CASE REPORT

Spontaneous resolution of a recurrent axillary cystic hygroma following acute infection

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SUMMARY

We report a case of spontaneous resolution of a recurrent axillary cystic hygroma in a 4-year-old boy. He presented with a 1-year history of a cystic lump in the left axilla, which intermittently changed in size. Ultrasound suggested it was a lipoma, with raised suspicions of vascular malformation. Scans were discussed in a multidisciplinary meeting and provisional diagnosis of lymphocele or slow flow lymphovascular malformation was made. It was surgically excised and histology confirmed the lesion to be a cystic hygroma. However, it recurred within 3 weeks. The patient was booked for aspiration and treatment with sclerotic agent OK 432. He developed acute infection in the cystic hygroma a week before surgical intervention and was treated with antibiotics for 5 days by his general practitioner. Acute infection led to complete spontaneous resolution of the cystic hygroma within a week. There are no other reported cases in which recurrent cystic hygroma has resolved after a week of acute infection.

BACKGROUND

Although uncommon, there is a chance of spontaneous resolution of a cystic hygroma (CH) and thus a 'watch and wait' approach is sometimes adopted in infants unless complications occur.¹ The recurrence rate following surgical extirpation is nearly 10%, and is noted more with those in extra parotid and suprahyoid locations. Most of the recurrences manifest within the first year and are due to residual cysts that grow with the patient. Complications of surgery include cranial nerve palsies, cosmetic defects, dysphagia and airway compromise. We report a case of recurrent CH with spontaneous resolution within 1 week following an inflammatory response from acute infection. We searched the literature and found no case reports of this nature.

CASE PRESENTATION

A 4-year-old boy was referred to the paediatric surgery department because of a lump in the left axilla for 1 year; it had increased and then subsequently decreased in size over the past 3 months (figure 1). There was some mild discomfort but there was no pain, irritation, discharge or change of colour. Movement of his arm and neuromuscular examination was normal. His parents had noticed that, off and on, there was some bluish discolouration of the lump. It was about 5 cm in size, soft and mobile, did not appear to be attached to

any deeper structures, and had no associated lymphadenopathy.

INVESTIGATIONS

An ultrasound was carried out at the referring hospital, which suggested that it was a lipoma, but with suspicions raised that it might be a vascular malformation, as it increased as well as decreased in size (figure 2). Ultrasound scans were discussed in a multidisciplinary meeting and a provisional diagnosis of lymphocele or slow flow lymphovascular malformation was made.

DIFFERENTIAL DIAGNOSIS

Lipoma, vascular malformation, lymphocele, cystic hygroma, lymphovascular malformation.

TREATMENT

After discussion with the patient's parents, a decision was made to remove the cyst as it was causing some discomfort when it swelled up. Surgery was not our preferred approach as first choice in treating this child, but the reason for operating was that we were not clear about the diagnosis. During the surgical procedure, it was noticed that the lump was cystic in origin and had deeper extensions into the axilla, with fluid filled cystic components, and was very difficult to dissect. It was invading into deeper structures around the brachial plexus and it

Figure 1 A lump 5 cm in diameter in the left axilla, causing mild discomfort but not causing pain, irritation or discharge, and with no change of colour.

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Figure 2 Ultrasound images of axillary lump suggestive of lipoma or vascular malformation.

was not possible to dissect the entire cystic element. It appeared to be a cystic hygroma. A careful dissection was performed and the cystic lesion was removed and sent for histopathological examination. A drain was left in situ for 3 days, and the child was given antibiotics for 5 days. Histology showed connective tissue with vascular spaces focally featuring lymphoid tissue. Features were consistent with cystic lymphangioma/lymphovascular malformation (CH).

OUTCOME AND FOLLOW-UP

The child was seen again in our outpatient clinic, with re-occurrence of the lump 21 days after the surgery. Histology of the dissected cystic structure confirmed that it was a cystic hygroma. 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 to have the procedure, but there was no lump at all (figure 3). According to his parents, the child had a local

infection 7 days prior, for which he had been on oral antibiotics for 5 days (started by his general practitioner) and the swelling/lump resolved completely. There was no swelling at all and no clinical findings. There was no thickening of tissues felt. The local infection in the lump caused the spontaneous regression of the lump within 1 week without any surgical intervention. In this case, the recurrent CH had resolved within a week, due to local infection.

DISCUSSION

There are reports of watchful waiting for CH in patients who are asymptomatic. Medical treatment of CH consists of the administration of sclerosing agents, such as OK-432 (an inactive strain of group A *Streptococcus pyogenes*), bleomycin, pure ethanol, sodium tetradecyl sulfate and doxycycline.¹

An infected CH should be treated with intravenous antibiotics, and definitive surgery should be performed once the infection has resolved. Incision and drainage or aspiration results only in temporary shrinkage, and subsequent fibrosis can further complicate the resection. Radiotherapy has not been demonstrated to be effective. The mainstay of treatment of CH is surgical excision.² Microcystic lesions are much more difficult to remove because of their intimate association with nearby tissues. The exceptions to excision at the time of diagnosis are few and include premature infants who are small in size, and those with involvement of crucial neurovascular structures that are small and difficult to identify (eg, facial nerve). If no airway obstruction is present, surgery can be delayed until the child is aged 2 years or older, especially when the operation will be around the facial nerve in the parotid area. Signs of airway obstruction necessitate surgical intervention.

CH can present on routine prenatal ultrasonography as a large obstructing airway mass, as can other pathological conditions (eg, teratoma or rhabdomyosarcoma). If such a mass is visible on ultrasonography, MRI should be performed to delineate the mass further. In these cases, a multispecialty team, including a high-risk obstetrician, paediatric otolaryngologist, paediatric surgeon and neonatologist, should be present at the ex-utero intrapartum treatment (EXIT) procedure.

Several studies have shown an increase in rates of recurrence, morbidity and complications for lymphangiomas located in the suprahyoid versus infrahyoid region.³ CH is a benign pathology and there is no need for aggressive surgery at the cost of complications. Complete excision of a CH has been shown to have an 81% cure rate. When only part of the lymphatic malformation is excised, there is an 88% recurrence rate.⁴ Spontaneous

Figure 3 Appearance of axilla showing spontaneous resolution of cystic hygroma 7 days postlocal infection treated with oral antibiotics.

regression of a CH without treatment is occasionally reported and occurs with a frequency of 1.6–16%.⁵

Infections of CHs are common, especially in the suprahyoid microcystic and mixed variety forms.⁶ Infection causes inflammation and swelling of the cyst, which 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.⁶ Steroids also convey some benefit in acute infection.⁶ 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, with well-defined lymphangiomas confined to the posterior triangle having the best results after surgery.⁵ Treatment of cystic hygromas is usually sought when the lesions pose cosmetic problems, or get infected or traumatised. There are multiple treatment options including aspiration, laser, radiofrequency ablation, sclerotherapy and surgical excision.⁷ Sclerotherapy or surgical excision is the mainstay of treatment. Neither method has shown superiority over the other.⁸ Both treatments are associated with nerve damage and infection.⁸ In our case report, the resolution of the CH postinfection gave the same picture as resolution after a postsclerosing agent or blockage of lymphatic channels.

Sclerosing agents and radiation therapy have not been shown to play a role in the primary treatment of CHs. They may be indicated in lymphatic malformations that are macrocystic and not amenable to surgical resection. Sclerotherapy with OK-432, a lyophilised, low-virulence *S. pyogenes* strain of group A *S. pyogenes*, has been suggested as a possible therapy for macrocystic lesions. To date, however, the efficacy of OK-432 has not been proven in prospective, controlled trials.⁹ A sclerosant used in treating CH, OK-432 (picibanil), is derived from a low virulent strain of *S. pyogenes* treated with benzylpenicillin potassium.¹⁰ It is thought that an inflammatory reaction to the agent causes fibrosis and subsequent resolution of the CH. The infective agent in the case described probably incurred an inflammatory reaction in a similar manner causing fibrosis and resolution of the lymphangioma. Intralesional bleomycin as a sclerolant can cause pulmonary toxicity, which is the most serious potential side effect of bleomycin therapy. It is of importance to stress that old sclerosing materials such as bleomycin, ethibloc, tetracyclin, dextrose and sodium morrhuate, are known to cause scarring and contraction of the surrounding tissues, rendering subsequent surgery more difficult.

Learning points

- Cystic hygromas can re-occur despite surgical excision.
- Treatment options include sclerotherapy, aspiration, surgical excision, laser and radiofrequency ablation.
- Acute infection of a cystic hygroma can lead to rapid spontaneous resolution.
- Infection should be monitored closely as it can aggravate the cystic hygroma.

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