INTERNATIONAL JOURNAL OF MEDICAL AND APPLIED SCIENCES
Earthjournals Publisher
eISSN:2320-3137
www.earthjournals.in

CASE REPORT

LYMPHANGIOMA OF ANKLE : A CASE REPORT

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Publication history: Received on 25/8/2018, Published online 16/9/2018

ABSTRACT:

Lymphangiomas are rare vascular lymphatic malformation .The rate of occurrence in foot and ankle region have been scarcely reported.MRI is the best modality of investigation. Treatment option includes complete surgical resection, sclerotherapy, and the use of angiogenesis inhibitors. We report a case of lymphangioma at the ankle.

Keywords:Lymphangioma,lymphatic malformation, ankle, vascular malformation.

INTRODUCTION

Lymphangiomas are uncommon, hamartomatous, congenital malformations of the lymphatic system that involve the skin and subcutaneous tissues.
Lymphangiomas of the foot and ankle is a rare occurrence. Lymphangiomas are equally distributed among genders and races. In the literature, their incidence in children is estimated to amount to 6% of all benign tumors.. They are commonly misdiagnosed as hemangiomas. Hemangioma is the most common and is almost exclusive to infants. Vascular malformations are the result of abnormal development of vascular elements during embryogenesis and fetal life.. Vascular malformations do not generally demonstrate increased endothelial turnover. They are designated according to the predominant channel type as capillary malformations, lymphatic malformations (LMs), venous malformations (VMs), arteriovenous malformations, and complex forms such as capillary-lymphatico-venous malformations. Malformations with an arterial component are fast-flow while the remainders are slow-flow.

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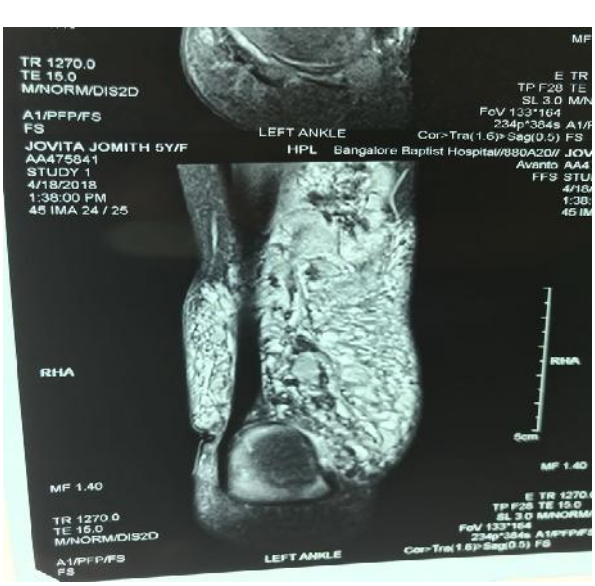
Figure 1 MRI of the swelling;



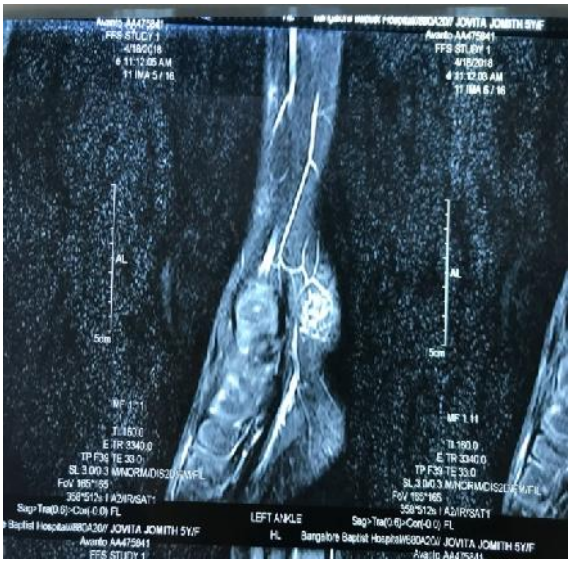
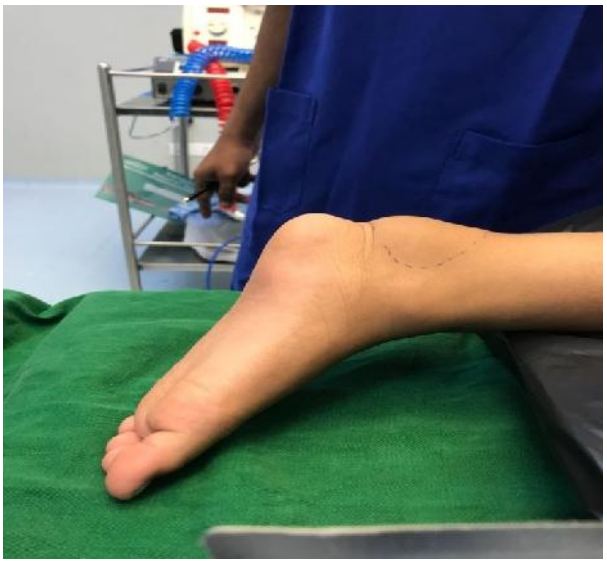
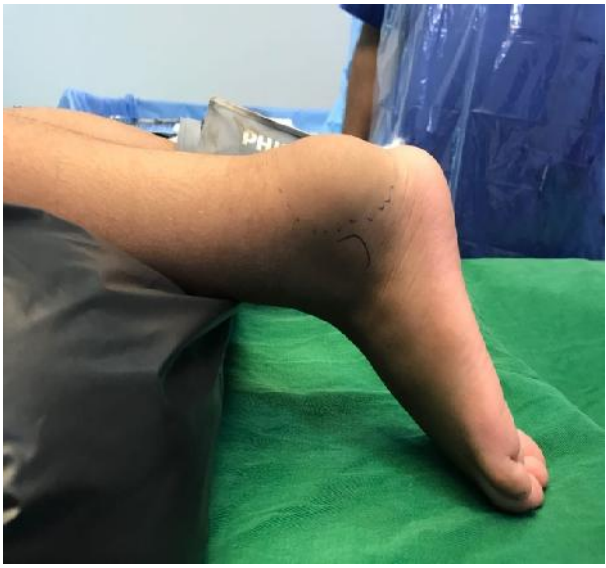


Figure 2 Pre op pics of lymphangioma;





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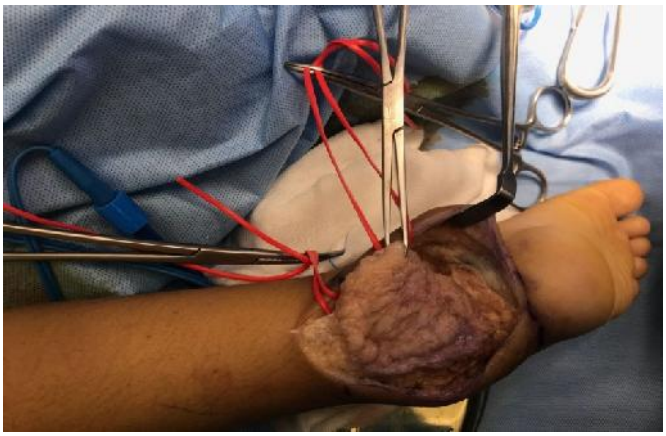
CASE REPORT

A 4 yr old girl presented with history of swelling over her left ankle since birth.
On examination the swelling of about 5*3cm was noted over the posterior aspect of the left ankle joint. The swelling was non tender ,firm to soft ,pear shape and fixed to the skin with smooth surface. The swelling was gradually increasing in size .
MRI showed lobulated multicystic mass.
Diagnosis of lymphangioma was made and planned for excision biopsy of the swelling.

On table during dissection it is noted that the swelling was encasing the sural nerve on the lateral aspect of the ankle. Strawcoloured lymph filled multiple sacs were noted. There was hardly any delineation of subcutaneous tissue from dermis to deep fascia. Every structure was replaced by a dense, dilated network of lymphatic channels and fibrous tissue.Nerve sparing technique was used to preserve the sural nerve .The swelling was dissected out of the tendo achilis as the swelling was just overlying the tendon.Grossly almost all the abnormal swelling was excised .Wound was closed over a suction drain.



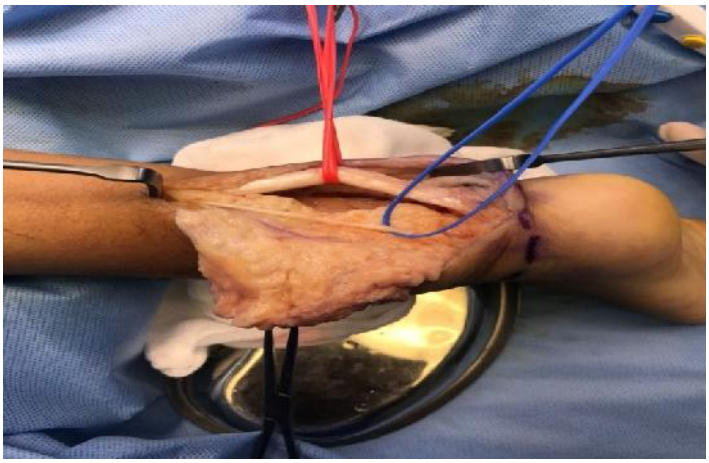


Figure 3 Intra op pics.

DISCUSSION
Lymphangiomas are developmental malformations of the lymphatic system . Prenatal ultrasonography can detect relatively large lesions as early as the second trimester, although lymphangiomas are frequently misdiagnosed as other pathologic entities. Lymphangiomas most commonly occur in the cervicofacial region, axilla/chest, mediastinum, retroperitoneum, buttock and perineum.
Lymphangiomas in the subcutis or submucosa manifest as tiny vesicles. Intravascular bleeding is evident by tiny, dark red domed shaped nodules.
Radiologic documentation is best performed by MRI .
Histologically, lymphangiomas consist of ecstatic lymphatic channels,occasionally containing a mild lymphocytic infiltrate.

Classification by histologic appearance includes three subtyes: capillary, cavernous and cystic. The capillary form is composed of small thinned walled lymphatic vessels. Cavernous consists of large lymphatic channels with adventitial coats.

The third and most common type, cystic lymphangioma is composed of macroscopic lymphatic spaces. These lesions are often multiseptated, which is suggestive of infiltration across the tissue planes.

Lymphangioma rarely affects the foot and ankle region.

The two main complications of that can arise from lymphangiomas are intralesional bleeding and infection. Lymphangiomas often swell in the event of a viral or bacterial infection. Most often this is a harmless event likely related to change in flow or alterations. Bacterial cellulitis, however, is more dangerous and requires prolonged intravenous antibiotics.

The two main strategies used to treat lymphatic anomalies are schlerotherapy and surgical resection. Sclerotherapy works through obliteration of the lymphatic lumen.

Macrocystic lymphangiomas is more likely than microcystic tissue to shrink after an injection of sclerosant.

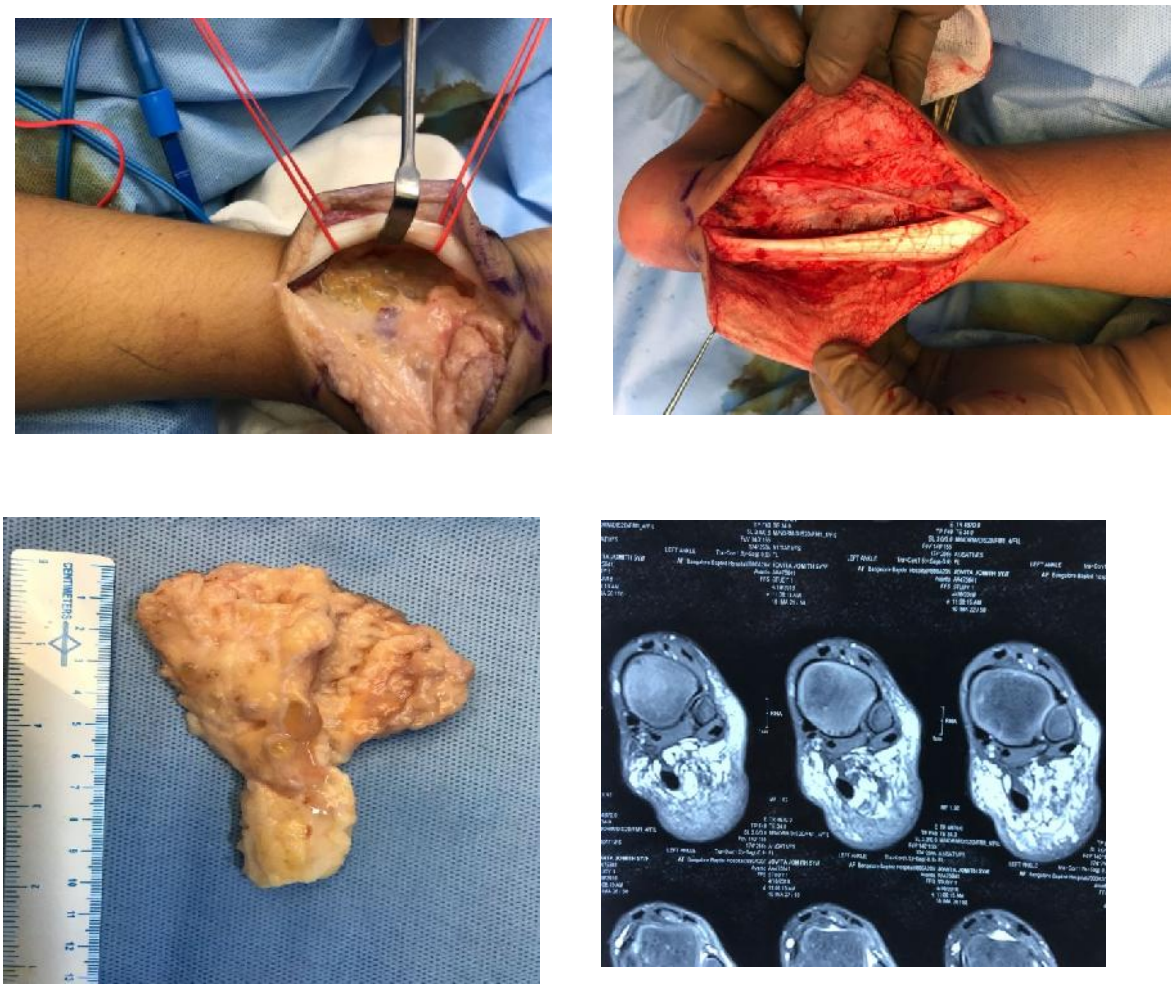


Figure 4 Intra operative removal of the lymphangioma . Removed lymphangioma on the back table.

On MRI, lymphangiomas are characterized by the absence of feeding vessels and lack of intense contrast enhancement. Because of the infiltrating nature of the lesion, complete excision rates are only 18-50%. It appears that there have not been a reported percentage of lymphangioma occurrences of the foot and ankle region.

In 2009, Itakura, et al., reported 114 cases of lymphangiomas, which of only 2 occurred in the foot. According to their findings the incidence of lymphangioma of the foot is 1.75%.

Although lymphangiomas are mainly a soft tissue pathology they can occur within bone, but remain extremely rare in the lower extremities.

Wu in 1996 described a case of a 12 year-old female with a lymphangioma of the anterior aspect of her ankle. He promotes the use of Lymphangiography for diagnosis and recommends for complete surgical excision for treatment. They usually appear in the cervicofacial area up to 75% of the time.

In 2004, Ly, et al., described another case of lymphangioma in a 2 year-old girl's foot.

A compressive dressing should be applied immediately post operatively to prevent complications. Even with an intensive approach to resection, the recurrence rate is reported to be 40% after an incomplete excision and 17% after a macroscopically complete excision.

In conclusion, lymphangiomas are rare in the foot and ankle region. A thorough history and physical examination are necessary to help distinguish from other benign soft tissue tumors. Imaging studies such as radiographs and MRI are highly recommended to aid in the diagnosis of these tumors.

Surgical resection of the mass is recommended for the treatment with the goal of complete excision. The specimen was sent to pathology for confirmation of the diagnosis Surgical resection was recommended for this patient. Overall, the patient had no complications with no sequela.

Infection is a common complication for this procedure and the patient should receive pre operative antibiotics and possibly continue them for 5 days post resection to avoid this complication. Suture removal was done on post op day 8 and compression dressing was applied.The patient was followed post resection of the tumor and was doing satisfactory, no pain was present upon stance or ambulation .On pathological examination sections showed fragments of fibroadipose tissue showing many dilated vascular channels focally filled with amorphous material consistent with lymphangioma.

Written consent was obtained from the patient/s or their relative/s for publication of this study.

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Paper cited as: **Kumar Nikhil, Ramani C V,Anoop T. LYMPHANGIOMA OF ANKLE : A CASE REPORT . International Journal of Medical and Applied Sciences. 2018;7(2): 12-17.**