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Giant Cervicofacial Haemolymphangioma: Case Report

Rim Adyel[1*], Ibtissam Naanani[1], Zineb Kihal[1], Salwa Hafoud[1], Daoud Bentaleb[1], Dallal Laoudiyi[1], Kamelia Chbani[1], Siham Salam[1]

[1]Department of Pediatric Radiology, A.Harouchi Mother and Child Hospital, CHU Ibn Rochd, Casablanca, Morocco

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***Corresponding author:** Rim Adyel
Department of Pediatric Radiology, A.Harouchi Mother and Child Hospital, CHU Ibn Rochd, Casablanca, Morocco

Abstract **Case Report**

In this article, we report a cervicofacial haemolymphangioma case whose particularity lies in its significant locoregional extension to the deep spaces of the face and the floor of the mouth, which makes its therapeutic management tricky, requiring a multidisciplinary approach despite its benign nature, the aim being a complete surgical cure to avoid the risk of recurrence. We also highlight the radiological features arguing in favor of the diagnosis.

Keywords: Haemolymphangioma, Cervicofacial, Infant, Multidisciplinary approach, Radiological features.

INTRODUCTION

Haemolymphangioma is a rare entity corresponding to the lesional association of lymphatic malformations and vascular tumors of the haemangioma type.

In this article, we report a rare case of cervicofacial haemolymphangioma in a 2-year-old infant.

OBSERVATION

A 2-year-old infant presented with a left cervicofacial swelling that had been present since birth and had progressively increased in size.

The swelling was soft, painless, and without inflammatory signs.

An ultrasound scan revealed a cystic mass, lateral cervical on the left, anechoic in content, multi-loculated, with multiple septae, pushing back the superficial lobe of the homolateral submandibular gland and extending to the overlying left jugal soft tissue, pushing back the superficial lobe of the parotid gland.

A cervicofacial lymphangioma was suggested.
A cervicofacial MRI was performed under sedation to assess the extent of the lesion and better characterize it.

The MRI revealed a voluminous left cervicofacial formation with a dual cystic and solid contingent. The cystic contingent was predominant, multi-located in T2 hypersignal, and T1 hyposignal, with fine septa, enhanced after gadolinium injection.

Peripheral contrast was also noted.

The solid contingent was represented by three formations with T1 iso signal, intermediate T2 hypersignal, more marked on the T2 sequence with suppression of fat, and intensely enhanced after injection of gadolinium on the dynamic TWIST sequences.

This formation extended into the deep spaces of the left face (parotid cavity, pterygoid cavity, pre, and retrostylial spaces) and the floor of the mouth without crossing the midline.

All the radiological findings led us to suspect a left cervicofacial haemolymphangioma of uncomplicated appearance.

Histopathology confirmed the diagnosis.

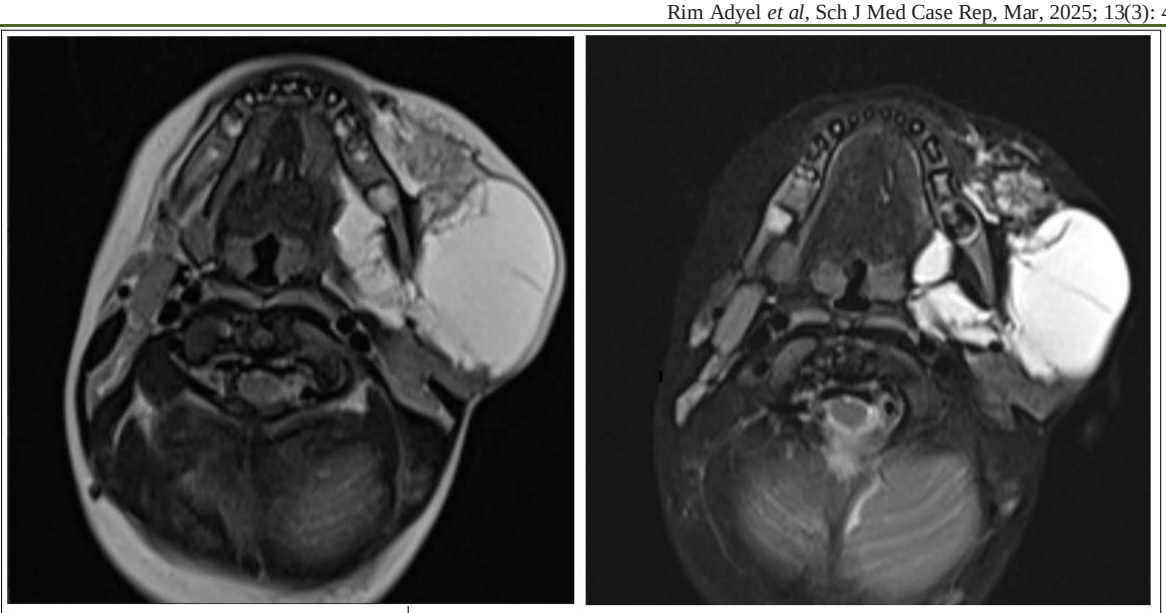

Figure 1: Axial-section T2-weighted MRI images without and with fat saturation showing a multi-loculated, solid cystic left jugal formation extending through the free edge of the mylohyoid muscle at the level of the floor of the mouth.

Note the increased signal of the solid portion on the fat-saturated T2-weighted sequence.

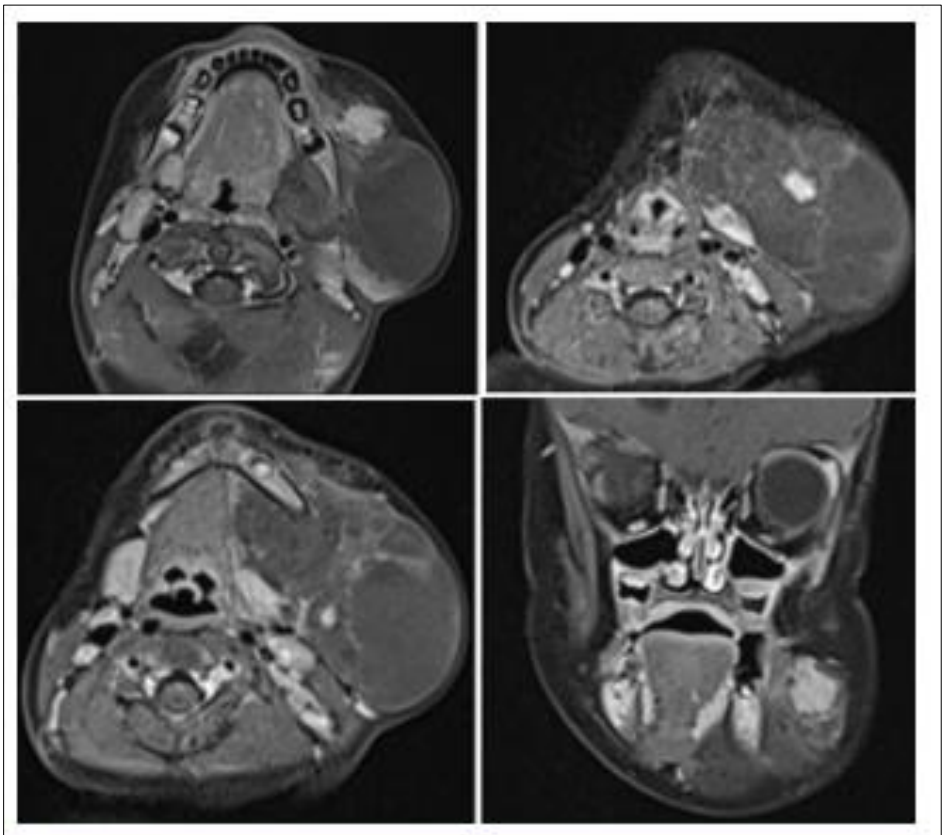

Figure 2: T1-weighted MRI images after gadolinium injection and fat saturation in axial and coronal sections, showing enhancement of the periphery of the solid cystic lesion, the septa and the solid portions.

DISCUSSION

Lymphangiomas are lymphatic vascular malformations that include several histopathological variants. We distinguish cystic, capillary, cavernous lymphangiomas and haemolymphangiomas corresponding to a lesional association comprising a lymphatic contingent and a vascular tumor contingent corresponding to a haemangioma [1].

Haemolymphangioma is a rare entity, most often congenital; a few acquired post-traumatic or post-surgical forms also exist but are even more anecdotal [2, 3].

It is most often diagnosed in children under the age of 2, with a clear predominance of females [3].

The pathophysiology of these malformations is still poorly understood.

Several risk factors and hypotheses have been reported for congenital forms, all of which have in common a state of fetal hypoxia leading to overexposure of the fetus to VEGF [4].

Haemolymphangiomas are most often found in the anterolateral or posterior part of the neck and in the face, other heterogeneous and ubiquitous locations have also been reported in the literature, and multifocal forms have been reported [5, 6].

Clinically, haemolymphangioma will appear as a soft, swelling that is mobile to the various planes, painless, and without inflammatory signs, apart from possible complications such as superinfection or rupture, whether hemorrhagic or not.

Ultrasound is the examination of the first choice because it is accessible, inexpensive, and safe due to its non-irradiating nature.

It will reveal the cystic nature of the mass, which will be multi-loculated and the site of septations, with fleshy portions, highly vascularised on color Doppler, corresponding to the haemangiomatous contingent.

The deep solid contingent may be missed, and color Doppler analysis may be impaired.

Thyroglossal duct cysts, epidermoid cysts, dermoid cysts, and branchial cysts are the main differential diagnoses.

Due to its high resolution in soft tissue contrast, MRI is the reference examination to be carried out pre-operatively.

Due to its volume rendering in all three planes, MRI can provide an exhaustive analysis of the lesion's relationship with neighboring organs, and vascular and neural structures.

It will also provide a precise assessment of the lesion depth.

CT is an irradiating examination, and its soft tissue contrast performance is inferior to MRI, so it is not the preferred examination for diagnosing such lesions.

The treatment of choice for haemolymphangiomas remains surgery with the aim of complete resection, as cases of recurrence have been reported [7, 8].

However, surgery can be invasive and debilitating if the haemolymphangioma has spread locally or regionally. Administration of a sclerosing agent, or embolization in acute active bleeding cases, may be carried out as a first step [9].

CONCLUSION

Haemolymphangioma is a rare entity, despite its benign nature, it remains difficult to manage in case of wide regional spread. Radiology has a central role in assessing the spread of the lesion and thus determining the lesion resectability.

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