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A rare case of cervical cystic lymphangioma in a preschool child: Timely diagnosis and treatment

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Introduction: *Cystic lymphangiomas are rare congenital lymphatic malformations that usually present before two years of age, most commonly involving the cervical region. Reporting this case is important due to its atypical age of presentation, deep cervical involvement, and successful non-surgical management.*

Case presentation: *We report a five-year-old boy who presented with an eight-day history of a progressively enlarging, painless right-sided neck swelling. Clinical examination revealed a well-defined, mobile cervical mass. Ultrasound and magnetic resonance imaging demonstrated a multilocular cystic lesion consistent with cystic lymphangioma. The child was treated with a single intralesional dose of bleomycin, resulting in significant regression of the swelling. He has remained asymptomatic with no evidence of recurrence during two years of follow-up.*

Conclusions: *This case highlights the need to maintain a broad differential diagnosis for paediatric neck swellings, especially in atypical presentations. Early imaging and prompt initiation of sclerotherapy resulted in complete regression and prevention of potential complications. The excellent response to a single intra-lesional bleomycin injection underscores its effectiveness as a minimally invasive treatment, while continued follow-up remains essential due to the risk of recurrence.*

Keywords: LYMPHANGIOMA, CYSTIC; SCLEROTHERAPY; CHILD, PRESCHOOL; LYMPHATIC VESSELS

INTRODUCTION

Lymphangiomas are rare, benign congenital malformations of the lymphatic system resulting from abnormal embryologic development of lymphatic channels. They occur in approximately 1 in 2 000 to 6 000 live births worldwide (1). Although comprehensive national epidemiological data from India remain limited, available Indian paediatric case series demonstrate a similar age distribution and clinical profile (2). These lesions arise from sequestration or failure of communication between primitive lymphatic sacs and the central lymphatic system, leading to progressive dilatation and eventual cyst formation (3, 4).

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The cervical region is the most common site of involvement, accounting for approximately 75% of cases, followed by the axilla (15%) and less commonly the mediastinum, abdomen, and other anatomical regions (2, 5). Nearly 50% are present at birth, and about 80–90% are diagnosed by two years of age (6, 7). Cervical lymphangiomas typically present as painless, soft, and compressible swellings that may transilluminate; however, larger or deep-seated lesions can result in airway compromise, dysphagia, or neurovascular compression depending on their location and extent (8, 9). In view of the broad differential diagnosis of paediatric neck masses – including branchial cleft cysts, thyroglossal duct cysts, reactive lymphadenitis, and tubercular lymphadenitis – careful and systematic evaluation is imperative to ensure an accurate diagnosis.

Although diagnosis of lymphangiomas is primarily clinical, imaging is essential for confirmation and treatment planning. Ultrasonography serves as the initial investigation of choice, effectively differentiating cystic from solid lesions and evaluating internal septations (10). Magnetic resonance imaging (MRI) is considered the gold standard, accurately delineating lesion extent, internal architecture, and relationships with adjacent neurovascular structures, thereby guiding management and excluding mediastinal or intracranial extension (11).

Management of lymphangiomas has advanced considerably over time. Although surgical excision was traditionally regarded as definitive, it carries risks of neurovascular injury, incomplete resection, and recurrence, particularly in multiloculated or infiltrative lesions (7). Over recent decades, sclerotherapy has emerged as a safe and effective minimally invasive alternative, especially for macrocystic lymphangiomas, with agents such as bleomycin, OK-432, and doxycycline demonstrating favorable outcomes, reduced morbidity, and superior cosmetic results compared to surgery (12,13). Notably, intralesional bleomycin has achieved high rates of lesion regression with minimal adverse effects when administered appropriately (14). Despite these advances, delayed presentation beyond early childhood remains rare and can present significant diagnostic challenges. We report a rare case of cervical cystic lymphangioma in a five-year-old child successfully managed with timely sclerotherapy with bleomycin.

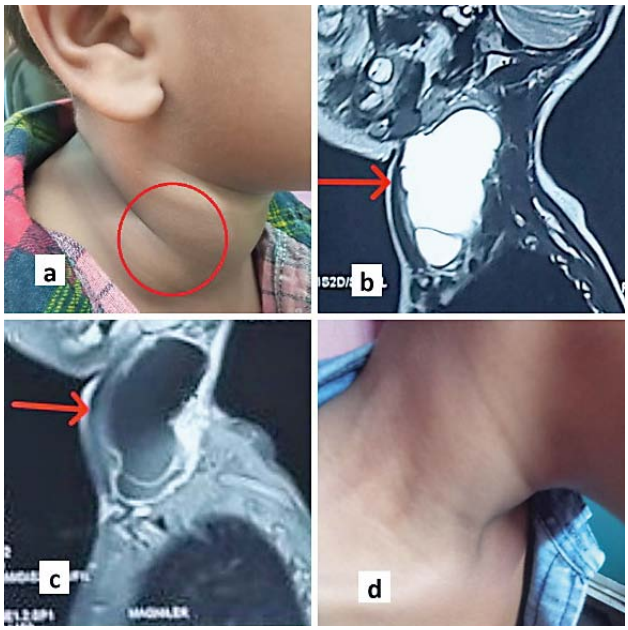

Figure 1. Cystic lymphangioma (encircled in red) measuring about 6X6cm in the right side of the neck at presentation **(a)**; Magnetic resonance imaging of the neck demonstrating a well-defined loculated cystic lesion with septations within the extent of lymphangioma on T2 weighted sagittal section **(b)**; hypointense loculated cystic lesion in post contrast section **(c)** and regression of the lymphangioma following sclerotherapy **(d)**.

CASE REPORT

A five-year-old boy, second child of a non-consanguineous marriage, presented with an 8-day history of painless swelling on the right side of the neck. There was no fever, respiratory distress, or difficulty in swallowing. The swelling gradually increased in size. His perinatal history was uneventful, immunizations were up to date, and development was age-appropriate. No significant past or family history was noted.

On examination, the child was afebrile and haemodynamically stable, weighing 15.5 kg and measuring 98 cm in height. A 6x6 cm, well-defined, mobile, non-tender swelling was noted on the right side of the neck, extending from below the ear to the clavicle (Figure 1a). It was smooth, non-pulsatile, and not fixed to the skin, with normal overlying skin. No other abnormalities were found.

Ultrasound revealed a multilocular cystic mass suggestive of lymphangioma. MRI (Figure 1b and

1c) showed a well-defined loculated cystic lesion with septations within the extent of lymphangioma on T2 weighted sagittal section and hypointense loculated cystic lesion in post contrast section. MRI brain and abdominal ultrasound were normal.

Given the macrocystic nature of the lesion, its well-defined margins, and the absence of acute complications such as airway compromise or infection, sclerotherapy was selected over primary surgical excision. This approach aligns with current evidence recognizing sclerotherapy as an effective first-line treatment for macrocystic lymphangiomas, associated with lower complication rates and superior cosmetic outcomes. Following a comprehensive discussion with the child's parents regarding available treatment options, potential risks and benefits, the likelihood of requiring multiple sessions, and the possibility of recurrence, informed consent was obtained for intra-lesional sclerotherapy. The child was treated with a single dose of intra-lesional bleomycin (0.5 mg/kg), resulting in regression of the swelling (Figure 1d). No adverse events were noted following the procedure. Three-month follow-up ultrasonography revealed near-complete resolution of the cystic lesion, with only minimal residual septations and no detectable vascularity, and given the significant clinical and radiological regression, repeat MRI was not considered. The child has remained asymptomatic with no evidence of recurrence during two years of follow-up.

DISCUSSION

Cystic lymphangiomas are benign congenital malformations of the lymphatic system, with approximately 80–90% of cases diagnosed before the age of two years (1). The cervical region is the most frequently involved site, accounting for nearly three-quarters of reported cases (2). Classical lesions are superficial, soft, compressible, and transilluminant, facilitating early diagnosis in infancy.

The diagnostic challenge in this case arose from the unusual age of presentation, as lymphangiomas typically present before two years of age. Additionally, the relatively rapid increase in size over a short duration raised concern for alternative diagnoses. Differential diagnoses considered included reactive or tubercular lymphadenitis or thyroglossal duct cyst. The absence of systemic symptoms, normal overlying skin, non-pulsatile nature of the swelling, and lack of solid components on imaging made infectious, vascular, and malignant causes less likely. MRI played a crucial role in delineating the extent of the lesion, confirming its cystic nature, and excluding intracranial or mediastinal extension, thereby aiding definitive diagnosis.

The present case is notable for its delayed presentation at five years of age, which is uncommon but increasingly recognized in the literature. Delayed presentation is often attributed to deep-seated lesions, which may remain clinically silent until sudden enlargement due to infection, hemorrhage, or lymphatic obstruction (5, 6). *Chakravarty et al.* reported a giant cervicofacial lymphangioma in a 14-year-old adolescent, emphasizing that lymphangiomas should remain a diagnostic consideration even beyond early childhood (15). Similarly, *Opoko et al.* described cervical cystic lymphangioma in a child presenting later than infancy, reinforcing the heterogeneity in age at diagnosis (16).

Another distinguishing feature of our case was the deep posterior cervical location with proximity to major neurovascular structures, yet without airway compromise, dysphagia, or neurological deficits. Deep cervical lymphangiomas pose diagnostic challenges due to reduced compressibility and atypical consistency, often mimicking other cystic neck masses such as branchial cleft cysts or salivary lesions (8, 15). In the case reported by *Nurzawani et al.* (9), a 3-year-old child presented with a progressively enlarging posterior cervical cystic mass, which caused restricted neck movements and intermittent discomfort, necessitating surgical excision. In contrast, our patient remained asymptomatic despite lesion size and depth, highlighting the variable clinical behavior of cervical lymphangiomas.

Imaging plays a pivotal role in diagnosis and treatment planning. While ultrasonography serves as a useful initial modality, MRI is the imaging modality of choice, as it accurately delineates lesion extent, internal septations, and relationships with adjacent neurovascular structures (4 ,7). MRI

findings in our case were instrumental in confirming the diagnosis and excluding mediastinal or intracranial extension (8,15).

Management strategies for lymphangiomas have evolved over time. Surgical excision has traditionally been considered definitive treatment; however, it is associated with risks such as neurovascular injury, incomplete resection, and recurrence rates of up to 20–27%, particularly in multiloculated or infiltrative lesions (7, 8). Both *Chakravarty et al.* (15) and *Nurzawani et al.* (9) opted for surgical excision, with favorable short-term outcomes but inherent operative risks due to deep anatomical involvement.

In contrast, sclerotherapy has emerged as a safe and effective minimally invasive alternative, especially for macrocystic lesions. Agents such as bleomycin, doxycycline, and OK-432 have demonstrated comparable success rates with reduced morbidity, better cosmetic outcomes, and shorter hospital stays (12). The complete regression following a single intralesional dose of bleomycin (0.5 mg/kg) in our patient underscores the efficacy of sclerotherapy, even for sizeable and deep cervical lesions.

The role of sclerotherapy is particularly relevant in low- and middle-income countries (LMIC), where access to specialized pediatric surgical care and postoperative intensive monitoring may be limited. Ultrasound-guided sclerotherapy offers a cost-effective, daycare-based treatment option with minimal infrastructure requirements, making it an attractive first-line modality in resource-constrained settings (3,12).

Although recurrence remains a recognized concern – particularly in multilocular or microcystic lymphangiomas – long-term follow-up is essential. The sustained remission observed over two years in the present case reinforces evidence that early diagnosis and timely sclerotherapy can prevent complications such as airway obstruction, infection, neurovascular compression, and cosmetic deformity.

CONCLUSION

This case highlights the need to maintain a broad differential diagnosis for paediatric neck swellings, especially in atypical presentations. Early imaging and prompt initiation of sclerotherapy resulted in complete regression and prevention of potential complications. The excellent response to a single intra-lesional bleomycin injection underscores its effectiveness as a minimally invasive treatment, while continued follow-up remains essential due to the risk of recurrence.

Abbreviations:

LMIC – Low and Middle Income Countries

MRI - Magnetic resonance imaging

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SAŽETAK

Rijedak slučaj cističnog limfangioma vrata kod predškolskog djeteta: pravovremena dijagnoza i liječenje

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Uvod: *Cistični limfangiomi su rijetke kongenitalne limfne malformacije koje se obično javljaju prije druge godine života, najčešće zahvaćajući cervikalnu regiju. Prikaz ovog slučaja važan je zbog atipične dobi pojave, duboke zahvaćenosti vrata i uspješnog nekirurškog liječenja.*

Prikaz slučaja: *Izvještavamo o petogodišnjem dječaku koji se javio s osmodnevnom anamnezom progresivno rastuće, bezbolne otekline vrata s desne strane. Kliničkim pregledom otkrivena je dobro definirana, pokretna masa na vratu. Ultrazvuk i magnetska rezonancija pokazali su multilokularnu cističnu leziju u smislu cističnog limfangioma. Dijete je liječeno jednom intralezijskom dozom bleomicina, što je rezultiralo značajnom regresijom otekline. Ostao je asimptomatski bez znakova recidiva tijekom dvije godine praćenja.*

Zaključci: *Ovaj slučaj naglašava potrebu razmatranja široke diferencijalne dijagnoze oteklina vrata u djece, posebno kod atipičnih prezentacija. Rana slikovna dijagnostika i brzi početak skleroterapije rezultirali su potpunom regresijom i sprječavanjem potencijalnih komplikacija. Izvrstan odgovor na jednu intralezijsku injekciju bleomicina naglašava njegovu učinkovitost kao minimalno invazivnog liječenja, dok je kontinuirano praćenje i dalje ključno zbog rizika od recidiva.*

Ključne riječi: LIMFANGIOM, CISTIČNI; SKLEROTERAPIJA; DIJETE, PREDŠKOLSKA DOB; LIMFNE ŽILE