SPONTANEOUS RESOLUTION OF PRIMARY AND RECURRENT CYSTIC HYGROMA FOLLOWING ACUTE INFECTION

Kashif Chauhan1, Richard Gan1, Bala Eradi2, Brian Davies1, Shailinder Singh1
1Paediatric Surgical Department, Nottingham University Hospital, UK
2Paediatric Surgical Department, Leicester Royal Infirmary, Nottingham, UK

Abstract

Introduction:
There are reports of chronic infection leading to spontaneous regression of primary cystic hygromas (CH). To the best of our knowledge there is no report of acute infection in primary and recurrent CH leading to a rapid and spontaneous resolution. We report case series of four patients with primary (three) and recurrent (one) CH, which resolved spontaneously following an episode of acute infection within a short span of 1-2 weeks.

Patients and Methods:
A retrospective case note review of four cases of CH was done in two tertiary paediatric surgical centres in UK.

Results:
Four-year-old boy presented with CH in the axilla. It recurred within 3 weeks of surgical excision. He developed acute infection in the recurrent CH while waiting for sclerotherapy. This was treated with oral antibiotics and led to complete resolution of the swelling within a week. Follow up was 3 months.
A neonate presented with congenital left sided CH in the neck. On day 9 of life there was an infection in the CH, which was treated with intravenous antibiotics. There was a complete resolution of the neck swelling within 2 week. Follow up was 12 years.
A 2-month-old girl presented with right-sided CH in her neck. A decision was made to treat it conservatively. The CH got infected at 6 months of age while on conservative management. It was treated with intravenous antibiotics. There was a complete resolution of the neck mass in 2 weeks. Follow up was 2 years.
A neonate presented with congenital right neck CH. Sclerotherapy was planned at 6 months of age. The infant developed an acute infection in CH at 4 weeks, requiring intravenous antibiotics. There was complete resolution of CH within weeks. Follow up was 1 year.

Conclusion:
This case series raises our awareness of spontaneous resolution of primary as well as recurrent cystic hygromas following acute infection.

Keywords: cystic hygroma, acute infection, lymphangioma

Correspondence

Kashif Chauhan
Paediatric Surgical Department
Nottingham University Hospital
Queen Medical Campus
Derby Road, NG7 2UH, United Kingdom
E-mail: mkashifdr@yahoo.com
**Introduction**

Treatment of cystic hygroma is usually sought when lesions pose cosmetic problems, or get infected or traumatised. There are multiple treatment options including aspiration, laser, radiofrequency ablation, sclerotherapy and surgical excision [1] Sclerotherapy or surgical excision is the mainstay of treatment. Neither method has shown superiority over the other [2] Both treatments are associated with nerve damage and infection [2] Although uncommon, there is a chance of spontaneous resolution of cystic hygromas and thus a “watch and wait” approach is sometimes adopted in infants unless complications occur [1] [3]. We report 4 cases of cystic hygroma with spontaneous resolution within 1-2 weeks following an inflammatory response from acute infection. We have searched the literature and there are no case reports of this nature.

**Case Series**

**Case 1**

A 4-year-old boy was referred to the paediatric surgery department complaining of a lump in the left axilla for one year but over the last three months it had increased and then subsequently decreased in size. There was some mild discomfort but there were no complaints of pain, irritation, discharge or change of colour. Movement of arm and neuro-muscular examination was normal. Parents noticed that off and on there was some bluish discoloration of the lump. It was about five centimetres in size, soft, mobile and did not feel to be attached to any deeper structures with no associated lymphadenopathy. An ultrasound was done at the referring hospital (Fig.1A and 1B), which suggested that it was a lipoma, but with suspicions raised that it may be a vascular malformation as it increased as well as decreased in size. Ultrasound scans were discussed in a multidisciplinary meeting and a provisional diagnosis of lymphocele or slow flow lymphovascular malformation was made.

After discussion with parents, a decision was made to remove it as it was causing some discomfort when it swelled up. Surgery has complications and it was not our preferred approach as first choice in treating this child, but the reason for operation was that we were not clear about the diagnosis. During the surgical procedure, it was noticed that the lump was cystic in origin with deeper extensions into the axilla with fluid filled cystic components and was very difficult to dissect. It looked likely to be a cystic hygroma. A careful dissection was done and cystic lesions were removed and sent for histopathology examination. A drain was left in situ for a few days and the child was given antibiotics for five days. Histology of the dissected cystic structure showed microscopic appearances of adipo-fibrous connective tissue and fascicles of skeletal muscle. The connective tissue featured vascular spaces of varying size including large cavernous forms with thin walls, focally featuring lymphoid tissue in littoral regions, and focally containing oedema coagulum. There were large muscular vessels. The features were consistent with cystic lymphangioma/lymphovascular malformation. The child was seen again in our outpatient clinic with re-occurrence of lump 21 days after the surgery. The lump was much bigger than before with the same signs and symptoms (Fig.2). With a confirmed diagnosis of cystic hygroma, on histology with represented same sign and symptoms before the child was re-booked for aspiration of cystic fluid and insertion of OK 432 in the cystic lesion. The child was seen 4 weeks later at the time of operation and was noted to have no lump at all. According to parents the child had some local infection 7 days back and was on oral antibiotics for 5 days (started by General Practitioner) and the swelling/lump resolved completely (Fig. 3). There was no swelling at all with any clinical findings. There was no thickening of tissues felt. The local infection in the lump caused the spontaneous regression of the lump within one week without any surgical intervention. The recurrent cystic hygroma was resolved within a week due to local infection. Child was reviewed in outpatient clinic after 6 months with no re-occurrence and followed up by a telephone call 12 months after the resolution of cystic hygroma with no re-occurrence.

**Case 2**

A 9 days old girl referred to Accident and Emergency Department with swelling left side of the neck. She was born full term and this swelling was noticed at the antenatal scans. The swelling rapidly increases in size in few days after birth. At presentation it was red in colour, erythematous with a purple hue, fluctuant and a diagnosis of infected cystic hygroma was made and intravenous antibiotics were started. The features were consistent with cystic lymphangioma/lymphovascular malformation. The baby was already been referred to surgeons soon after birth for further management of cystic hygroma and an MRI of head and neck was in pipe line. She was discharged home after 10 days. An MRI of the neck was done a month later and no cystic hygroma or any other abnormality was identified. She was managed conservatively and was seen in outpatient clinic two months after the discharge and no swelling was noticed only small area of scarring was noticed. She was further followed up in the outpatient department 10 month post infection of cystic hygroma with no recurrence and there was a small scar mark visible. This is the case report of primary resolution of cystic hygroma within 2-3 weeks with acute episode of infec-
tion without requiring any surgical intervention. She was reviewed in outpatient clinic after 12 months with no re-occurrence.

**Case 3**
A term baby, birth weight of 4 kg born with large right neck swelling. All the antenatal scans were normal. He had an ultrasound, which showed multiseptate lobulated cystic mass with clear fluid. There is no clear solid component or calcification. A diagnosis of cystic hygroma was made and a conservative approach is taken with a view to do sclerotherapy after 6 months (Fig. 4A). He was admitted in the hospital at 4 weeks of age with enlargement and marked tenderness in the cystic hygroma. A diagnosis of infected cystic hygroma is made (Fig. 4B) and he was started on intravenous antibiotics for 5 days leading to complete resolution of cystic hygroma following this acute infection episode. He was seen in outpatient clinic with some redundant skin for which he was referred to plastic surgery. Cystic hygroma completely resolved within a week following an acute infection episode. He was reviewed in out patient clinic after 6 months with no re-occurrence.

**Case 4**
A 2-month-old girl is seen in outpatient clinic with complaint of cystic lesion on right side of the neck since birth. Antenatal scans were all normal. On examination the cystic lesion is about 5 cm into 6 cm. It was soft, non-tender with no deeper attachments. An ultrasound scan was done which showed cystic lesions with multiple septations with a diagnosis of cystic hygroma. She was managed conservatively and reviewed again after 2 months with complaint of intermittent swelling and some discomfort. The lesion was increased in size to...
5 cm into 8 cm and the decision was made to continue conservative management. She was seen in outpatient clinic after 3 months and the cystic swelling was reduced in size (4 cm into 6 cm) and was not causing any discomfort or problem. Again she was managed conservatively without any intervention. Six months later she was admitted in the hospital with redness, swelling and pain the cystic hygroma and was diagnosed with an acute infection. Intravenous antibiotics were given for 10 days. She recovered well and was discharged home. She was seen in the outpatient clinic about two weeks later and there was no cystic swelling noticed. Cystic hygroma resolved completely. She was reviewed in outpatient clinic after 12 months with no re-occurrence. Again an acute infection has caused the cystic hygroma to resolve completely with 2 weeks without any surgical intervention.

Discussion
Lymphangiomas are congenital benign lymphatic malformations [3-5]. They can be broadly classified into capillary, simplex or cystic lymphangiomas. Cavernous lymphangiomas affect dense tissue like the tongue [5]. Cystic lymphangiomas are also known as cystic hygromas and affect loose tissue forming cysts of varying sizes [3, 5]. They arise anywhere in the body, most commonly (75%) in the head and neck but have also affected the trunk, extremities, internal organs and axilla (15%) [6]. Cystic hygromas have an incidence of 1 in 12000 live births and make up 6% of benign tumours in children [3, 4]. Both sexes are affected equally [4]. Several genetic disorders are associated with the development of these lymphangiomas such as Turner’s syndrome and Trisomy 21 [3]. These lesions are thought to arise from sequestration of lymphatic tissue during development that fails to drain into main lymphatic or venous systems [1]. Infections of cystic hygromas are common especially in the suprahyoid microcystic and mixed variety forms [7]. Infection causes inflammation and swelling of the cyst that can aggravate symptoms by putting increased pressure on adjacent structures and obstructing the aerodigestive tract. Initial treatment of the acute episode involves using broad-spectrum antibiotics [7]. Steroids also convey some benefit to acute infection [7]. Repeated infection is usually an indication for surgical excision. The success of surgical excision is most dependent on the site of the lesion being excised, well defined lymphangiomas confined to the posterior triangle having the best results after surgery [5]. Surgical excision has been the mainstay of treatment for many years, completely removing the lymphangioma or to remove as much as possible, sparing all vital neurovascular structures. Complete excision has been estimated to be possible in roughly 40% of cases. Unfortunately, there is a significant complication rate of 12-33% and recurrence of 15-53% [1]. Complications include
Intra-thoracic and intra-abdominal lesions are also preferably managed with a surgical approach. Sclerotherapy involves initial aspiration of the cyst with subsequent injection of a sclerosant and transient compression. There are several sclerosing agents that have been used including doxycycline, alcohol, hypertonic glucose, bleomycin and OK-432. Results have been variable but good outcomes have been demonstrated using bleomycin and OK-432 [1]. OK-432 (Picibanil) is derived from a low virulent strain Streptococcus pyogenes treated with benzylpenicillin potassium [5, 8]. It is thought that an inflammatory reaction to the agent causes fibrosis and subsequent resolution of the cystic hygroma. Bleomycin is considered a poor choice because of its toxicity (pulmonary fibrosis). Absolute alcohol as a sclerosing agent has been used with some success in some patients; alcohol works well a sclerosing agent has been used with some success in some patients; alcohol works well in vascular malformations. Imperizzilli et al. [9] studied CT guided ethanol injection and obtained complete resolution in seven of eight patients without complications. Interferon alfa-2a has been used in the treatment of hemangiomas, and its use has been proposed in lymphangiomas. However, its efficacy has never been documented and it carries a serious side effect profile. Doxycycline has been reported as a potential sclerotherapy agent, with both safety and efficacy. As with most other sclerosing agents, it showed the highest efficacy in microcystic lesions and lowest efficacy in microcystic ones. A limited case series has been reported using sildenafil for severe lymphatic malformations [10]. Cystic hygromas are benign lesions; however, complications may arise. These lesions can get infected any time. The usual source of infection is seeding of microorganisms from a secondary focus of infection (respiratory tract infections), though they may get infected primarily also. During the course of infection, the cystic hygroma increases in size and becomes warm, red and tender. The patient may become febrile. The infection may involve the entire cystic hygroma or a few of its cysts. During active infection, it may not remain trans illuminant. Sometimes cystic hygroma turns into an abscess, which needs to be drained for amelioration of the symptoms. The treatment is conservative with antibiotics, antipyretics and analgesics. It is our observation that in some instances, post-infection, the size of the cystic hygroma reduces remarkably. The infective agent in the case described probably incurred an inflammatory reaction in a similar manner causing fibrosis and resolution of the lymphangioma.

Conclusion
We have reported four cases of cystic hygroma with spontaneous regression due to acute infection. There are reports of chronic infection in them, which subsequently led to regression, but we did not find any report in the literature in which there was spontaneous regression of primary as well as recurrent regression of cystic hygroma due to acute infection within 1-2 weeks.

REFERENCES