https://doi.org/10.33472/AFJBS.6.13.2024.2354-2359

African Journal of Biological Sciences

Journal homepage: http://www.afjbs.com

Research Paper Open Access

First Description of Mandibular Cystic Hygroma in a Crossbred Goat

Nassim Sid [1,2], Nora Mimoune [3], Choayb Mecherouk[4*], Nassima Ait Issad [3], Nor El Houda BELALMI [1,2], Rédha BELALA[4], Djamel KHELEF [3]

[1]Laboratory of Health and Environment. University Mohamed El Bachir El Ibrahimi of Bordj Bou Arreridj, Algeria.
[2]Department of Biology, Faculty of Nature and Life Sciences and Earth and the Universe, University Mohamed El Bachir El Ibrahimi of Bordj Bou Arreridj, Algeria.
[3]Animal Health and Production Laboratory (Animal Health & Production Laboratory), Higher. National Veterinary School, Issad Abbes, Oued Smar, Algiers, Algeria.
[4]LBRA, Veterinary Institute, Saad Dahleb, Blida University 1, Blida, Algeria.*

Corresponding author: [4*]choayb.mech@gmail.com

Article Info

Volume 6, Issue 13, July 2024

Received: 28 May 2024

Accepted: 30 June 2024

Published: 26 July 2024

doi: 10.33472/AFJBS.6.13.2024.2354-2359

ABSTRACT:

Cystic hygromas (cystic lymphangiomas, macro cystic lymphatic malformations) are seldom observed in different animal species and they have not yet been reported in goats. They are rare benign congenital vascular disorders of the lymphatic system that commonly localize in the cervico-facial region. A two-month-old, male crossbred goat-kid presented with a single swollen and painless mass (5 cm in diameter) on the left side of the mandible area occurring from birth. Ultrasonography showed a fluid-filled multi-lobular cystic lesion with internal separations. The cytological examination revealed the presence of cloudy serous fluid with a predominance of lymphocytes. To the authors' knowledge, this is the first description of a congenital lymphatic anomaly in goats.

Keywords: Cystic hygroma, lymphatic malformation, mandible, goat.

1. INTRODUCTION

Cystic hygroma, also known as cystic lymphangioma or macro cystic lymphatic malformation, is a rare benign congenital fluid-filled lesion due to the absence of normal communication between the lymphatic and venous systems that originate from an aberrant

proliferation of lymphatic vessels (ERÖKSÜZ et al., 2021; SHRESTHA et al., 2022). This failure of communication eventually causes the accumulation of lymphatic fluid, which forms a cystic mass (YARSO et al., 2023). Cystic hygroma is defined as the most frequent type of lymphangioma. There are several different classification schemes of lymphangiomas. The most frequently used divides them based on cyst size into microcystic, macrocystic and mixed lymphangiomas (LIU et al., 2021). In humans, this lymphatic malformation is characterized by the formation of a single or multiple cysts within the soft tissues, most commonly present at birth or by 2 years of age. It can occur anywhere in the body, but the most common sites are the head and neck. Clinically, cystic hygroma is usually asymptomatic but complications such as dysphagia and complete or incomplete airway obstruction may arise (MIRZA et al., 2010). Surgical excision is the treatment of choice for cystic hygromas; however, other options include sclerotherapy, drainage, radio-frequency ablation, or cauterization (YANI et al., 2023). Its incidence is approximately 1/6000 live births. In animals, sporadic cases of cystic hygroma have been reported in sheep (SANTOS et al., 2014), cattle (WANKE et al., 1990), swine (HOLLEBOOM et al., 2022), horses (ZUVIRIA et al., 2022) and dogs (LODZINSKA et al., 2019). The precise embryonic origin of cystic hygroma in humans and animals is still uncertain. However, it was associated with chromosomal abnormalities particularly Trisomy 21, 13, 18 and several genetic syndromes. It can be inherited in autosomal recessive and autosomal dominant manner in humans (LETKO et *al*., 2021; ZUVIRIA et *al*., 2022). One report has suggested the existence of an autosomal recessive type of cystic hygroma in cattle (WANKE et al., 1990). A little knowledge is present about lymphatic malformations in ruminants and remains poorly understood condition. The aim of this work is to present the first case report of cystic hygroma in a 2-month-old mal crossbred goat-kid.

Case History

In February 2020, a two -months-old, farm-bred, male crossbred goat-kid (Arabia x Saanen) weighing about 9 kg was presented with a history of swelling on the left side of the body of the mandible are associated with facial asymmetry (Fig. 1). According to the owner, there is no farm history of previous similar cases. The swelling was approximately 5 x 3 cm in size and it did not interfere with the viability of the animal. It was present since birth and has slowly decreased in size. The goat-kid was in good body condition and its weight is similar to other animals of the same age. The body temperature, respiration rate, and heart rate were within the normal limits. 62

Fig. 1. Clinical presentation of the cystic hygroma, a 5 × 3 cm mass in on the left side of the mandible region of a two-month-old crossbred goat-kid 66

Clinical examination revealed a solitary, soft to tender in consistency, painless, compressible cystic mass with no changes in the overlying skin. The facial nerve function was normal. The left pre-scapular lymph node was not deemed abnormal by palpation. Ultrasonography examination, using an ultrasound machine (Kaixin® KX 5500; Xuzhou Kaixin Electronic Instrument Co., Ltd., Xuzhou, China) with 5.0 MHz frequency sector transducer, confirmed a fluid-filled multiloculated anechoic cystic mass with internal septations. Fine-needle aspiration was performed under aseptic precautions, demonstrating that the swelling contained serous cloudy fluid compatible with chyle fluid (Fig. 2). Biochemical analysis revealed a fluid pH of 6.0, density of 1.025, total protein of 0.8 g/dL, albumin of 0.6 g/dL, LDH of 7 UI/L, glucose of 2 mg/dL, and no trace of nitrite. The cytological investigation did not show any malignant cells but revealed nucleated cells consisting predominantly of lymphocytes and a small amount of red blood cells, which can be attributed to the process.

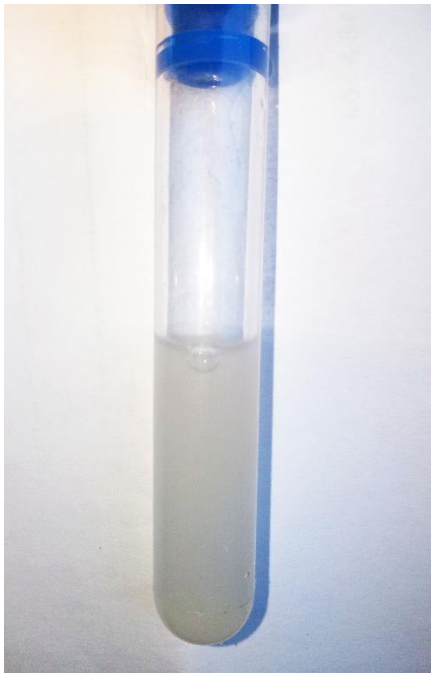

Fig. 2. Aspirated serous cloudy fluid from the cystic mass.

2. DISCUSSION

Cystic hygroma is an uncommon congenital abnormality of the lymphatic vessels that affect humans and various animals. Until now, this congenital defect in goats has not yet been described in the literature. In our study, we evidenced cystic hygroma in a 2-month-old cross- breed goat-kid. The clinical features, findings on ultrasonography and cytological examination were compatible with cystic hygroma. The etiology of cystic hygroma is unclear. During the gestation period, the lymphatic system normally develops at fifth week. The developing embryo has six lymphatic sacs located in different regions of the body. These lymphatic sacs are two jugular sacs, two iliac, one is cisterna chyli and the last one is retroperitoneal (CHOWDHURY et al., 2023). Theories in the literature suggest that a common cause of cystic lymphangioma is the failure of jugular lymphatic sacs to drain into the internal jugular vein. This causes dilatation of the jugular lymphatic sac, which can lead to the development of single or numerous fluid-filled lesions in various locations, especially in the neck (AL-ZAMZAMI, 2022). Cystic hygroma is a type of lymphangioma

with one or multiple cysticlesions. The classification of lymphangioma proposed by the International Society for the Study of Vascular Anomalies (ISSVA) is based on clinical features and complications (Liu et *al*., 2021). According to themicroscopic aspect, lymphangiomas are subdivided into capillary, cavernous, and cystic. They can also be classified into mixed, macrocystic (diameter > 1 cm) and microcystic lymphangiomas (diameter <1 cm), according to the size of the cysts. Mixed lymphangiomas have cysts of varying sizes, ranging from over 2 cm to under 2 cm (Mirza et *al*., 2010). Currently, there is no standard classification of vascular malformations in veterinary medicine due to their infrequency (DRIESSEN et al., 2020).

Very few clinical cases of lymphatic malformations have been reported in ruminants. These anomalies were described in the neck of two calves (WANKE et al., 1990) and one lamb (SANTOS et al., 2014). However, in the present case, the cyst was found in the mandible area. To our knowledge, this rare location of lymphatic malformations in animals has not been reported in the veterinary literature. In humans, cystic lymphangiomas are ubiquitously located but the common locations are cervico- facial regions (~75%), probably because of the highly dense lymphatic system in this area (BHATNAGAR et al., 2020). They are most often unilateral and frequently located on the left side, as in our case. In the neck, they are especially found in the posterior triangle**.** Other associated areas are parotid and submandibular regions (AL-ZAMZAMI, 2022). Although, 60% of cystic lymphangiomas are identified at birth as an asymptomatic mass and about 90% of the lesions are diagnosed before the age of 2 years. It is less commonly observed during adulthood and has no gender predilection (Yarso et *al*., 2023)**.** In this case report, the goat-kid was two months old and had the anomaly since birth, which indicates a congenital malformation. Clinically, cystic hygroma commonly presents as diffuse, soft, slow-growing, painless and movable cystic mass as reported in our case. Cysts vary in size from a few millimetres to more than several centimetres in diameter. Our case showed a macrocystic lymphangioma of about 5 cm in diameter. In order to confirm this diagnosis, ultrasonographic examination and fine- needle aspiration (FNA) for cytology and biochemical testing were performed. Ultrasonography is a widely available and non-invasive diagnostic tool to consider as a first choice for investigating head and neck lumps (CHOWDHURY et al., 2023). It is particularly performed to distinguish between cystic and solid mass and its relationship to the surrounding structures (PALLAGATTI et al., 2012). An ultrasound of the lesion described herein showed a well-defined, multi-loculated cystic with internal septations without significant vascularization, as reported in the previous study (HOLLEBOOM et al., 2022). Usually, cystic lymphangiomas can contain different types of fluids, such as milky, watery, serous, serosanguinous or straw-coloured (CHOWDHURY et al., 2023). Fine-needle aspiration revealed serous cloudy fluid consistent with lymphatic fluid as confirmed by biochemical testing. Cytological examination displays the presence of a massive infiltrate of lymphoid cells and occasional red blood cells, but no malignant cells. These findings were similar to those of previous studies in humans and animals (MIRZA et al., 2010; LETKO et al., 2021). In our study, spontaneous regression of cystic hygromas was observed without any treatment. This is in concordance with spontaneous regression in humans that can occur only in macrocystic and mixed lymphangiomas with a rate of up to 16 %, but not in the microcystic variant. Macrocystic and mixed lymphangiomas differ from microcystic lymphangiomas in the balance of lymph fluid in-flow and out-flow (KATO et al., 2017). The present clinical case represents the first description of lymphatic malformation in goats. Cystic hygroma should be included by veterinarians in the differential diagnosis of any cystic mandibular swelling, especially in goat-kids.

Availability of Data and Materials

The datasets during and/or analyzed during the current study available from the

corresponding author on reasonable request.

Acknowledgments

We thank Dr. Mohamed Ezzine Zebiri for his expertise in clinical examination of the animals.

Funding Support

There is no funding source.

Competing Interests

The authors declare that they have no known competing financial interests or personal relationships that could have appeared to influence the work reported in this paper.

Author Contribution

NS, NM, NAI, NB, DK: Conceptualization, Methodology, Validation, Formal Analysis, Investigation, Writing – Original Draft, Writing – Review & Editing.

3. REFERENCES

1. L-ZAMZAMI, A. A. (2022): Cervical Cystic Hygroma in Infant among Yemenis, A Case
2. Report. J Dent Rep. 3(1), 1-8.
3. BHATNAGAR, A., V.D. UPADHYAYA, R. YADAV, B. KUMAR (2020): Management of macrocystic lymphatic malformation at uncommon site with aqueous bleomycin sclerotherapy. National Journal of Maxillofacial Surgery. 11(2), 193.
4. CHOWDHURY, M.A.T., M.Z. AHMED, N. HAIDER, A.F. AHMED, N. HAQUE, N.
5. AKTAR, M. MAGDUM (2023): Unusual Presentation of a Large Multi-Septated Cystic
6. Hygroma: A Case Report and Comprehensive Literature Review. Surgical
7. Science. 14(7), 486-495.
8. DRIESSEN, F., T. CUSHING, S.J. BAINES (2020): Retroperitoneal lymphatic malformation in a dog. Acta Veterinaria Scandinavica. 62, 1-7.
9. 171 ERÖKSÜZ Y., Emine ÜNSALDI, Murat TANRISEVER, Canan AKDENİZ İNCİLİ, Hatice ERÖKSÜZ. Subcutaneous Cavernous Cervicofacial Lymphangioma and It"s Surgical Treatment in a Calf. Kafkas Univ Vet Fak Derg, 27 (6): 811-814, 2021. DOI:
10. 175 10.9775/kvfd.2021.26330 176
11. HOLLEBOOM, R.G., D. BOMBOSCH, M.M.H. WISPELS, G. GIGLIA, T.J. TOBIAS
12. (2022) Cystic hygroma in a female suckling pig: a case report. Porcine Health
13. Management. 8(1), 40 KATO, M., S. WATANABE, R. KATO, H. KAWASHIMA, T. IIDA, A. WATANABE (2017): Spontaneous regression of lymphangiomas in a single center over 34 years. Plastic and Reconstructive Surgery Global Open. 5(9).
14. LETKO, A., A.M. SCHAUER, M.F. DERKS, L. GRAU-ROMA, C. DRÖGEMÜLLER, A. GRAHOFER (2021): Phenotypic and genomic analysis of cystic hygroma in
15. 185 pigs. Genes. 12(2), 207.
16. LIU, X., C. CHENG, K. CHEN, Y. WU, Z. WU (2021): Recent progress in lymphangioma. Frontiers in Pediatrics. 9, 735832.
17. LODZINSKA, J., M. GRACZ, M. SILKSTONE, E. MUNRO (2019): Presumed congenital lymphangioma in a dog: ultrasonographic and computed tomography findings, presentation and treatment. Veterinary Record Case Reports. 7(4), e000901.
18. MIRZA, B., L. IJAZ, M. SALEEM, M. SHARIF, A. SHEIKH (2010): Cystic hygroma: an overview. Journal of cutaneous and aesthetic surgery. 3(3), 139.
19. PALLAGATTI, S., S. SHEIKH, N. PURI, A. MITTAL, B. SINGH (2012): To evaluate the efficacy of ultrasonography compared to clinical diagnosis, radiography and histopathological findings in the diagnosis of maxillofacial swellings. European

Journal of radiology. 81(8), 1821-1827 SANTOS, N., L.M. FERRER, D. LACASTA, J.J. RAMOS, A. UIXERA, M. CLIMENT, L.

20. FIGUERAS (2014): Lamb congenital lymphatic malformation-a case report.
21. ActaVeterinaria Brno, 83(3), 239-241.
22. SHRESTHA, S. D., G.B. CHALISE, D. BISHANKHA, T.B. PANTHA, D. GAUTAM (2022): Giant cystic hygroma in an adult male; A case report. Otolaryngology Case
23. Reports. 25, 100492.
24. WANKE, R., O. DISTL, P. SCHMIDT, W. HERMANNS (1990): Cervical hygroma I the calf as an absolute cause of dystocia. An unusual malformation possibly of hereditary genesis in Braunvieh x Brown-Swiss breeds. Berliner und Munchener Tierarztliche Wochenschrift. 103(1), 18-22.
25. YANI, A., R.P. ULIMA, K. RINALDHY, T.H. RAHAYATRI, R. AMALIAH, W.
26. AYUDYASARI (2023): Management of prenatally diagnosed multiple giant cystic lymphangioma with combination of surgery and intralesional bleomycin injection: A case report. International Journal of Surgery Case Reports. 108391.
27. YARSO, K.Y., M. BELLYNDA, K. MUYASARAH, F. MUHAMMAD (2023): Ethanol ablation for management of large cyst hygromacolli in adult: A case report. International Journal of Surgery Case Reports. 105, 108092.
28. ZUVIRIA, S.M., M. CIURKIEWICZ, P. WOHLSEIN, G. MADARIAGA, G. ZUCCOLILLI (2022): First Description of Fetal Cystic Hygroma Associated with Early Equine Pregnancy Loss. Journal of equine veterinary science. 119, 104148. 217