptically prepared and draped in a standard fashion. A 20-cm preumbilical ventral midline incision was made, and the abdomen was carefully explored. The caecum and ileum were first identified and exteriorised followed by the jejunum. The most aboral part of the duodenum appeared to be thickened on palpation. Tracing the duodenum orally, a nonreducible intussusception of approximately 1-m length was identified in the middle part of the duodenum. The most aboral segment of the descending duodenum and the transverse part of the duodenum were intussuscepted into the ascending duodenum. The mesentery of the affected intestinal segment was highly haemorrhagic and oedematous. Manual reduction of the intussusception was not successful, because of the marked oedema of the intestinal wall. Multiple, focal haemorrhages and marked oedema were present in the visible part of the most oral jejunum, which completely lacked intestinal motility. As the affected part of the duodenum could not be exteriorised, controlled resection and anastomosis of the affected segment of duodenum was deemed technically impossible and the mare was subjected to euthanasia on the surgery table (Pentobarbital 90 mg/kg bwt i.v.; Esconarkon ad us. vet., Streuli Tiergesundheit AG).

Post-mortem findings

On post-mortem examination, the intussusception was located 50 cm aboral to the stomach and affected part of the descending duodenum as well as the transverse and part of the ascending duodenum (Figure 2). The intussusceptum was invaginated over a length of 100 cm into the intussuscipiens. The mucosa of the intussusceptum was severely thickened and diffusely oedematous and dark red, consistent with haemorrhagic venous infarction (Figure 3). Immediately oral to the intussusception, a diverticulum measuring 5 cm in diameter was found on the antimesenterial side of the intestinal wall (Figure 4). The tunica muscularis was moderately thickened in the area of the intussusception as well as up to 20 cm aboral to it. In addition, the serosal surface of the duodenum immediately aboral to the intussusception showed several multifocal, red and slightly

raised plaque-like lesions consistent with hemomelasma ilei. In the mucosal lining of the duodenum near the diverticulum, several multifocal ulcerations measuring up to 4 cm in diameter were present. Histological examination confirmed the diffuse oedematous thickening of the submucosa as well as the hypertrophy of the muscular layer of the small-intestinal wall. The diverticular wall was lined by mucosa, submucosa and serosa. The diverticular wall was lacking the tunica muscularis and was therefore classified as a pseudodiverticulum (Figure 5). The diverticular wall was infiltrated by moderate amounts of adipocytes and was moderately oedematous.

DISCUSSION

In horses and other equids, intestinal intussusceptions most frequently occur in foals and adolescent animals younger than 3 years of age and ileocecal intussusceptions seem to be the most prevalent type (Abraham et al., 2014; Edwards, 1986; Greet, 1992; Haddad et al., 2021; Matsuda et al., 2013; Nelson & Brounts, 2012). One study reported an apparently higher risk for ponies (Edwards, 1986). In adult humans, intussusceptions are considered rare, causing less than 5% of gastrointestinal obstructions. Similar to what is described for horses, 95% of all intussusceptions in people occur in children (Yalamarthi & Smith, 2005). In humans, duodenoduodenal intussusception is an extremely rare condition. The causes of duodenoduodenal intussusceptions in humans are usually changes in the intestinal wall of the duodenum or irritants within the lumen leading to altered motility (Pradhan et al., 2015). Ninety-five per cent of intussusceptions in children are idiopathic, whereas in adults, a cause for intussusceptions can be identified in 80%-90% of the cases (Potts et al., 2014).

To the authors' best knowledge, this is the first description of a true duodenoduodenal intussusception in a horse. Buchanan et al. (2006) described a pyloric-duodenal intussusception in a 15-year-old Tennessee-walking horse. The mare in that report showed recurrent colic when given access to feed or water (Buchanan et al., 2006). In the reported cases of duodenoduodenal intussusceptions in humans, patients commonly present with nonspecific signs of obstructive intestinal disease, which may be acute, chronic or intermittent. Weight loss and a trans-abdominally palpable mass may also be present (Pradhan et al., 2015). In the case described here, the horse showed an acute onset of severe abdominal pain with no previous history of recurrent colic or weight loss. A mass could be identified at transrectal palpation, and the lesion was visualised by means of transrectal and transabdominal ultrasonography.

Four lesions were concomitantly observed with the intestinal intussusception: a pseudodiverticulum, a segmental hypertrophy of the tunica muscularis, few multifocal intestinal ulcers and *hemomelasma ilei*-like lesions. Given their proximity, it is likely that at least some of these lesions are related to each other. However, with the available information, it is not possible to determine with certainty, which of them are primary and which are secondary. The diverticulum was located on the antimesenterial side just oral to the

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FIGURE 2 Post-mortem image of the excised stomach and proximal jejunum, showing the intussusception 50 cm aboral to the stomach. The most aboral part of the descending duodenum and the transverse part of the duodenum (intussusceptum) invaginated over a length of approximately 100 cm into the ascending duodenum (intussuscipiens). Note the multifocal, red, irregular, slightly raised areas in the serosa of the duodenum, immediately aboral to the intussusception (hemomelasma ilei-like lesions) (black arrow)

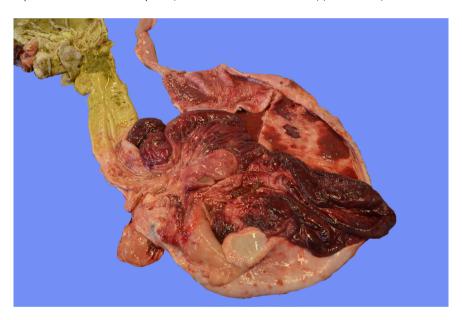


FIGURE 3 Intussusception after opening of intussuscipiens. Mucosa of intussusceptum is severely and diffusely oedematous and dark red colour (haemorrhagic venous infarction). The pyloric antrum of the stomach can be seen on the upper left corner of the image

intussusception, it was lacking the tunica muscularis of the intestinal wall and was therefore considered a pseudodiverticulum (Kumar et al., 2010). While some authors divide acquired diverticula into true and pseudodiverticula (Robertson, 1990; Southwood et al., 2010), others consider all diverticula as acquired and thus pseudodiverticula (Assenza et al., 2007). In the case subject to this report, given the age of the horse and concurrent presence of intestinal muscular hypertrophy, the pseudodiverticulum is most likely acquired. In acquired pseudodiverticula, the mucosa and submucosa are thought to herniate through the tunica muscularis because of increased intraluminal pressure (Bettini et al., 2003; Chaffin et al., 1992; Cordes &

Dewes, 1971; Mair et al., 2011). In most reported cases of pseudo-diverticula in cats and horses, the increased intraluminal pressure is thought to result from the hypertrophy of the tunica muscularis in the small intestine (Bettini et al., 2003; Chaffin et al., 1992; Cordes & Dewes, 1971). Pseudodiverticula can occur either on the mesenteric or the antimesenteric side of intestines. In case of concurrent muscular hypertrophy, they most often occur on the mesenteric side of the small intestines. It is thought that the mesenteric vessels penetrate the tunica muscularis and therefore create weaker areas predisposed for pseudodiverticula (Lindsay et al., 1981). In the current case, however, the pseudodiverticulum was located on the

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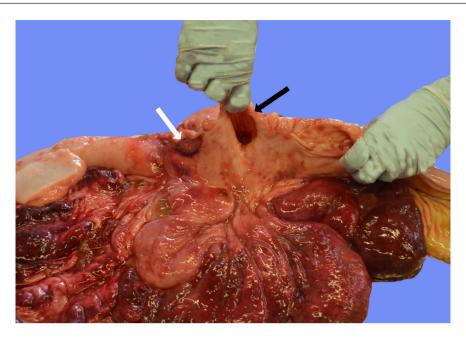


FIGURE 4 Diverticulum of approximately 5 cm in diameter (black arrow) and a focal ulceration of approximately $3 \times 3 \times 0.3$ cm (white arrow) are present immediately oral to intussusception. The oral part of the intestine is located on the right of the image

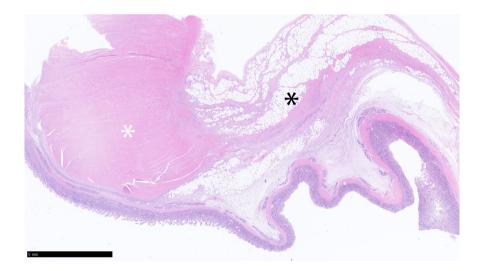


FIGURE 5 Duodenum, pseudodiverticulum. Transition between the pseudodiverticulum (right, black asterisk) and adjacent duodenal wall (left, white asterisk). The pseudodiverticular wall lacks tunica muscularis, is oedematous and is infiltrated by numerous mature adipocytes. Haematoxylin eosin stain

antimesenterial side of the duodenum. So far, a pseudodiverticulum on the antimesenterial side has only been described in three horses, all of them had an associated lymphoma (Matsuda et al., 2013).

The observed hypertrophied area of the intestinal muscular layer was limited to the duodenum and most pronounced in the central portion of the intussusception. Hypertrophy of the musculature of the small intestine and diverticulosis may exist concurrently or independently (Chaffin et al., 1992; Cordes & Dewes, 1971; de Solís et al., 2015). Muscular hypertrophy of the tunica muscularis of small intestines in combination with a pseudodiverticulum has been described in several species including horses, cats, pigs, rabbits and sheep. Muscular hypertrophy can

be idiopathic (if no predisposing cause can be identified) or secondary to chronic partial obstruction in an aboral intestinal segment that increases the workload on the smooth muscle (Bertram et al., 2019; Bettini et al., 2003; Chaffin et al., 1992; Cordes & Dewes, 1971).

In the horse of this report, the duodenoduodenal intussusception most likely occurred due to an underlying abnormality in the intestinal motility for which several possible causes may be accountable. For one, as mentioned above, idiopathic segmental muscular hypertrophy of the small intestines could have led to an increased intraluminal pressure resulting in the development of the pseudodiverticulum. Changes in intestinal motility caused by the thickened

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tunica muscularis, and the pseudodiverticulum may have contributed to the intussusception of the duodenum.

For another, aboral to the intussusception lesions compatible with hemomelasma ilei were present. Hemomelasma ilei is most often localised in the ileum but it can occur anywhere in the serosa of the intestines. It is thought to be caused by migrating larvae of Strongylus vulgaris and Strongylus edentatus (Barker & Druemel, 1985; Chaffin et al., 1992; Gelberg, 2008). Therefore, possible parasitic infestation may have altered the intestinal motility, and this could then have led to the development of segmental muscular hypertrophy, the pseudodiverticulum and the subsequent intussusception. However, no worms were detected during the post-mortem examination.

The presence of the ulcerations in the mucosa of the duodenum cannot be explained with the here proposed pathogeneses. Concurrent pathologies such as the occurrence of duodenal ulcers in association with equine gastric ulcer disease seem unlikely, as no ulceration was present in the stomach. Other possible causes for ulcerations in the duodenum, like a foreign body, is doubtful, as no such foreign material was found in the post-mortem examination. Therefore, a definitive causative agent for the multiple ulcerations could not be determined in the case presented here.

The acute component of the colic in the case described here can certainly be attributed to the haemorrhagic infarction of the intussusceptum and the acute obstruction of the intestinal passage. Since the horse did not show any previous colic symptoms or weight loss, it can be assumed that the additional changes such as the hypertrophy of the tunica muscularis and the diverticulum had no previous clinical relevance in this horse. This is in contrast to other studies that have shown that horses with hypertrophy of the tunica muscularis tend to have chronic intermittent colic and weight loss (Chaffin et al., 1992; Robertson, 1990). Diverticula can lead to colic symptoms in case of constipation, dilatation or rupture of the diverticulum (Cordes & Dewes, 1971; Mahne et al., 2017; Robertson, 1990).

In general, small-intestinal intussusceptions are surgical lesions that either require manual reduction or if the affected intestines are too oedematous and haemorrhagic, resection of the entire intussusceptum with subsequent end-to-end anastomosis. In a recent study, Haddad et al. could determine that the overall survival rate after surgical correction of small-intestinal intussusception is 53% (Haddad et al., 2021). No significant difference in short-term survival rate was depicted between horses that received manual reduction and horses that underwent resection and anastomosis (Freeman, 2019; Haddad et al., 2021). The length of the affected segment seems to be crucial. Previous studies showed that the longer the affected segment of the small intestine, the worse the chance of survival for the horse (Haddad et al., 2021; Morton & Blikslager, 2002). A gastroduodenostomy or a gastrojejunostomy with or without concurrent duodenojejunostomy or jejunojejunostomy (modified Roux-en-Y-procedure) would have been the only conceivable surgical therapy in this case (Gillis et al., 1994; Hanson et al., 1993; Mespoulhès-Rivière & Simon, 2008). Previous studies evaluating the surgical treatment of pyloroduodenal obstructions in foals revealed a long-term survival rate of 46%-69%

with post-operative stenosis of the created stoma being a frequent complication leading to new obstruction and subsequent gastric rupture and/or post-operative colic (Coleman et al., 2009; Orsini & Donawick, 1986; Southwood, 2021; Zedler et al., 2009). These surgical options to create a duodenal bypass are described for foals (Coleman et al., 2009; Orsini & Donawick, 1986; Zedler et al., 2009), whereas in adult horses, bypass procedures of the duodenum are rarely attempted (Hanson et al., 1993) and greatly restricted by the limited surgical access. In foals, the smaller size and depth of the abdomen make this region more accessible, but more importantly, because of the chronicity of the underlying gastric outflow obstruction in most cases, the stomach is greatly distended and chronically enlarged, making access through a ventral midline approach much easier (Orsini & Donawick, 1986). Other previously described approaches such as the J-incision or an approach through the dorsal paralumbar fossa could have additionally facilitated access to intussusception in the present case (Bauck et al., 2021; Durham, 1998). The J-incision as an approach to the cranial abdomen has also been used successfully in an adult horse that required a duodenojejunostomy (Ross et al., 1989). Because of the haemorrhagic venous infarction of the intussusceptum, a complete resection of the affected intestines would have been necessary. Complete exteriorisation of the duodenum for resection would still have been very difficult in the present case. In the authors' opinion, surgical therapy would therefore not have been possible.

In conclusion, this case report describes a duodenoduodenal intussusception with associated muscular hypertrophy and a pseudodiverticulum in a mature horse. Although only speculations regarding the cause for this extremely rare pathology can be made, a duodenoduodenal intussusception should be considered as a differential diagnosis in case of colic in adult horses where the clinical examination is indicating a small-intestinal pathology.

AUTHOR CONTRIBUTIONS

R. Mathys, C. Graubner, T. Kaiponen, L. Grau Roma and H. Brünisholz contributed to case management and preparation of the manuscript. E. Van der Vekens and C. Koch helped in the preparation of the manuscript. All authors gave their final approval of the manuscript.

ACKNOWLEDGEMENT

Open access funding provided by Universitat Zurich.

CONFLICT OF INTEREST

No conflicts of interest have been declared.

ETHICAL APPROVAL

The authors worked with the ethical standards of the use of animals in research. Furthermore, the owner of the affected horse has given her informed consent for publishing the information pertaining to the case by signing an informed consent form.

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How to cite this article: Mathys, R., Graubner, C., Kaiponen, T., Grau-Roma, L., Van der Vekens, E., Koch, C. et al. (2022) Duodenoduodenal intussusception in a 16-year-old German Warmblood mare. *Equine Veterinary Education*, 00, 1–7. Available from: https://doi.org/10.1111/eve.13688