

## Cutaneous vascular hamartoma in a lamb

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**Abstract:** A male Polwarth lamb with a good body score was born with a pendulous, oval, red mass adhering to the skin in the left ventrolateral thoracic area. The nodule was removed when the lamb was five days old and sent for a gross and histopathological evaluation. Macroscopically, the mass was reddish, slightly oval and flat, well delineated, and contained a central ulcerated area. Microscopically, it showed a marked proliferation of well-differentiated endothelial cells with the formation of new blood vessels. An immunohistochemical analysis showed mild cytoplasmic immunostaining for the von Willebrand Factor in the endothelial cells. To the best of our knowledge, this is the first report of a cutaneous vascular hamartoma in a sheep.

**Keywords:** congenital disorder; sheep; skin; vascular malformation

Vascular hamartoma is defined as a vascular malformation characterised by the disorganisation and excessive proliferation of the vascular tissue, visualised in an anatomically normal site (Smith 2001; Yasuno et al. 2011; Borel et al. 2014). The abnormal cell growth often starts during gestation, and, in most cases, the hamartoma is detected on clinical evaluation at birth (Smith 2001; Brunson et al. 2006). Sometimes, hamartomas are difficult to differentiate from vascular neoplasms, the main indication being limited growth. Structures such as large well-defined vessels and lack of mitotic activity are suggestive of hamartoma (Bildfell et al. 2002; Shirota et al. 2013).

This vascular malformation has been described in horses (Colbourne et al. 1997; Borel et al. 2014; Nolf et al. 2014), cattle (Benoit et al. 2005; Macedo et al. 2011; Brisville et al. 2012), dogs (Gualtieri et al.

2009), cats (Martin-Vaquero et al. 2011), and goats (Bildfell et al. 2002), along with one case in the lung and heart of a sheep (Bradley et al. 1980; Anderson et al. 1990). In humans, although several organs can be affected, cutaneous vascular hamartomas are the most frequent and is also known as eccrine angiomatous hamartoma (Requena et al. 1997).

There is wide discussion about the classification of cutaneous vascular anomalies in humans, mainly with regard to the differentiation of vascular neoplasms and malformations (Requena et al. 1997). In veterinary medicine, vascular hamartomas are also classified as angiomatous hamartomas, telangiectatic hamartomas, and angiomas (Smith 2001; Sakurai et al. 2011), but should be considered as the same entity. Here, we report a case of cutaneous vascular hamartoma in a sheep.

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### Case report

A male Polwarth (Ideal) lamb with a good body score was born with a pendulous, oval, red nodule on the skin of the left ventrolateral thoracic area (Figure 1A). The nodule was surgically removed five days after birth and submitted for a histopathological evaluation. Macroscopically, the mass was reddish, slightly oval, and flattened, measuring  $9.0 \times 8.0 \times 3.0$  cm. It was well delineated, with a rounded border and contained areas of ulceration in the central region. Upon dissecting, the nodule was soft and red and contained blood in cavities (Figure 1B). A rim of fibrovascular tissue marked the transition between the lesion and the surrounding normal skin.

After the macroscopic evaluation, the material was preserved in 10% neutral buffered formalin and fragments of the nodule were embedded in paraffin and stained with haematoxylin-eosin and Masson's trichrome. The histological sections were submitted for an immunohistochemical evaluation using the streptavidin-biotin-peroxidase method and incubated with the primary polyclonal anti-von Willebrand Factor (vWF) antibody (Dako, Carpinteria, CA, USA) with a dilution of 1 : 200 and heat-induced antigen retrieval in a citrate buffer (pH 6.0) for 3 min at 125 °C under pressure.

Microscopically, the nodule was characterised by a marked proliferation of well-differentiated endothelial cells arranged within the vascular structures with multiple lobes extending from the

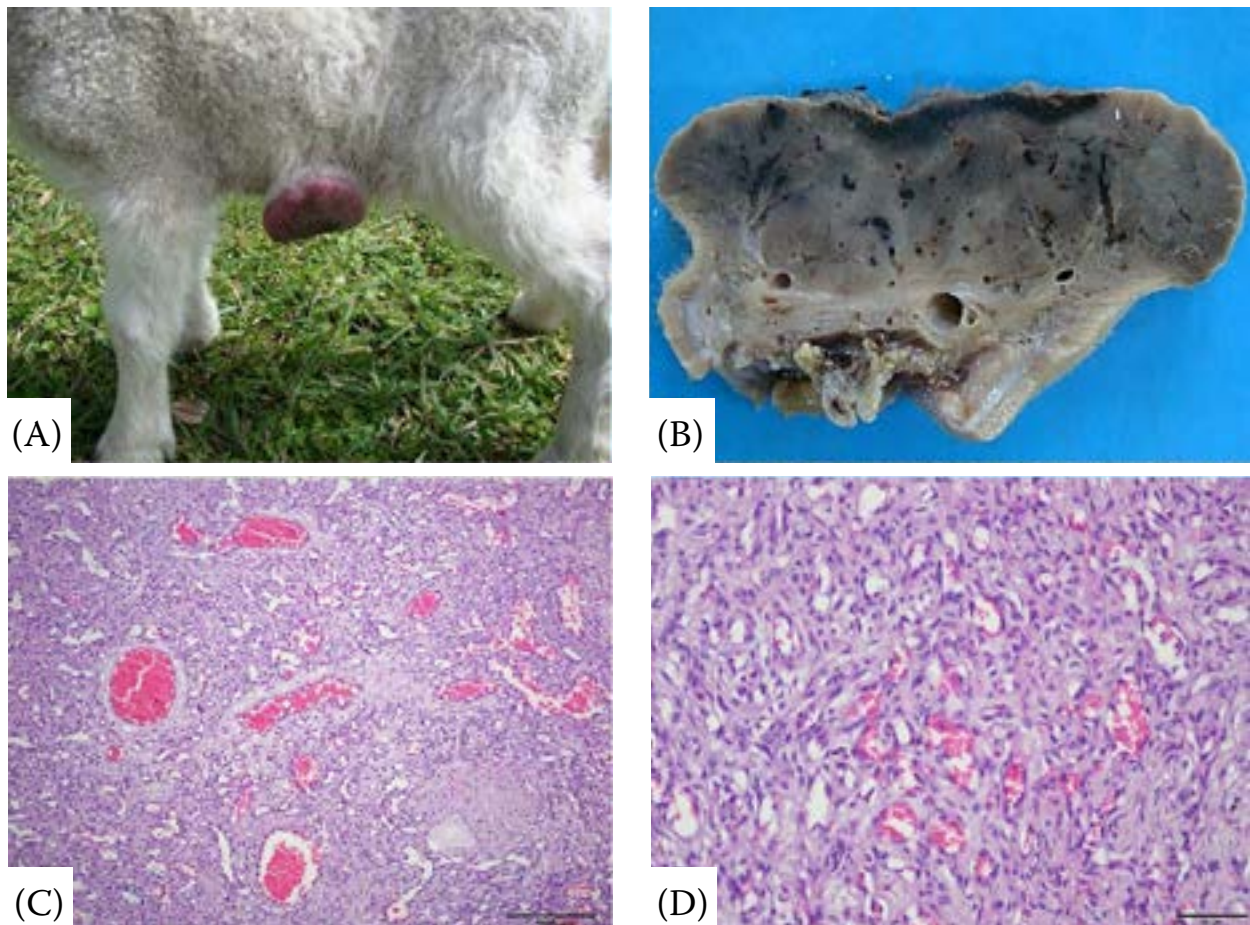


Figure 1. A cutaneous hamartoma in a lamb. (A) A well-defined reddish pendulous nodule is observed on the skin of the left ventrolateral thoracic region. (B) The cutaneous surface of the dermal nodule fixed in 10% formalin. The border between the skin and the surface of the nodule contains a fibrovascular tissue. (C) Significant proliferation of the endothelial cells forming vascular structures supported by a moderate amount of fibrous connective tissue can be observed. Dilated vessels filled with erythrocytes are noted. Haematoxylin and eosin staining. Bar = 200 µm. (D) The nodule shows a pattern of well-differentiated endothelial cells with formation of capillaries structures. Haematoxylin and eosin staining. Bar = 50 µm

superficial dermis to the deep dermis. The vascular structures consisted of small capillaries lined by a layer of flattened endothelial cells (Figure 1C) as well as large vessels containing a layer of endothelial cells and filled with erythrocytes, sustained by a connective stroma. The endothelial cells exhibited either a spindle-shape or oval nucleus with dense chromatin and minimal anisokaryosis, and scarce eosinophilic cytoplasm, with indistinct borders (Figure 1D). The vascular proliferation was supported by a moderate stromal fibrous connective tissue, which was stained using Masson's trichrome. A moderate orthokeratotic hyperkeratosis and areas of ulceration were observed in the epidermis. The immunohistochemical analysis showed a vWF-positive immunoexpression, limited to the cytoplasm of the endothelial cells in the anomalous blood vessels.

## DISCUSSION AND CONCLUSIONS

The diagnosis of a vascular hamartoma was established based on the epidemiological, macroscopic, and microscopic findings (Bildfell et al. 2002; Gualtieri et al. 2009; Borel et al. 2014). In this case, the lamb's birth with the lesion, as well as the delimitation and non-invasive mass growth with an irregular vessel formation and minimal cellular pleomorphism suggest that it was a vascular malformation. The vascular proliferation was confirmed by the immunohistochemistry examination. It is believed that some growth factors are involved in the pathogenesis of such lesions, e.g. interacting fibroblast growth factor and endothelial growth factor (Yasuno et al. 2011) may trigger the start of a hyperplastic process with neovascularisation.

Vascular hamartomas, although infrequent, may develop in different regions of the body. Although not frequently, they have been described in goats (Bildfell et al. 2002), in horses and humans (Requena 1997). In horses, some cases have been described in the brain (Borel et al. 2014), tongue (Brunson et al. 2006), caecum (Nolf et al. 2014), ovary (Rhyan et al. 1981), and hind limb (Colbourne et al. 1997; Saifzadeh et al. 2006). This being the specie most affected by this malformation.

In cattle, these lesions have been observed in the heart (Sugiyama et al. 2007; Brisville et al. 2012), ovary (Benoit et al. 2005; Macedo et al. 2011), gingival tissue (Mohammadi et al. 2007), and liver

(Braun et al. 2011). Cases of dogs with hamartoma in the brain have also been reported (Sakurai et al. 2011), as has one case in the uterus of a rat (Shirota et al. 2013). In these cases, vascular changes were similar to those described in the sheep in this study, all of them with lesions characterized by irregular growth of neoformed blood vessels.

Hamartomas in sheep are rare (Anderson et al. 1990). Cases of cardiac and pulmonary hamartomas were described in sheep (Bradley et al. 1980; Anderson et al. 1990). However, these cases corresponded to hamartomas characterized by proliferation of cardiac muscle and lung tissue, respectively, with irregular growth of these tissues, but without vascular changes. To date, no case of cutaneous vascular hamartoma has been reported in the sheep.

In the present case, the vascular malformation did not cause any clinical symptoms and was successfully removed surgically without any sequelae. Although considered a benign lesion, the severity of the clinical signs would depend on its location. A case of a hamartoma was described in the brain of a dog that had clinical signs of blindness and decreased proprioception (Sakurai et al. 2011). A vascular hamartoma has also been described as a cause of laminitis in a horse (Saifzadeh et al. 2006).

Vascular hamartomas can be easily confused with haemangiomas (Bildfell et al. 2002; Yasuno et al. 2011; Nolf et al. 2014). When the microscopic aspects of a hamartoma are very similar to a neoplastic lesion, the macroscopic findings and their presence at birth help define the diagnosis of a vascular malformation. However, the presence of well-differentiated cells is characteristic of a lesion that is not a tumour (Benoit et al. 2005; Brunson et al. 2006). In this case, the affected vascular structures were observed to be lined by endothelial cells with flattened nuclei, morphologically compatible with the vascular structure. However, in cases of haemangiomas, the behaviour is more invasive and occurs more frequently in adult animals (Requena 1997; Bildfell et al. 2002; Sakurai et al. 2011). Hamartomas should be differentiated from choristomas, another congenital malformation, in which histologically normal tissue grows in ectopic sites (Batra 2012).

As previously described, these vascular malformations are generally solitary well-circumscribed nodules (Bildfell et al. 2002; Brunson et al. 2006), but, in some cases, they may be infiltrative and poorly delineated (Yasuno et al.



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2011; Nolf et al. 2014), which hinders their surgical removal (Saifzadeh et al. 2006). The lesion in the present case was present at birth, similar to reports in other species (Smith 2001; Brunson et al. 2006; Mohammadi et al. 2007). However, cutaneous vascular hamartomas have been diagnosed in goats that were approximately two years of age (Bildfell et al. 2002).

A microscopic evaluation along with histochemistry and immunohistochemistry examinations are tools that can help to identify the type of proliferating vascular tissue, determine the degree of the cellular pleomorphism, and differentiate this malformation from a neoplastic disorder (Smith 2001). The anti-vWF antibody is the most commonly used one to detect vascular structures, but the labelling may vary according to the degree of maturity of the proliferated cells. In this case, the marking basically limited a single layer of endothelial cells, which reinforces the diagnosis of a hamartoma. The alpha smooth muscle actin antibody can also be used to aid in the diagnosis (Yasuno et al. 2011; Borel et al. 2014).

In summary, to the best of our knowledge, this is the first report of a vascular hamartoma with cutaneous involvement in a sheep. Although rare, this disorder should be included in the differential diagnosis for neoplastic epidermal lesions in this species.

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## Conflict of interest

The authors declare no conflict of interest.

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