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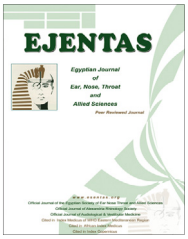

CASE REPORT

A rare case of spontaneous regression of huge neck lymphangioma post primary infection

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KEYWORDS

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Abstract Spontaneous regression of lymphangioma is a rare entity. This case report describes a case of huge lymphangioma over the left submandibular region for a twelve-year-old boy, which had clinically regressed completely two months after recovery from an episode of infection with no clinical recurrence two years upon follow-up. Antibiotic treatment and needle aspiration for pressure relief was done during acute infection. In view of possibility of spontaneous regression, expectant management can be considered for cases of asymptomatic lymphangioma.

1. Introduction

Lymphangiomas are rare benign lesions which result from localized congenital lymphatic malformations. They attribute to about 6% of benign tumours in paediatric age group.[1] About 50% of lymphangiomas are present at birth while 90% of lymphangiomas are diagnosed in children younger than 2 years old.[2] From our literature review, spontaneous regression of lymphangioma is very rare. We would like to share our experience in managing a case of spontaneous infection of huge neck lymphangioma which later achieved complete clinical regression with no recurrence clinically at 2 years of follow-up.

2. Case report

A 12-year-old boy had been under paediatric surgical team follow-up since the age of three months for the complaint of left submandibular swelling, which had been present since birth. The diagnosis of lymphangioma was confirmed by ultrasonography which showed septated multicystic lesion over the left submandibular region about 5.3 × 1.8 cm.

The swelling gradually increased in size and was fluctuant. The largest size measured about 10 cm × 10 cm at the age of five. Magnetic resonance imaging done showed multiloculated cystic lesion with thin septations extending from the left submandibular region to the left parotid region.

There were no signs of infection during follow-ups. As per parental choice, in view of possible inflammatory reaction from OK-432 which might cause airway obstruction, a conservative management was opted for him.

He remained asymptomatic till 12 years of age upon which he presented to Department of Otorhinolaryngology, Head and Neck Surgery, Sibu Hospital for infected left submandibular lymphangioma. The left neck swelling acutely increased in

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size over a duration of four days which was associated with pain, fever and reduced oral intake due to limited mouth opening. There was no associated recent upper respiratory tract infection or trauma to the neck. On examination, there was a huge left submandibular swelling about 15 × 10 cm, erythematous, warm, tense and tender. There was no obvious source of infection either from intraoral or from the external neck. Immediate computed tomography (CT) of neck (Fig. 1) revealed a cystic mass of about 6.1 × 8.9 × 7.6 cm which is centred within the left submandibular space. Multiple thin septae were noted within the cystic mass. The mass extended medially into the left parapharyngeal space, laterally till the left parotid space, anteriorly to the floor of mouth on the left side not crossing midline, posteriorly onto the left sternocleidomastoid muscle and displaced it posteriorly. There was a small component which extended into the left carotid space compressing onto the left internal jugular vein. There was no evidence to suggest hematoma within. Streakiness around the lesion suggested infected lesion.

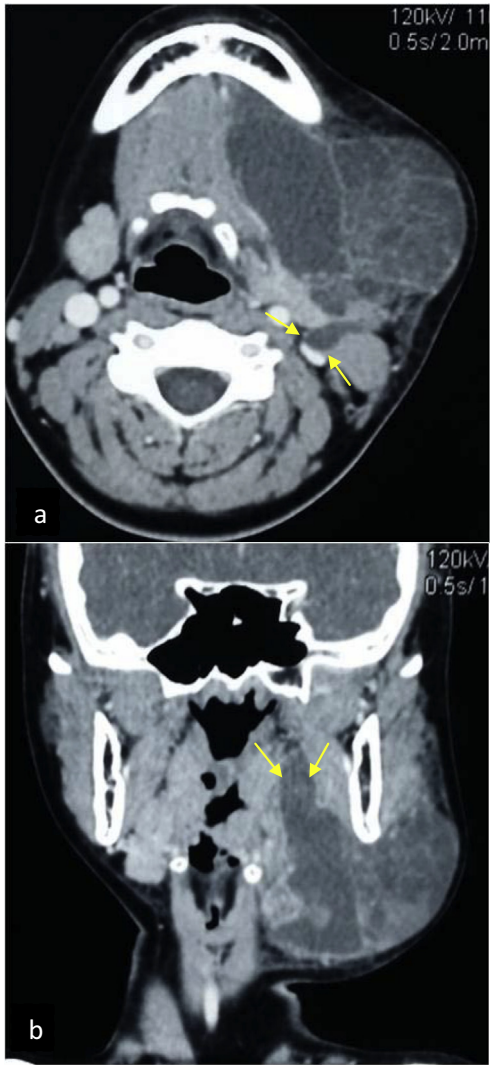

Figure 1 CT neck showed a large multiseptated cystic lesion centred within the left submandibular space, extended medially into left parapharyngeal space, laterally till left parotid space, anteriorly to the left sided floor of mouth, posteriorly onto the left sternocleidomastoid muscle; (a) axial view: extension into the left carotid space, (b) coronal view: extension to left parapharyngeal space.

Needle aspiration with 23G needle was done and about 20 cc hemoserous fluid was aspirated. Trismus spontaneously resolved upon aspiration. Intravenous Augmentin was commenced and continued for a week.

Upon completion of antibiotics, infection subsided and the swelling slightly reduced in size. He was then planned for OK-432 injection two months later. However, the swelling further regressed and upon follow-up in two months it was not palpable clinically (Fig. 2). Ultrasonography showed multiloculated cyst with the largest cyst measuring just less than 1 cm. With this the decision for intralesional OK-432 injection was withheld.

Repeated ultrasonography of the neck one year later showed ill-defined multiseptated cystic lesion over the left submandibular region measuring 1.1 × 2.1 × 0.7 cm, with no significant change in size compared to the previous scan. At 2 year follow-up after the infection, there was no palpable neck swelling.

3. Discussion

Lymphangiomas probably occurs as a result of failed connection between lymphatic system and jugular vein on the fourteenth day of pregnancy, causing stasis of lymph and formation of cysts which are covered by an endothelial layer.[3] Lymphangiomas can occur in any part of the body with head and neck region being the most common site. It can be found less commonly in axilla, mediastinum, internal organs, trunks and extremities.[1]

Lymphangiomas usually present in children as painless mass which raises parental concern. However, they may present along with complications of respiratory distress, feeding difficulty and signs of infection. Lymphangiomas can get infected either primarily or secondarily where usually it is associated with an upper respiratory tract infection. In our patient, the left submandibular region swelling was present at birth and the child remained asymptomatic up to the age of 12, until a primary infection took place. During this period, the swelling increased in size, was erythematous, warm, tender and causing limited mouth opening. Aspiration of the swelling was performed for symptomatic relief and treated with intravenous antibiotics for one week duration. Aspiration is not a definitive treatment but may be useful for emergency decompression.[1]

Lymphangiomas can usually be diagnosed from history and physical examination. Nevertheless, radiological imaging plays an important role to confirm the diagnosis and to exclude other possible differentials of congenital lesions namely branchial cyst, thyroglossal cyst, teratoma and lipoma. Ultrasonography is also useful for prenatal diagnosis of lymphangiomas from the fourth month of gestational period.[3] Computed tomography and magnetic resonance imaging also play a significant role in surgical management of lymphangiomas. They help to delineate extension and anatomy study before operation to avoid iatrogenic injury to vital structures.[4] In our case, the diagnosis of lymphangiomas was confirmed with ultrasonography and magnetic resonance imaging which showed