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CASE REPORT

Treatment of congenital atypical haemangiosarcoma in a foal

Francisco José Vázquez Bringas[1,2] | Antonio Romero Lasheras[1,2] | Alicia Laborda García[1,3] | Javier Gómez-Arrue Azpiazu[3,4] | José Antonio García Jalón Ciércoles[2] | Arantza Vitoria Moraiz[1,2]

[1]Hospital Veterinario de la Universidad de Zaragoza (HVUZ), Zaragoza, Spain

[2]Departamento de la Patología Animal, Facultad de Veterinaria, Universidad de Zaragoza, Zaragoza, Spain

[3]Grupo de Investigación en Técnicas de Mínima Invasión de la Universidad de Zaragoza (GITMI), Zaragoza, Spain

[4]Instituto Universitario de Investigación en Nanociencia de Aragón (INA), Zaragoza, Spain

Correspondence: Francisco Vázques Bringas
Email: pvazquez@unizar.es

Present address
Javier Gómez-Arrue Azpiazu, Instituto Aragonés de Ciencias de la Salud, Zaragoza, Spain

Summary

Haemangiosarcoma is a rare vascular tumour in horses, usually originating from blood vessel endothelial cells. We present the case of an 8-day-old foal, referred for an atypical large subcutaneous mass on the left side since birth. Ultrasonographically, it showed multiple cavities with hypoechoic content, marked vascularisation and fluid movement between cavities. As the nature of the mass suggested that surgery could result in profuse bleeding, we decided to perform an initial arteriography to identify the pattern and calibre of the main vessels and embolisation of this vascular supply, which allowed surgical removal with less bleeding than expected. This approach, with pre-surgical transarterial embolisation of the tumour, is not commonly used in equine surgery. Histology established a diagnosis of cutaneous haemangiosarcoma. During 1-year post-surgery, clinical and ultrasound examinations were carried out without any signs of recurrence or metastasis. One year later, the foal was euthanised due to a limb fracture. No macroscopic signs of metastasis were observed at necropsy. Histology showed no signs of recurrence. Cutaneous haemangiosarcomas, though rare, should be included in the differential of masses and growths with compatible ultrasound or cytological findings. Transcatheter arterial embolisation of highly vascularised neoplasms can reduce bleeding and facilitate subsequent surgical resection.

KEYWORDS
horse, congenital, endovascular embolisation, haemangiosarcoma

INTRODUCTION

Haemangiosarcoma, also known as angiosarcoma or malignant haemangioendothelioma, is a malignant neoplasm derived from the endothelial cells of blood vessels (Taintor, 2014).

It is a rare tumour in horses (Southwood et al., 2000). Reported cases include neoplasms originating from the endothelium of vessels in dermal or subcutaneous tissues (Schaffer et al., 2013) or from many other locations, predominantly ocular/periocular (Bischofberger et al., 2008; Sansom et al., 2006; Scherrer et al., 2018). Disseminated cases have also been described (Jean et al., 1994; Southwood et al., 2000). These types of metastatic forms usually affect adult or old horses (Ferrucci et al., 2012), which is the age range most commonly affected (Knottenbelt & Clegg, 2004; Taintor, 2014), although they can also be found in young animals (Johns et al., 2005) and even congenitally (Dunkel et al., 2004).

The clinical presentation of these tumours is highly dependent on the wide variety of possible locations (Beaumier et al., 2020;

Hughes et al., 2018; Segabinazzi et al., 2021). Haemangiosarcomas vary in size and can be solitary but are often multiple. They may be lobulated, and some are encapsulated and subdivided by thick fibrous septa. Cutaneous and subcutaneous masses in young horses often present first as an enlarged paw or joint effusion (Johns et al., 2005). Congenital masses can be multiple, affecting large areas (Taintor, 2014).

The aim of this case report is to summarise clinical and diagnostic imaging findings and treatment of a neonate with an atypically large subcutaneous congenital haemangiosarcoma over the left shoulder.

CASE HISTORY

A Spanish pure breed (PRE) foal, 8 days old, was referred to our hospital, presented with a mass of approximately 40×20 cm since birth, next to the left shoulder (Figure 1).

CLINICAL FINDINGS

The mass was of a non-painful fleshy consistency. Cardiac and respiratory frequency, rectal temperature, mucous membranes colour, capillary refill time, locomotion and behaviour of the foal were normal.

Ultrasonographic exploration of the mass showed a cavitated appearance, with thin echogenic tissue delimiting multiple caverns occupied by hypoechoic content of fluid appearance (Figure 2). Pulsed Doppler mode showed marked vascularisation and fluid movement between cavities (Figure 2).

Several fine-needle puncture aspirations were performed, obtaining abundant liquid material, with a macroscopic appearance of blood and flowing with pulsatile pressure. Cytological analysis showed a blood-like appearance (but with a higher haematocrit than the venous sample); no neoplastic findings were observed in any of these samples.

DIAGNOSIS

Based on these findings, a presumptive diagnosis of cavernous haemangioma was made. A preoperative biopsy of the mass was not performed because of the risk of bleeding (Hurcombe et al., 2022), and because the owner requested surgical excision, even without a definitive anatomopathological diagnosis.

TREATMENT

The nature and ultrasonographic findings of the mass suggested a surgical intervention would likely result in abundant haemorrhage (Hurcombe et al., 2022), so we decided to carry out a preliminary arteriography with the double objective of identifying the pattern, origin and calibre of the vessels, as well as embolising the main vessels to facilitate subsequent surgical excision.

Under general anaesthesia, the foal was positioned in right lateral recumbency on an interventional radiology radiolucent table to allow the use of a fluoroscopy C-arm. The first attempt was made to access through the common carotid artery, using ultrasound-guided arterial puncture, without arteriotomy, as described in a case series report that includes this case (Vitoria et al., 2022). Digital subtraction angiography, using iohexol as contrast media, showed that, from this carotid access, it was possible, but complicated, to reach the vessels that supplied the tumour, due to the complexity of the required rotation of the catheter at the level of the subclavian artery. The foal was then placed in the left lateral recumbency position to

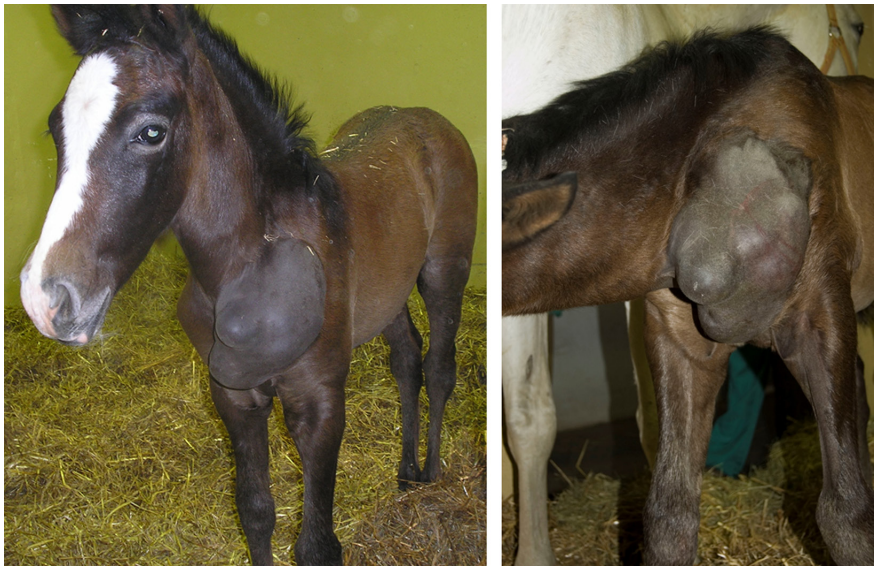

FIGURE 1 Images of the foal with the congenital mass at the base of the neck and over the left shoulder.

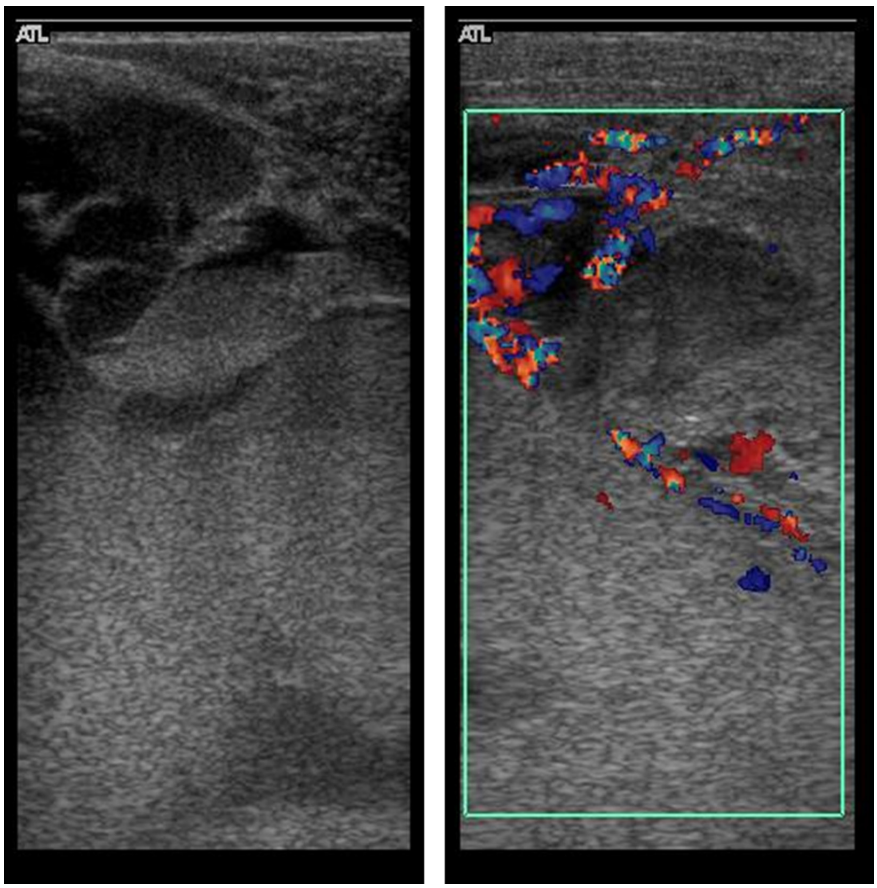

FIGURE 2 Ultrasonographic aspect of the mass. Left: (B mode) cavitated appearance, with thin echogenic tissue delimiting multiple caverns occupied by hypoechoic content of fluid appearance. Right: (colour Doppler) marked vascularisation.

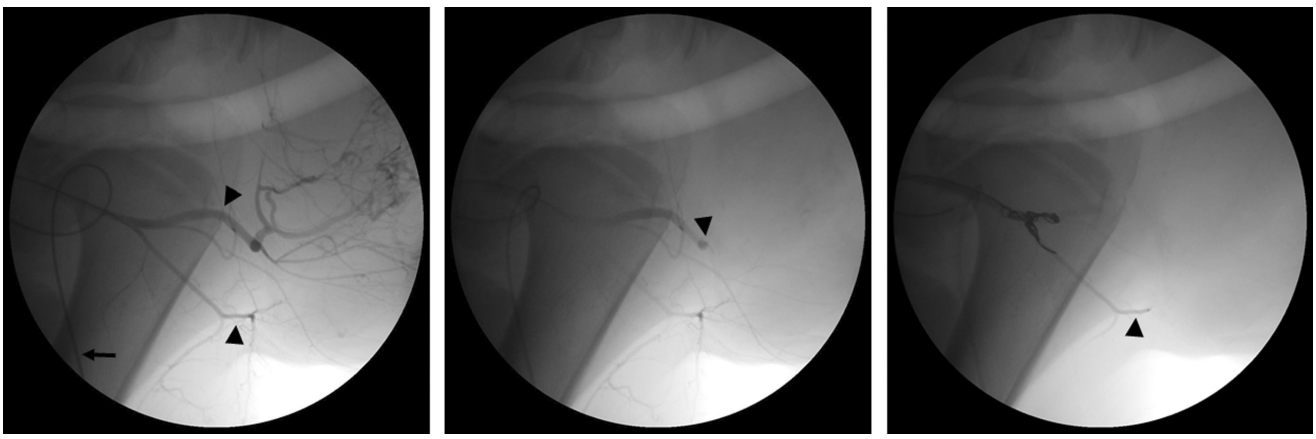

FIGURE 3 Fluoroscopic images of the endovascular procedure prior to surgical resection. Left: selective arteriography showing the artery supplying the tumour, with two main branches (arrow heads). Catheter is coming from the median artery (arrow). Middle: arteriography after polyvinyl alcohol embolisation of the upper branch, showing interruption to the flow to the tumour. Right: immediate control arteriography after final embolisation of the proximal branches with platinum coils.

allow percutaneous puncture of the median artery on the medial aspect of the forearm, using a 5Fr introducer sheath to gain arterial access.

A new angiographic examination from the introducer sheath allowed identification of the vascular pattern and origin. Embolisation of the main vessels was performed with a 5 Fr C2 diagnostic catheter, starting with distal embolisation of the smaller vessels with polyvinyl alcohol (PVA) particles (300–500 μm), until flow into the tumour was significantly reduced. When the diagnostic angiography showed no more contrast filling of the small vessels surrounding the mass, the same catheter was used to deploy a 0.035″ embolisation coil in the main two supplying vessels (Figure 3), to achieve proximal closure.

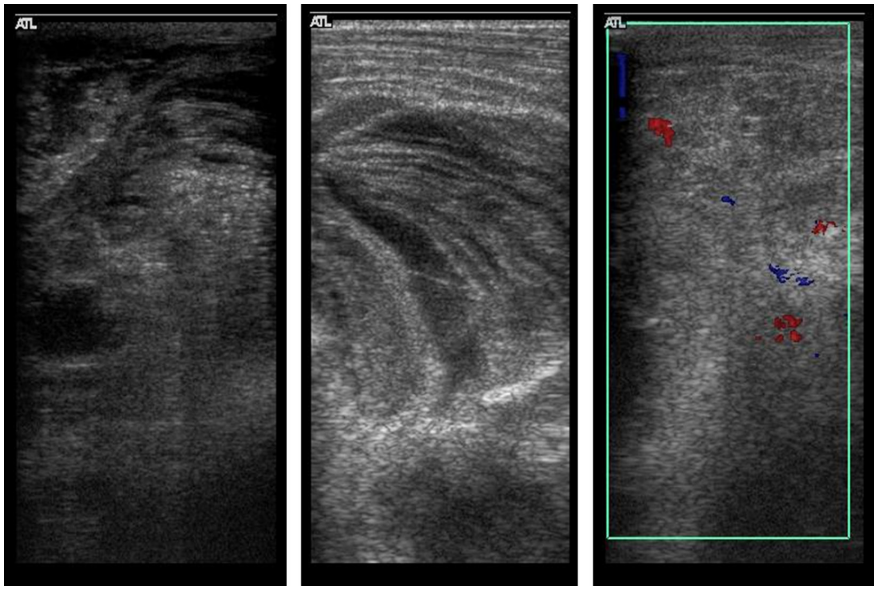

FIGURE 4 Ultrasonographic appearance 7 days after embolisation. Left and middle: changes in the ultrasound pattern (B-mode). Right: obvious changes were seen with colour Doppler.

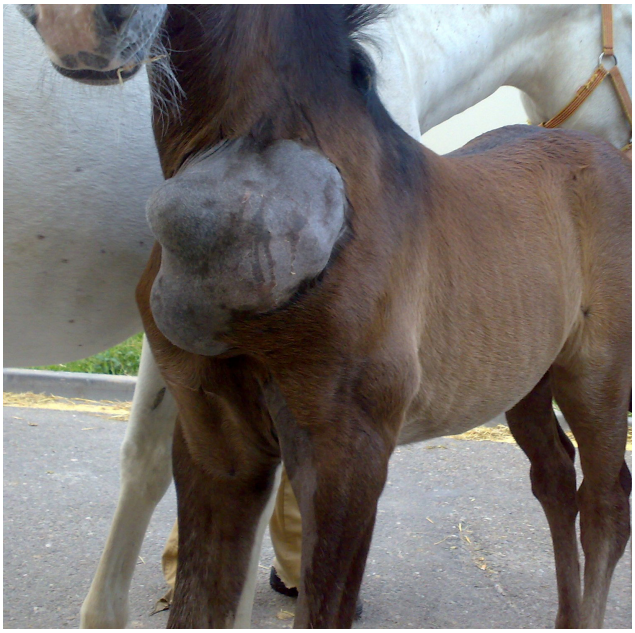

FIGURE 5 External appearance of the mass 5 days after the embolisation.

After embolisation, there was some swelling at the site and slight hyperthermia (up to 39.3°C). Although obvious changes were seen with pulsed Doppler (Figure 4), only some discrete changes were seen in the ultrasound pattern (B-mode) (Figure 4), and in the size and external appearance of the mass (Figure 5).

After 21 days, the foal was again anaesthetised for surgical excision, performing a previous post-embolisation control angiography at the same intervention. Despite the imperceptible external changes in the size of the mass, this second arteriography showed a significant decrease in blood supply (Figure 6), which allowed for a very easy surgical excision with less bleeding than expected. A single large curved skin incision was made over the mass, 4 cm cranial to its dorsocaudal border. Subcutaneous tissue was carefully dissected over the mass. The tumour could be fairly easily separated from surrounding tissues by blunt dissection (Figure 7) with the fingers, except in the distocaudal area where it infiltrated muscle tissue. Only a few vessels needed to be ligated or thermo-sealed in the area where the mass was invading the muscle. Some excess skin was removed and traction stitches were made in the subcutaneous space, trying to eliminate dead space. A drainage tube was externalised through declive skin incision. The skin was sutured with a single interrupted suture.

Postoperatively, flunixin (1.1 mg/kg bwt IV twice daily) was administered for 5 days, as well as sodium penicillin (25,000 IU/kg bwt IV, every 6 h) and gentamicin (6.6 mg/kg bwt IV, once daily) for 7 days.

OUTCOME

After surgery, macroscopic evaluation of the excised mass revealed tumour-like subcutaneous tissue, affecting muscle tissue, highly vascularised, with intense hyperaemia, and multiple cavities filled with partially coagulated blood (Figure 8).

Microscopically oval, spindle-shaped, tumour-like cells were evident, which attempted to form vascular lumina by grouping together to form solid septa, giving rise to cavities with coagulated blood and infiltrating between the skeletal muscle fibres, causing degeneration and atrophy. Their nuclei were large and vesicular with a moderate mitotic index (Figure 9). These findings permitted the diagnosis of the resected mass as a congenital cutaneous haemangiosarcoma.

During the postoperative period, the foal showed no signs of pain. In the first week, there was dehiscence of the central part of the suture line and it was left to heal by secondary intention.

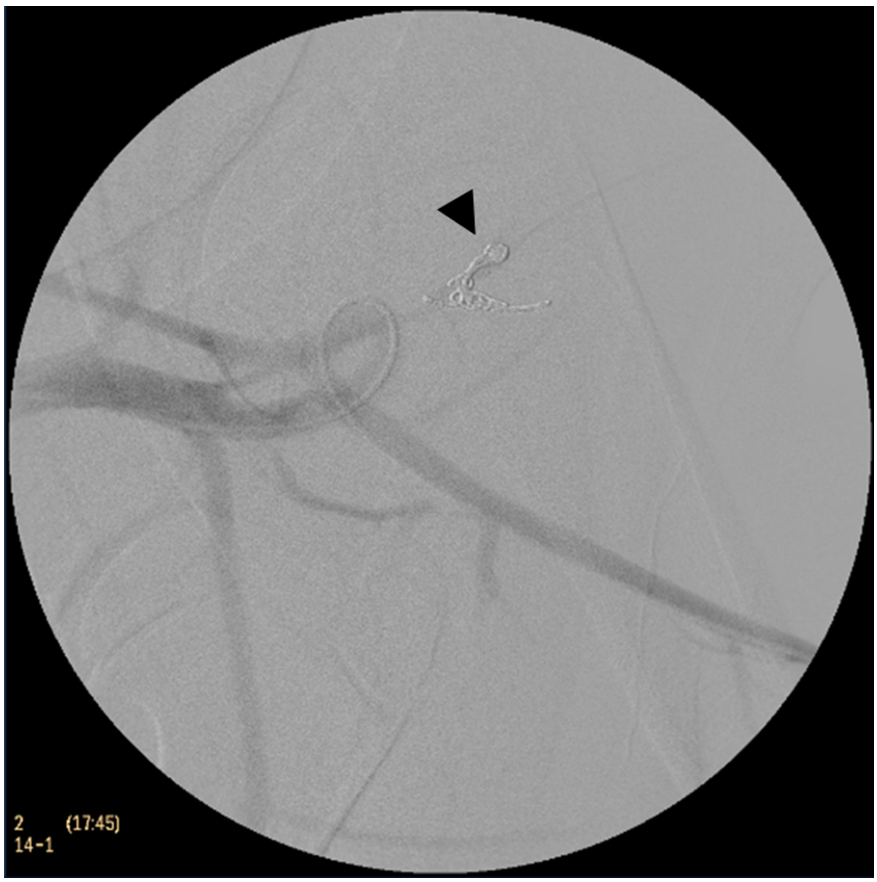

FIGURE 6 Non-selective digital subtraction arteriography previous to surgical excision, 21 days after embolisation. Platinum coils remain at the same position (arrowhead), no flow is coming from the original supplying arteries; there is no revascularisation of the neoplasm from new branches of the subclavian artery.

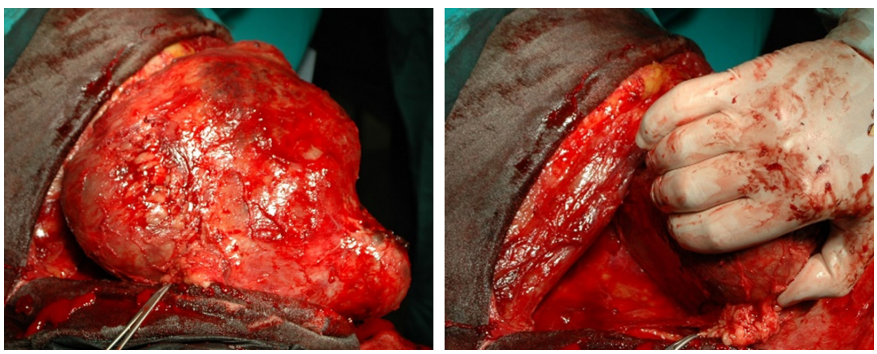

FIGURE 7 Images of the surgical excision.

Postoperative follow-up with clinical and ultrasonographic examinations was carried out during the first year after the surgery. At 5 months, the wound was healing by second intention and the defect created by the removed muscle tissue was visible in the neck (Figure 10). No clinical or ultrasonographic findings of recurrence or metastasis were observed. Considering the size and appearance of the tumour before surgery, the clinical and aesthetic result was considered satisfactory by the owner.

One year later, the yearling suffered an open fracture of the femur and due to the poor prognosis was euthanised.

POST-MORTEM FINDINGS

No metastases were observed at necropsy in the field. Histology of samples taken from the operated area showed no signs of recurrence.

DISCUSSION

A case of congenital cutaneous haemangiosarcoma in a foal is reported, with an atypical presentation, due to the large size of the

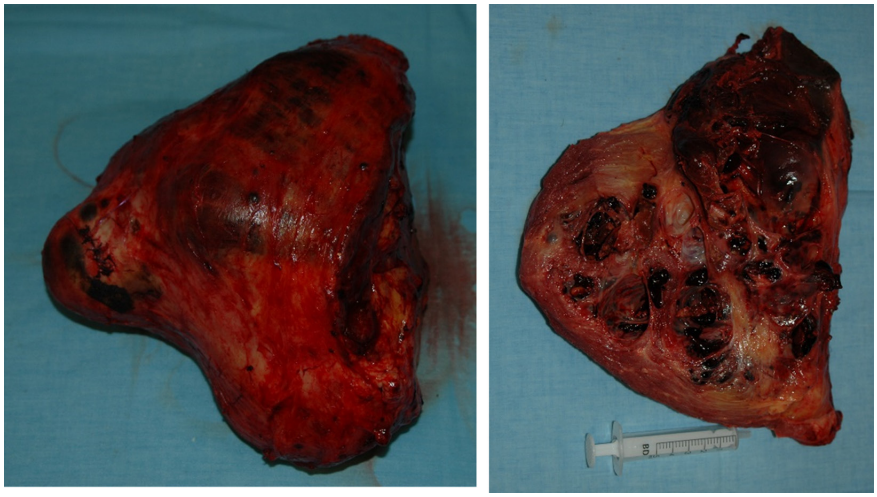

FIGURE 8 Macroscopic appearance of the tumour after excision (left) and following longitudinal section (right), showing multiple cavities filled with coagulated blood.

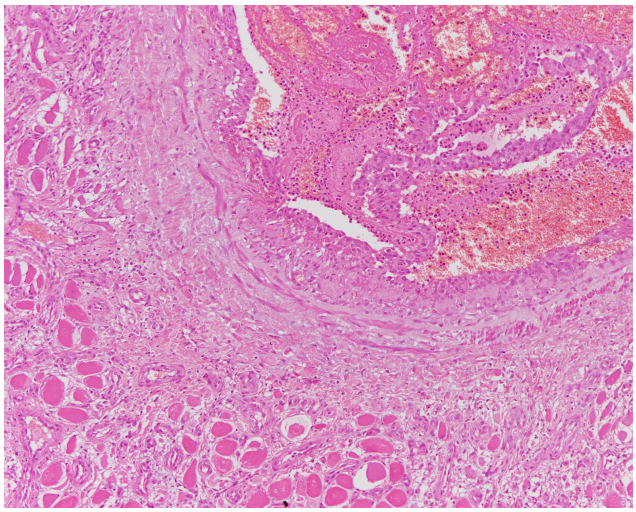

FIGURE 9 Microscopic (×100) examination showing, oval, spindle-shaped, tumour-like cells, which attempted to form vascular lumina by grouping together to form solid septa, giving rise to cavities with coagulated blood. Their nuclei were large and vesicular with a moderate mitotic index.

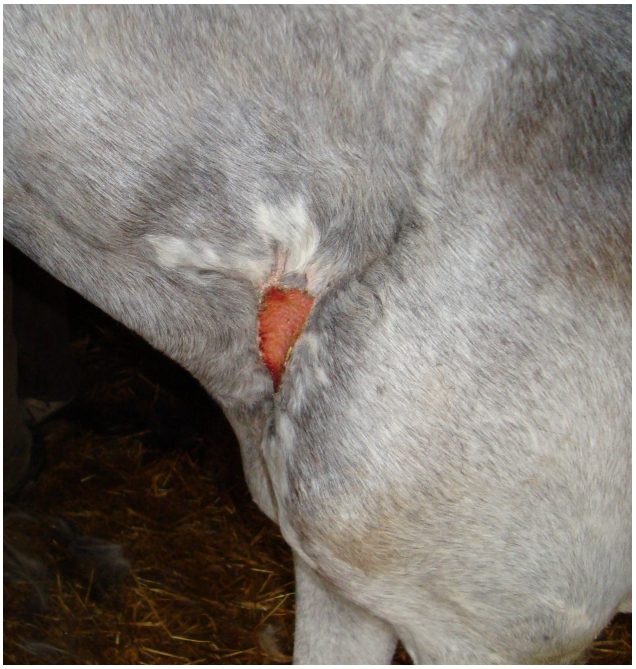

FIGURE 10 Aspect of the wound healing by second intention 5 months after the surgery, with the defect created by the removed muscle tissue visible in the neck.

mass and multiple vascular cavities, which exceed those described in similar cases (Cian et al., 2015).

Haemangiosarcoma is a rare neoplasm in horses, whose symptomatology is highly dependent on the multiple possible locations. In this case, the initial presumptive diagnosis was congenital arteriovenous malformation or cavernous haemangioma (Taintor, 2014). Fine-needle aspiration was performed, with no signs of neoplastic disease. There was no initial suspicion of malignancy and therefore no thoracic radiographs or ultrasound were performed to rule out the presence of metastases. Definitive diagnosis of haemangiosarcoma requires histopathology, which is often achieved only at post-mortem examination (Burks et al., 2009). In this foal, histopathology of the excised mass allowed a definitive diagnosis, although it would have been better characterised by immunohistochemistry (Cottle et al., 2008), which was not performed.

Proposed treatments for haemangiosarcomas with disseminated or undefined margins include topical or systemic chemotherapy, radiotherapy and interstitial brachytherapy (Burks et al., 2009). Although haemangiosarcoma can be a malignant tumour, it has been described that surgical resection can be curative in non-metastatic and well-localised tumours (Taintor, 2014). It has even been suggested that cutaneous presentations are less aggressive than

visceral presentations, with a longer clinical course and survival time and possible cure of dermal lesions by excision alone (Knottenbelt et al., 2015). In any case, this cutaneous presentation has been successfully excised in horses, without recurrence during years of follow-up. Moreover, some cases suggest that in young horses (<3 years), haemangiosarcomas grow more slowly and some animals have survived long term even without surgery; in these animals, dissemination is also less likely (Taintor, 2014). Unfortunately, in our case, the horse had to be euthanised after 1 year due to an unrelated accidental fracture, with no possibility of knowing its longer-term evolution.

Haemorrhage has been reported to be common in these tumours (Hurcombe et al., 2022), as the delicate vascular channels that form these neoplasms are prone to rupture (Knottenbelt et al., 2015). The ultrasound and colour Doppler appearance and the large size of the mass and its vessels aggravated that possibility in this case. It was therefore decided to perform arteriography and embolisation prior to surgical excision. In human medicine, transarterial embolisation of tumours have been routinely practiced worldwide in the last decades (Huszty et al., 2021; Schnapauff et al., 2018). However, in veterinary medicine, very few cases have been reported with tumour embolisation, either with (Kawamura et al., 2021) or without later surgical excision (Cave et al., 2003; Culp et al., 2021; de La Villeon et al., 2011; Iwai et al., 2015; Oishi et al., 2019; Sun et al., 2002; Weisse et al., 2002). Most of these cases are in small animals. Our patient showed slight hyperthermia with no concomitant leucocytosis after embolisation which could be consistent with a post-embolisation syndrome (PES) described in human and canine patients. This syndrome is described in human medicine as one of the most common complications of transarterial embolisation and chemoembolisation. The main symptoms in humans are hyperthermia, pain and anorexia/nausea/vomiting—those last associated mainly with intra-abdominal embolisation (Romero Ubillus et al., 2011). In dogs undergoing transarterial embolisation, vague, flu-like symptoms consistent with a PES have been appreciated at a frequency of 26%–100% (Gibson et al., 2022). In equine surgery, transarterial embolisation is mainly used in cases of mycosis of the guttural pouch (Lepage & Piccot-Crézollet, 2005), but to our knowledge there are no described cases where this type of procedure is used to treat tumours in horses. In this case, the use of this technique may have contributed to a much less bleeding surgery.

CONCLUSIONS

Cutaneous haemangiosarcoma, although rare in horses, should be included in the differential diagnosis of masses and growths with compatible ultrasound or cytological findings. Not all cutaneous haemangiosarcomas recur or metastasise, especially if they are congenital or affect very young animals. Presurgical transcatheter arterial embolisation of highly vascularised large tumour masses may reduce bleeding and facilitate subsequent surgical resection.

AUTHOR CONTRIBUTIONS

F. J. Vázquez and A. Romero contributed to study design, study execution, data analysis/interpretation and preparation of manuscript. F. J. Vázquez and A. Romero contributed equally to this paper. A. Laborda, J. Gómez-Arrue Azpiazu and A. Vitoria have contributed to study design, study execution, data analysis/interpretation and preparation of manuscript. J. A. García-Jalón has contributed to study execution, data analysis/interpretation and preparation of manuscript. All authors have approved the final version of the manuscript.

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CONFLICT OF INTEREST

No conflicts of interest have been declared.

ANIMAL ETHICS AND OWNER CONSENTS

According to the Spanish Policy for Animal Protection RD 53/2013, which meets the European Union Directive 2010/63 on the protection of animals used for scientific purposes (Article 1), non-experimental 'clinical veterinary practices' are exempted from the scope of above-mentioned Directive. 'Clinical veterinary practice' includes procedures and techniques performed by veterinary surgeons in the course of their professional duties, which ensure the health and welfare of animals committed to their care. Owner consent for the use of their horse's data and recordings, as part of a general consent form, was obtained upon admission at the Equine Surgery and Medicine Service of the Veterinary Hospital of the University of Zaragoza.

ORCID

Francisco José Vázquez Bringas https://orcid.org/0000-0002-8712-2275

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