J Orthop Spine Trauma. 2019 December; 5(4): 107-9. DOI: http://dx.doi.org/10.18502/jost.v5i4.4381

Case Report

Huge Post-Traumatic Cystic Hygroma, Combination of Surgical and Non-Surgical Treatment Methods: A Case Report

Ehsan Hedayat[1], Mohammad Hossein Nabian[2,*]

[1] Resident, Department of Orthopedic Surgery, Shariati Hospital, School of Medicine, Tehran University of Medical Sciences, Tehran, Iran
[2] Assistant Professor, Department of Orthopedic Surgery, Center of Orthopedic Trans-Disciplinary Applied Research, Shariati Hospital, School of Medicine, Tehran University of Medical Sciences, Tehran, Iran

[*]Corresponding author: Department of Orthopedic Surgery, Center of Orthopedic Trans-Disciplinary Applied Research, Shariati Hospital, School of Medicine, Tehran University of Medical Sciences, Tehran, Iran. Tel: +98-9126305095, Email: dr.nabian@gmail.com

Received: 10 September 2019; **Revised:** 14 October 2019; **Accepted:** 19 November 2019

Abstract

Background: Cystic hygroma (cystic lymphangioma) is a malformation of the lymphatic system. It is rare in adults and trauma may be the cause in some cases. Surgical and non-surgical treatment methods have been described in the management of cystic hygroma.
Case Report: A 38-year-old woman, known case of systemic lupus erythematosus (SLE), presented a huge cystic formation in the posterior aspect of her thigh following trauma. We treated the patient using a combination of three treatment methods including surgical excision, intra-cavity bleomycin injection, and post-operative use of compression pants.
Conclusion: Huge post-traumatic cystic hygroma in adults is rare. By the combination of the above three therapeutic regimens, there was no cystic lesion recurrence after eight months.

Keywords: Lymphangioma; Cystic; Surgery

Citation: Hedayat E, Nabian MH. **Huge Post-Traumatic Cystic Hygroma, Combination of Surgical and Non-Surgical Treatment Methods: A Case Report.** *J Orthop Spine Trauma* 2019; 5(4): 107-9.

Background

Cystic hygroma is a malformation of the lymphatic system that usually is found in the first 3 years of life (1).

It is suggested that lymphangiomas are the result of a failure of connection between the lymphatic system and the venous system (2).

It is rare to find cystic hygroma in adults which could be due to its congenital source and early childhood appearance (3).

Some case reports have described post-traumatic cystic hygroma formation in adults (4).

Surgical and non-surgical treatment options for cystic hygroma have been introduced over the past decades (5-8).

Here, we report an adult patient with a huge cystic lymphangioma after surgery in her thigh. We consider the surgery as a major trauma.

Case Report

A 38-year-old woman, known case of systemic lupus erythematosus (SLE), referred to the emergency department of Shariati Hospital. Her SLE was in the remission phase and she was taking oral corticosteroid 10 mg daily. She complained of pain and swelling in the posterior aspect of her left thigh. In clinical examination, she had an oral temperature of 40 °C. The posterior aspect of her thigh and knee was warm and erythematous. There was crepitation in touch. In the initial radiography of the thigh, gas lucencies were seen in the posterior thigh muscles (Figure 1). Sonographic evaluation reported gas echo and fluid accumulation in the posterior compartment. Serum erythrocyte sedimentation rate (ESR) and C-reactive protein (CRP) levels were elevated (Table 1). She underwent drainage and staged debridement of the posterior compartment of her thigh with suspicion to necrotizing fasciitis (NF). Her tissue culture came back positive [Escherichia coli (E. coli)]. Long-term intravenous antibiotic therapy was started for her. Her infection was completely treated and resolved.

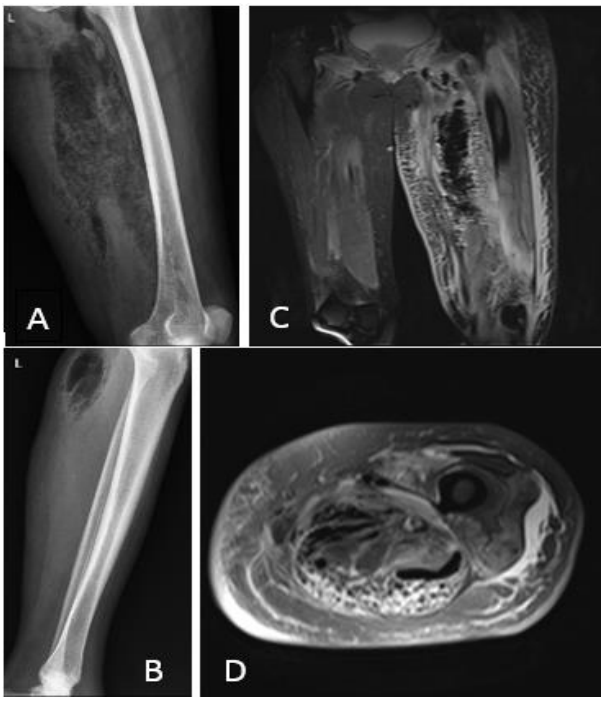

Figure 1. A) Lateral radiograph of the left femur, gas lucencies are obvious in the posterior compartment of the thigh; B) Lateral radiograph of the left tibia; C) T2 coronal image and D) T2 axial section of the left thigh magnetic resonance imaging (MRI)

Table 1. Initial laboratory data

Variable	Patient value	Normal range
WBC (/µl)	7870	4500-12500
Hb (g/dl)	7	12-16
PLT (/µl)	79000	150000-400000
BUN (mg/dl)	43	7-20
Cr (mg/dl)	1.81	0.60-1.30
ESR (mm/h)	50	< 20
CRP (mg/dl)	183	< 10

WBC: White blood cell; Hb: Hemoglobin; PLT: Platelet; BUN: Blood urea nitrogen; Cr: Creatinine; ESR: Erythrocyte sedimentation rate; CRP: C-reactive protein

15 months later in the follow-up, she complained of swelling and mild pain in the posterior aspect of her thigh and knee.

In physical examination, a 12 cm × 40 cm cyst-like mass was found on the posterior aspect of her left thigh. There was no tenderness on palpation, erythema, or warmth (Figure 2A). Anteroposterior (AP) and lateral view radiographs were unremarkable (Figures 2B, 2C). Needle aspiration of the cyst revealed a light-brown fluid. Magnetic resonance imaging (MRI) of the thigh showed a huge cystic mass on the posterior aspect of the posterior muscular compartment of the thigh (Figures 2D, 2E, and 2F).

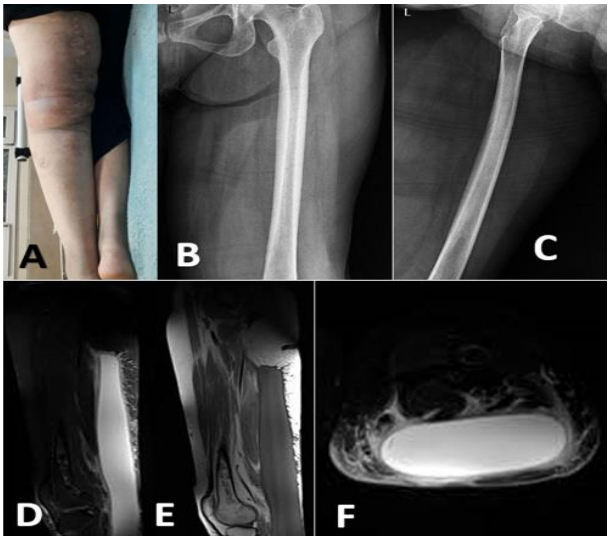

Figure 2. Preoperative photography (A), radiography imaging anteroposterior and lateral X-ray (B, C), and magnetic resonance imaging (MRI) of the patient (D, E, F)

Fluid analysis showed no evidence of hematoma or abscess collection and the fluid culture was negative. Pathologist detected inflammatory cells in the fluid.

The patient underwent surgical excision of the cystic mass by the primitive diagnosis of post-traumatic cystic hygroma. Unfortunately, we do not have tissue sample for pathology analysis to confirm the diagnosis and exclude malignancies.

The cyst was approached by direct posterior incision and was well encapsulated and filled with a light-brown fluid (Figure 3A).

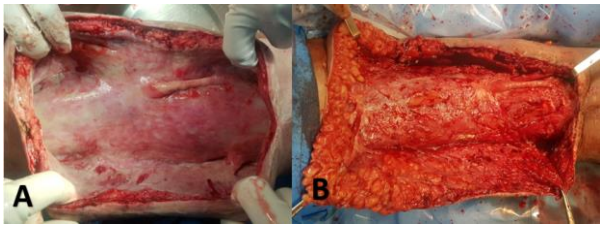

Figure 3. Intraoperative photography before (A) and after (B) excision of the encapsulated cyst

The capsule was completely excised (Figure 3B) and wound was closed in layers followed by a compressive pant for 6 weeks. Based on previous studies a 1 mg/kg dose of bleomycin was injected in the dead space with 3 weeks interval for two times (9-12).

After 7 months of follow-up, she had no pain and there were no tenderness, swelling, or erythema in physical examination. Her left knee and hip range of motion (ROM) was complete without any pain. Follow-up MRI showed no evidence of collection, edema, or inflammation in her thigh and knee (Figure 4).

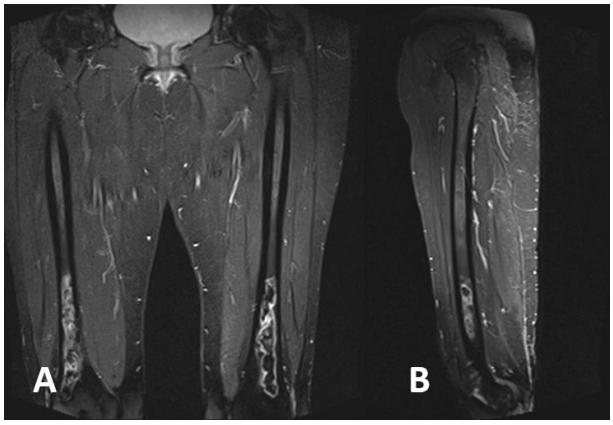

Figure 4. Seven month follow-up magnetic resonance imaging (MRI); (A) Coronal view; (B) Sagittal view

Discussion

Cystic lymphangioma is a rare lymphatic anomaly that usually occurs in children (1, 13). It may rarely be seen in adults (3). The etiology of lymphatic disorder is not fully understood.

Delay in the proliferation of lymphoid nests maybe the etiology of lymphangioma in adults. This delayed proliferation may be due to a stimulus such as trauma or infection (14, 15). Our patient had an irrigation and debridement surgery for her NF 23 months ago.

Paraclinical evaluations for the diagnosis of cystic hygroma include ultrasound scan, MRI, computed tomography (CT), and fine-needle aspiration (FNA) (especially to exclude malignancy) although postoperative histology is necessary for definitive diagnosis (16).

The treatment of choice is complete surgical excision of the cyst (especially in well-encapsulated cysts), but recurrence is common and may require multiple operations (5-7).

Another option is the injection of sclerosing agents such as bleomycin in the cyst, especially to prevent the complications and morbidities of surgical excision (8). Bleomycin is an antitumor agent developed in 1966. It inhibits deoxyribonucleic acid (DNA) synthesis (9). Yura et al. were the first clinicians that used bleomycin to treat cystic hygroma (10).

Bleomycin dosage is different in studies ranging from 0.3 to 3 mg/kg (15-17); we used 1 mg/kg. Intra-cystic bleomycin injection therapy has some side effects such as transient increase in the size of swelling, intra-cystic hemorrhage, infection, fever, pulmonary fibrosis, and leukocytosis.

To the best of our knowledge, huge post-traumatic cystic hygroma has not been reported in adults so far. Our case had a huge encapsulated single cyst. We excised the

entire cyst. To treat residual microscopic lesions, we used a 2-time injection of bleomycin. Also, in post-operative care, compression pant was used to decrease dead space and prevent re-accumulation of fluid.

By the combination of the above three therapeutic regimens, there was no recurrence after eight months.

Cystic hygroma may be caused by physical trauma such as surgery and it is not always congenital. Lymphangioma is one of the differential diagnoses of cystic lesions after trauma.

There are surgical and non-surgical options to treat cystic hygroma; combination of these two methods maybe helpful in huge cystic lesions.

Conflict of Interest

The authors declare no conflict of interest in this study.

Acknowledgments

None.

References

1. James DR. Craniofacial anomalies. In: Kerr AG, Editor. Scott Brown's otolaryngology. London, UK: Butterworth-Heinemann; 1987. p. 267.
2. Guarisco JL. Congenital head and neck masses in infants and children. Part II. *Ear Nose Throat J*. 1991;70(2):75-82. [PubMed: 2044478].
3. Baer S, Davis J. Cystic hygroma presenting in adulthood. *J Laryngol Otol*. 1989;103(10):976-7. doi: 10.1017/s0022215100110679. [PubMed: 2685151].
4. Kim HS, Kim EA, Chung SM, Shin YR. Cystic lymphangioma: Trauma may be a significant cause? *Int J Pediatr Otorhinolaryngol*. 2007;71(12):1921-3. doi: 10.1016/j.ijporl.2007.08.018. PubMed: 17913248].
5. Aneeshkumar MK, Kale S, Kabbani M, David VC. Cystic lymphangioma in adults: Can trauma be the trigger? *Eur Arch Otorhinolaryngol*. 2005;262(4):335-7. doi: 10.1007/s00405-004-0780-6. [PubMed: 15841412].
6. Bill AH Jr., Sumner DS. A unified concept of lymphangioma and cystic hygroma. *Surg Gynecol Obstet*. 1965;120:79-86. [PubMed: 14259790].
7. Tran NN, Tran XN. Cystic hygroma in children: A report of 126 cases. *J Pediatr Surg*. 1974;9(2):191-5. doi: 10.1016/s0022-3468(74)80120-x. [PubMed: 4825790].
8. Waner M, Suen JY. Treatment options for the management of vascular malformations. In: Waner M, Suen JY, Editors. Hemangiomas and vascular malformations of the head and neck. Hoboken, NJ: Wiley; 1999. p. 315-50.
9. Umezawa H. Recent study on biochemistry and action of Bleomycin. In: Carter SK, Crook ST, Umezawa H, Editors. Bleomycin: Current status and new developments. New York, NY: Academic Press; 1978. p. 15-20.
10. Yura J, Hashimoto T, Tsuruga N, Shibata K. Bleomycin treatment for cystic hygroma in children. *Nihon Geka Hokan*. 1977;46(5):607-14. [PubMed: 73364].
11. Baskin D, Tander B, Bankaoglu M. Local bleomycin injection in the treatment of lymphangioma. *Eur J Pediatr Surg*. 2005;15(6):383-6. doi: 10.1055/s-2005-872922. [PubMed: 16418953].
12. Niramis R, Watanatittan S, Rattanasuwan T. Treatment of cystic hygroma by intralesional bleomycin injection: Experience in 70 patients. *Eur J Pediatr Surg*. 2010;20(3):178-82. doi: 10.1055/s-0030-1247548. [PubMed: 20178075].
13. Hancock BJ, St-Vil D, Luks FI, Di LM, Blanchard H. Complications of lymphangiomas in children. *J Pediatr Surg*. 1992;27(2):220-4. doi: 10.1016/0022-3468(92)90316-y. [PubMed: 1564622].
14. Suk S, Sheridan M, Saenger JS. Adult lymphangioma: A case report. *Ear Nose Throat J*. 1997;76(12):881-3. [PubMed: 9431778].
15. Wiggs WJ Jr, Sismanis A. Cystic hygroma in the adult: Two case reports. *Otolaryngol Head Neck Surg*. 1994;110(2):239-41. doi: 10.1177/019459989411000216. [PubMed: 8108161].
16. Gow L, Gulati R, Khan A, Mihaimeed F. Adult-onset cystic hygroma: A case report and review of management. *Grand Rounds*. 2010;10:5-11. doi: 10.1102/1470-5206.2011.0002.
17. Kertesz Z, Bala G, Bancu S, Gozar H, Virgil G, Horvath E, et al. Bleomycin therapy for lymphangioma. *Chirurgia (Bucur)*. 2011;106(1):103-7. [PubMed: 21523964].
18. Mahajan JK, Bharathi V, Chowdhary SK, Samujh R, Menon P, Rao KL. Bleomycin as intralesional sclerosant for cystic hygromas. *J Indian Assoc Pediatr Surg*. 2004;9(1):3-7.