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Paediatric Cervical Cystic Hygroma, Managed Successfully at Temeke Regional Referral Hospital; Case Report and Literature Review.

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Abstract

Cystic hygroma is a benign congenital malformation associated with the lymphatic system. It occurs as result of obstruction or sequestration of developing lymphatic vessels at the junction with internal jugular vein. Commonly presents in infancy and childhood, rarely seen in adults. They present as painless large, diffuse, multi-component, non-tender, non-compressible swellings; located most on the cervico-facial region. Clinical manifestations and radiograph examinations are useful to do accurate diagnosis of this lesion. CT-Scan and MRI are useful particularly when surgical management is contemplated. Many treatment modalities are reported. Surgical treatment is mainstay treatment to excise the enlarging lesion, relief patient symptoms and exclude other pathologies. In some cases when surgical complete excision is impossible, sclerotherapy, radiotherapy, radio-frequency ablation and laser excision are reported as an alternative treatment. We report a case of cervical cystic hygroma which was managed successfully in resource limited area in Tanzania.

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Introduction

Lymphangioma was originally reported by Reden Backer in 1828, and "cystic hygroma" name was first given by Wernker in 1834 [1]. It is a benign malformation of lymphatic vessels which usually occurs when the lymphatic system fails to communicate with the normal internal jugular vein. It is a rare congenital abnormality with an estimated incidence of 1 case per 6000-16000 live births [1,2]. In most cases these malformations are visible at birth with approximately 80% to 90% becoming apparent by age of two [3,4]. Commonly cystic hygroma is a disease of head and neck, but can also affect other areas like the axilla, mediastinum, groin, tongue, and certain organs like the liver, spleen, kidney, and bowels. However, it rarely involves limbs. It predominantly affects children under 2yrs old, with rare occurrences in adults. It affects both genders and there are no reported cases of malignant transformation [5-7].

Case Report

A 5-years-old male patient presented at the surgical outpatient clinic with complaints of a painless swelling on the right side of the neck, which had been present since birth and seemed to gradually increasing in size over time. There was no supportive family, medical and surgical history. Physical examination determined a mass lesion in the right cervical region reaching approximately 6 cm in size, slightly mobile and pressurized, with no changes in skin color or temperature, leading to postural impairment of the neck (Figure 1). Routine laboratory results were normal. B-mode USG examination was directed to the cervical region. A complex cystic lesion was observed. Upon carefully pre-operative stabilization surgical excision was done and the gross mass was sent for histological study (figure 2,3). Microscopic evaluation revealed variable sized, cystically dilated lymphatic channels, some filled with lymphocytes. Hence, the final diagnosis of lymphangioma was confirmed (figure 4 and 5) post-operative course was uneventful and he was discharged home on fourth post-operative day. Outpatient follow up was uneventful as well so he was discharged from outpatient clinic when the wound healed completely (figure 6).

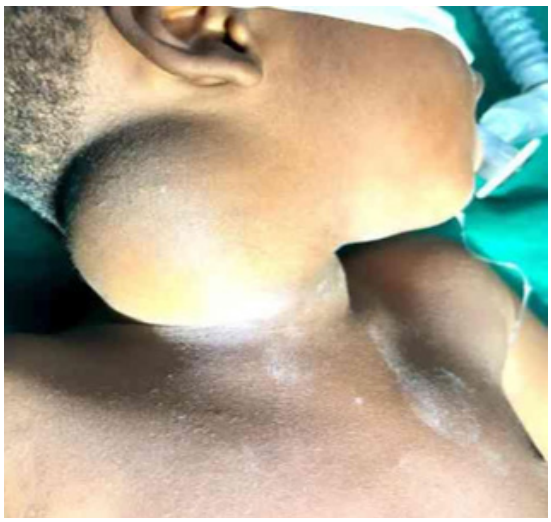

Figure 1: Pre-operative photograph showing a large right cervical swelling causing postural impairment of the neck

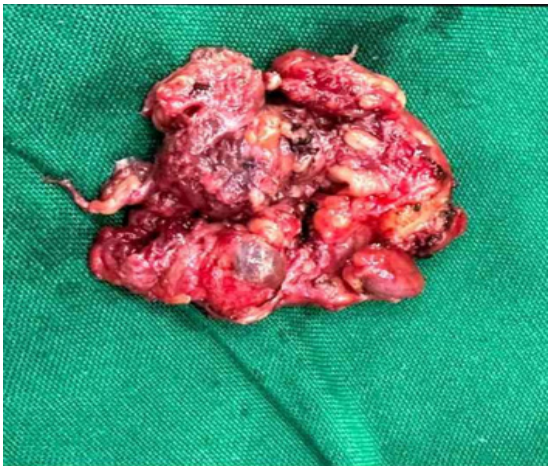

Figure 2: Gross specimen of the excised multi-cystic mass.

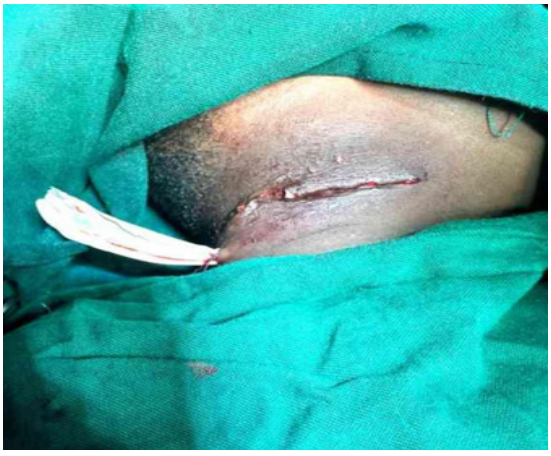

Figure 3: Immediate post-operative wound after layered closure

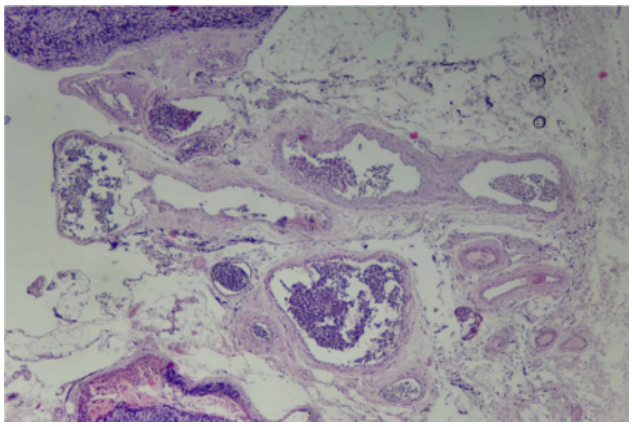

Figure 4: HE stains in 10× view. Multiple cystically dilated lymphatic spaces with intralumenal lymphoid cell aggregations

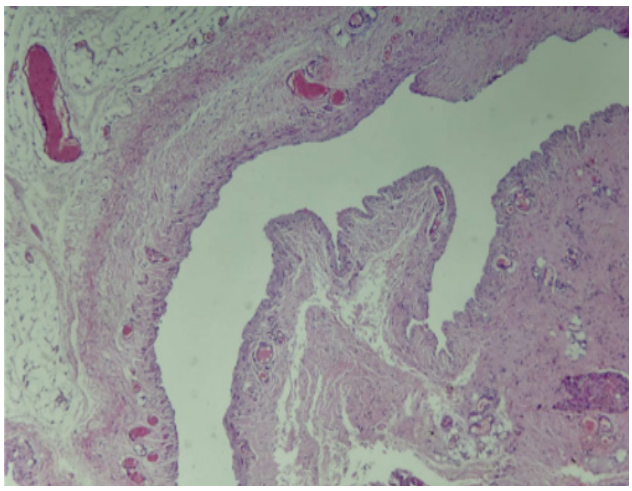

Figure 5: HE stains in 10× view. Large, irregular cystically dilated lymphatic vessel lined by flat endothelial cells and surrounded by smooth muscle layer

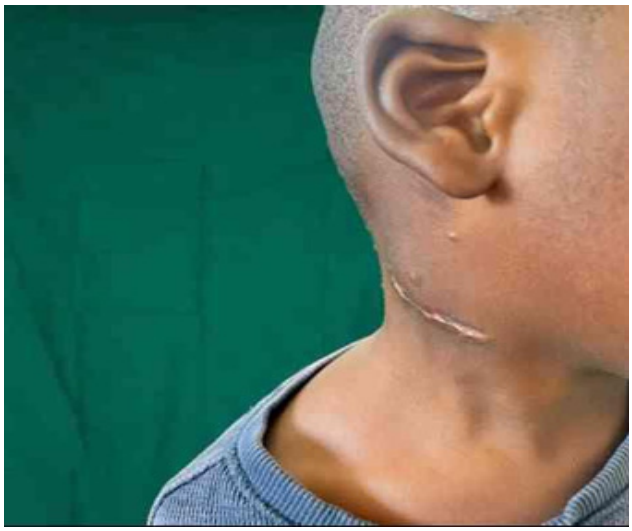

Figure 6: Late post-operative appearance showing well-healed scar.

Discussion

Cystic hygroma (lymphangioma) is a rarely seen congenital malformation of the lymphatic system, which most often involves the head and neck region. Half of cases are determined at birth and most are diagnosed under the age of two years. The most common presentation is usually a painless ill-defined swelling or mass. It never involves skin and it shows positive trans-illumination test. The common location of cystic hygroma is in the anterior and posterior triangles of the neck, with the posterior triangle of the neck being the most common site of occurrence [8,9]. The case narrated here had a mass lesion in the posterior triangle of the neck since birth and diagnosis was made late may be due to financial constraints as the child was not insured.

Radiologic investigations and biopsy are the cornerstone of the diagnosis of cystic hygroma. Ultrasonography is the least invasive option and can effectively show the relationship between the cyst and surrounding structures [10]. Preliminary diagnosis of the present case was done by clinical examination and neck ultrasound. However, after surgical excision histopathological examination confirmed the diagnosis (figs 4 and 5)The mainstay management of cystic hygroma is surgical excision, in order to excise the enlarged lesion, relief the patient symptoms and exclude other pathologies. Minimally invasive modalities include image guided aspiration and intralesional sclerotherapy (using bleomycin, OK-432, doxycycline, acetic acid, alcohol, hypertonic saline) and serial laser therapy has been reported as well [1,11]. Our reported case was managed by surgical excision as clinical assessment revealed possibility of complete open surgery. More over our hospital do not have the facilities for minimally invasive surgery for this particular pathology.

Conclusion

In summary, Cystic hygroma is a rarely seen congenital malformation of lymphatic systeam with variable presentations. The management of these lesions is complex and multidisciplinary, and depends on expertise and experience. The mainstay of treatment is complete excision. Complete excision, although not feasible in all lesions, has shown to result in the least recurrence rate among all treatment modalities. This case report provides valuable insights for both clinical practice and further research on cystic hygromas.

Declaration of Patient Consent

The authors certify that they have obtained all appropriate patient consent forms. In the form the patient(s) has/have given his/her/their consent for his/her/their images and other clinical information to be reported in the journal. The patients understand that their names and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

Conflicts of Interest

There are no conflicts of interest.

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