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Case Report

Cystic hygroma presenting as paediatric stridor

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ABSTRACT

Cystic hygroma, a fluid-filled lymphatic lesion, commonly presents in the head and neck but can occur anywhere. Paediatric stridor is a critical emergency demanding precise diagnosis and immediate airway intervention. We report the case of a 2-year-old male child presenting with a 7-day history of dysphagia and stridor, accompanied by high-grade fever, left neck swelling, and torticollis. Examination revealed a posterior pharyngeal wall bulge. Initial impression was retropharyngeal abscess, necessitating emergency tracheostomy for airway compromise. Midline aspiration yielded straw-coloured fluid, and histopathological examination confirmed cystic hygroma. Postoperative MRI revealed a large (9.6 x 7.7 x 3.4 cm) hyperintense cystic lesion with septations, extending from the oropharynx to the upper mediastinum, encasing vital structures. The patient subsequently underwent four cycles of sclerotherapy with intra-lesional bleomycin. This case highlights the challenges in diagnosing atypical causes of paediatric stridor. Sclerotherapy with bleomycin proved to be a highly effective treatment for this extensive cystic hygroma, demonstrating its superiority over surgical options due to lower associated risks and favourable outcomes.

Keywords: Cystic hygroma, Tracheostomy, Sclerotherapy, Bleomycin

INTRODUCTION

First described by Wernher in 1843, cystic hygroma is a cystic lymphatic lesion that can affect any anatomic subsite in the human body.[1] It is a fluid-filled sac resulting from a blockage in the lymphatic system. Although it can be located anywhere in the body, it is most commonly found in the neck or head region.[2] Symptoms vary depending on its size and specific location, with the potential to cause complications involving nearby structures or organs.[3]

Pediatric stridor constitutes a medical emergency requiring precise diagnosis and urgent attention for both airway management and definitive treatment. Common non-infectious causes include laryngomalacia, laryngeal web, subglottic stenosis, and vocal cord dysfunction. Infectious etiologies encompass croup, retropharyngeal abscess, and epiglottitis.[4] We present a case of cystic hygroma in a male child who presented with stridor.

CASE REPORT

A 2-year-old male child presented with a 7-day history of difficulty in swallowing accompanied by stridor. He also had associated high-grade fever, swelling on the left side of the neck, and torticollis. On examination, the child was irritable. Mouth opening was adequate. Oropharyngeal examination, performed with a headlight and tongue depressor, revealed a bulge in the posterior pharyngeal wall. Raised leukocyte counts were noted. An X-ray of the neck (lateral view) showed soft tissue swelling in the

retropharyngeal region. These symptoms and signs led to an initial impression of a retropharyngeal abscess.

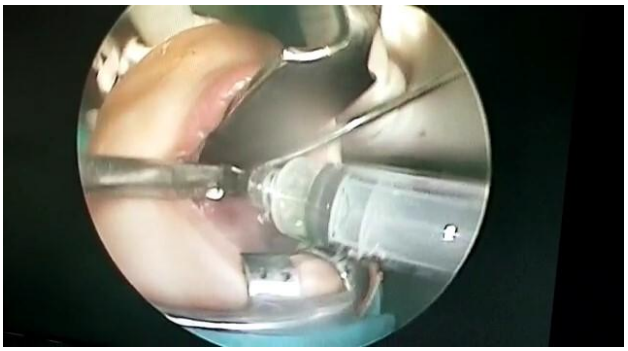

Figure 1: Intra-lesional injection of bleomycin into the cystic hygroma.

Consequently, the patient was admitted, and broad-spectrum antibiotics were initiated. Due to significant airway compromise, intubation was performed with difficulty using a size 3 endotracheal tube. A tracheostomy was then carried out, and a size 4.5 tracheostomy tube was inserted. The mouth was opened with a Boyle and Davis mouth gag, and midline aspiration was performed, yielding thick, straw-colored fluid. A tissue sample was obtained for histopathological examination, which subsequently revealed cystic hygroma.

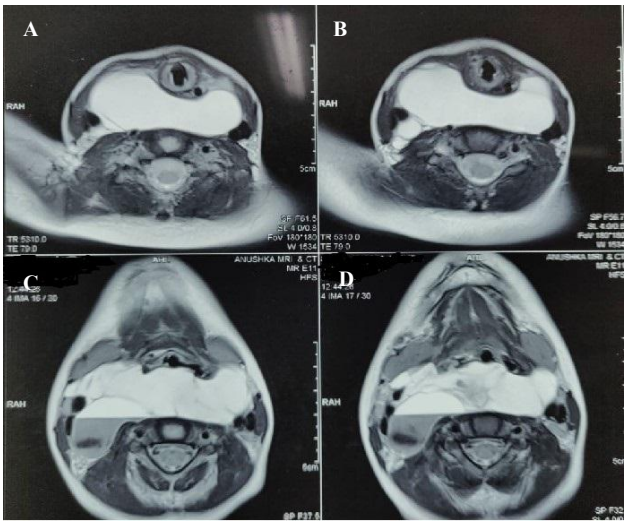

Figure 2 (A-D): Axial MRI scan of the neck demonstrating the extensive cystic hygroma. This hyperintense lesion, measuring 9.6×7.7×3.4 cm and featuring multiple thin walls and septations.

A postoperative MRI scan was conducted, which showed a cystic lesion measuring 9.6×7.7×3.4 cm. The lesion appeared hyperintense with multiple thin walls and septations, extending from the oropharynx up to the upper mediastinum, encasing the great vessels, airway, and esophagus. The patient was subsequently scheduled for sclerotherapy with bleomycin.

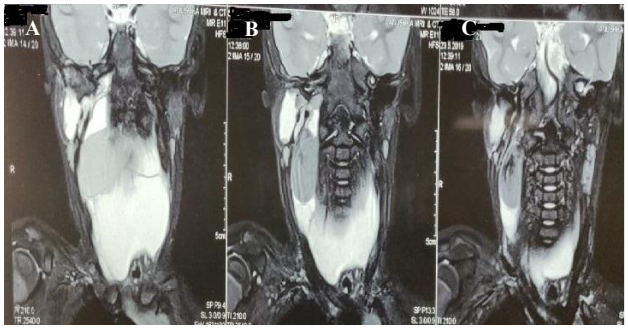

Figure 3 (A-C): Coronal MRI scan of the neck demonstrating the extensive cystic hygroma, measuring 9.6×7.7×3.4 cm and featuring multiple thin walls and septations, extends from the oropharynx down to the upper mediastinum.

Four cycles of sclerosant were administered, with one cycle given each month. The child remained tracheostomized during this period. Significant reduction in neck swelling was observed. After the 4th cycle, a repeat MRI was performed to assess the status of the cystic hygroma. The child showed good improvement and was comfortable. Subsequently, the patient was decannulated. He was kept on follow-up and advised to report immediately in case of recurrent stridor. A review after 6 months was planned to check the status of the cystic swelling.

DISCUSSION

Cystic hygroma originates in embryonal life, leading to a failure of communication and lymph drainage into the venous system.[5] Also known as cystic lymphangioma and macrocystic lymphatic malformation, it is often a congenital lymphatic lesion that can arise anywhere. However, the majority (45-52%) occur in the lymphatic-rich regions of the head and neck, classically found in the left posterior triangle of the neck and axillae. It can also present in the mediastinum, groin, and retroperitoneum.[6]

The indications for treatment include recurrent bouts of infection, respiratory distress, dysphagia, hemorrhage, sudden increase in lesion size, lymph-discharging sinus, and disfigurement. The respiratory distress can be severe, often necessitating a tracheostomy due to complete or significant laryngeal or tracheal compressions (as observed in the present case).

Management options include surgical excision and sclerotherapy. Surgical excision of complex cystic hygromas, especially those involving deep and vital structures, presents significant challenges. Meticulous care must be exercised to avoid perioperative complications. Possible complications during surgery include damage to the facial artery, carotid vessels, internal jugular vessels, thoracic duct, and pleura, as well as incomplete excision due to infiltration into surrounding structures.

Historically, sclerotherapy agents such as boiling water, quinine, sodium morrhuate, urethane, iodine tincture, doxycycline, and nitromin have shown low success rates and frequent complications.[7] Bleomycin, a chemotherapeutic agent typically used for several malignancies, was first utilized by Yura et al as an intra-lesional sclerosant. Sclerotherapy with intra-lesional bleomycin has since been tried, and various case reports have documented favorable responses to this therapy, a finding our case experience substantiates. Complications associated with bleomycin sclerotherapy include discoloration of the injection site, sudden increase in the size of the cystic hygroma, fever, vomiting, cellulitis, interstitial pneumonia, and pulmonary fibrosis.[8]

Sclerotherapy appears to be a superior treatment option for large cystic hygromas due to its lower associated risks compared to open surgery.

CONCLUSION

This case report underscores the critical importance of considering rare etiologies, such as cystic hygroma, in the differential diagnosis of acute paediatric stridor, particularly when initial presentations mimic more common conditions like retropharyngeal abscess. Despite the challenging initial clinical picture, which necessitated emergency tracheostomy for airway management, subsequent imaging and histopathological confirmation guided the definitive treatment.

Our experience corroborates the growing evidence supporting sclerotherapy with intra-lesional bleomycin as a highly effective and safer alternative to extensive surgical excision for large and complex cystic hygromas involving vital structures, leading to significant regression of the lesion and successful decannulation in this 2-year-old child. This case highlights the need for a comprehensive diagnostic approach and individualized management strategy to achieve optimal outcomes in such rare and challenging paediatric presentations.

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