

Lymphoscintigraphy for Cystic Hygroma

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We present the case of an infant with a cystic hygroma investigated by lymphoscintigraphy. The source of lymph flowing into the hygroma was identified as were some ectatic lymphatics feeding it. Lymph flow was very rapid. The implications for therapy are discussed. Lymphoscintigraphy can assess many aspects of lymph flow not accessible by other means.

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Investigation of cystic hygroma at present includes ultrasound (US) and computerized tomography (CT), with CT the method of choice for delineating the extent of the lesion if surgery is planned. Arteriography may also be done to be sure the lesion is not of vascular origin. These imaging techniques, however, cannot assess lymph flow. Technical difficulties make lymphangiography virtually impossible in the head and neck area, and in small infants.

Lymphoscintigraphy is a simple technique, yielding valuable information (*1*) such as, the source of lymph draining into the hygroma, rate of flow, the presence of ectatic lymphatics, rate and extent of spread of lymph through the hygroma, extent of the hygroma, and patency of lymphatics and lymphatico-venous anastomoses in various sites in the body. In addition lymph nodes can be visualized and lymph node drainage patterns determined by lymphoscintigraphy.

A case is presented of an infant with a cystic hygroma investigated by lymphoscintigraphy. The implications of the findings are discussed.

CASE REPORT

The patient was a 3-mo-old caucasian female who had previously undergone two surgical attempts at excision of a cystic hygroma on the right side of her neck. The hygroma recurred after each attempt. It was thought that the hygroma

was being fed by the thoracic duct and that ligation of connections to the thoracic duct would be required. The infant was referred for assessment of lymph flow, specifically to determine if the thoracic duct was feeding the hygroma.

A percutaneous drain, which had been situated in order to drain the hygroma, was clamped prior to lymphoscintigraphy. A subcutaneous injection of 250 μCi (10 MBq) $^{99\text{m}}\text{Tc-Sb}_2\text{S}_3$ (antimony sulfide) colloid in 0.1 ml was given just caudad to the right mid-clavicle. This site was chosen with the view that, if there were aberrant lymphatic drainage pathways, drainage should occur from this site via the right axillary channels into the abnormal channels, then into the cystic hygroma on the right side of the neck. With the infant in supine position, head turned to the right, imaging was carried out on a General Electric Gemini gamma camera attached to a General Electric Star II computer (Milwaukee, WI) using zoom mode and a 15-sec framing rate. A rapid framing rate was chosen since the rate of lymph flow in infants is not known. It became clear this framing rate was not sufficient for our needs.

At 15 min a faint amount of the radiocolloid could be seen in the liver and bladder, but none in the region of the cystic hygroma. One half-hour after the first injection, a subcutaneous injection was given in the right parietal area of the scalp. Within seconds, flow around the posterior of the right ear into the hygroma was seen on the monitor. The hygroma filling continued for a few minutes (Fig. 1). Another injection was given in the anterior midline at the hairline. Again flow into the hygroma was seen within seconds. What appeared to be ectatic lymphatics, draining around the anterior and posterior of the right ear, were also seen (Fig. 2). Foci of persisting collections around the right ear were seen, probably representing colloid uptake in the lymph nodes (Fig. 2). Images from other views, at 30 min after the third injection, showed drainage occurring via the left side of the head (Fig. 3).

DISCUSSION

Prior to lymphoscintigraphy, given that two previous attempts at surgical excision had resulted in recurrence of the cystic hygroma, it was felt that the thoracic duct was feeding the hygroma and that it would be necessary to ligate the connections with the thoracic duct. Lymphoscintigraphy showed that the lymphatics feeding the hygroma came, in fact, from the head and not from below the neck. This indicated that ligation of the thoracic duct connections would not have been successful, and resulted in an infant being spared a surgical procedure with a high morbidity rate.

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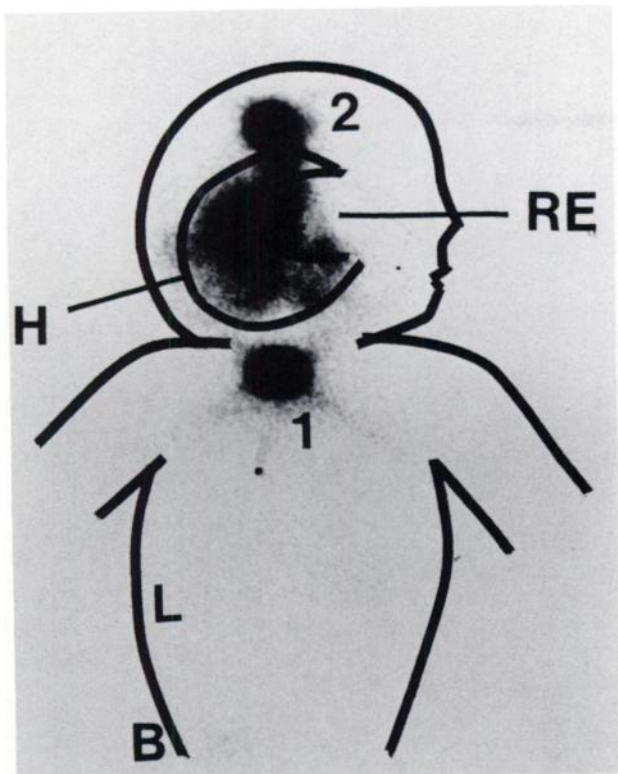


FIGURE 1
Right lateral view of head with anterior view of body 5 min after second injection. Injection sites 1 and 2 are labeled. L = liver, B = bladder, H = hygroma, RE = right ear.

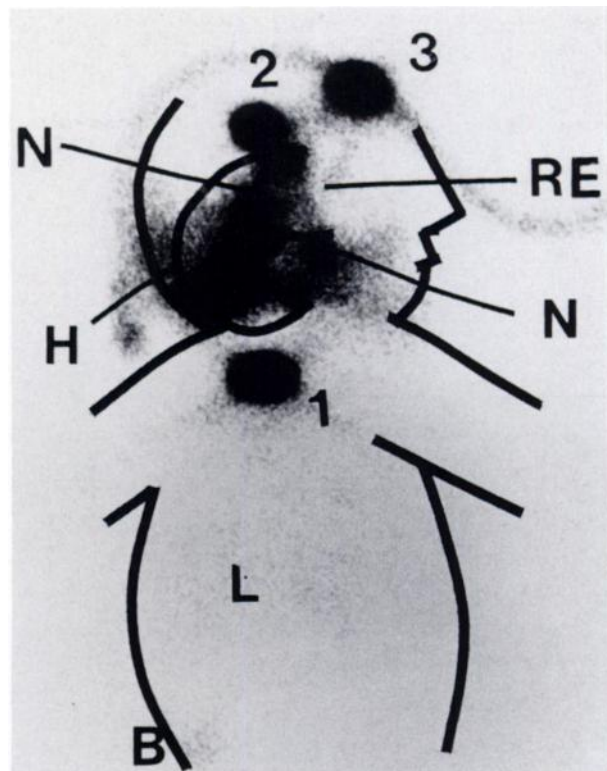


FIGURE 2
Fifteen minutes after third injection (labeled 3), a line marker is seen overlying the scalp from the nose to theinion. N = lymph node (only two are labeled). What appear to be ectatic lymphatics are seen between injection site 3 and the right ear (RE).

Imaging showed what appeared to be ectatic lymphatics feeding the hygroma. If a smaller field-of-view gamma camera with higher resolution were used, these vessels might have been seen more clearly. In retrospect, this might have raised the possibility of ligating these large vessels near their source or excising them back to their source, as an additional means of reducing the hygroma. Such a technique has not yet been tried in patients.

Since the previous interstitial injection had resulted in visible lymphatic movement, at a third operation Evans blue dye was injected subcutaneously, after surgical exposure, in order to visualize the leaking lymphatics on the operating table. The ectatic vessels were not visualized with the blue dye suggesting that these vessels were too deep. Leakage was seen within seconds and proceeded rapidly. The lymphatics leaking dye into the surgical field were ligated at their exposed ends but, unfortunately, the abnormal lymph drainage and accumulation recurred after surgery, possibly because the ectatic lymphatics were not excised or ligated at their source.

Surgical excision usually offers the best means for successfully reducing hygromas that do not resolve

spontaneously (2,3,4). A variety of sclerosing agents have been injected into cysts with varying success. Recently, bleomycin in a microsphere-in-oil emulsion has been injected intralesionally as a sclerosing agent (5), with promising results. However, those results appeared to be related to the ability of the bleomycin in oil to penetrate and remain in the hygroma for a prolonged period. The authors also found cystic hygromas more amenable to this sclerosing agent than capillary or cavernous ones. Lymphoscintigraphy might be helpful in delineating which hygromas could be successfully treated in this manner. The imaging agent could also be used in a microsphere-in-oil emulsion for this assessment, since different preparations do not have the same flow dynamics. A hygroma that could be fully delineated by injection and showed prolonged retention would be considered for sclerosis by this means. Promise has also been seen using group A streptococcus pyogens for sclerosis (6).

Various radiopharmaceuticals are available for lymphoscintigraphy. In situations in which assessment of lymphatic function is desired, it may be desirable to use a more physiologic agent such as ^{99m}Tc -labeled human serum albumin (7).

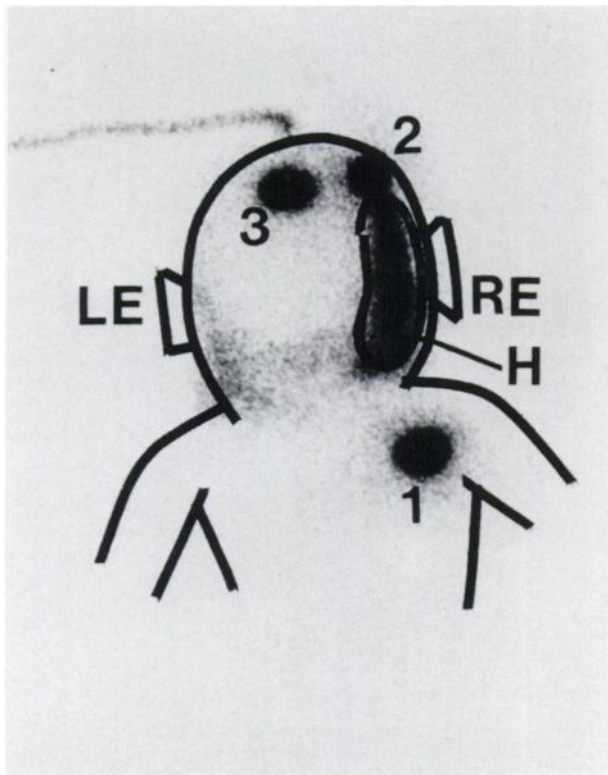


FIGURE 3
Posterior view of head 30 min after third injection. The linear marker has been lifted off the posterior scalp. LE = left ear. There may be collateral circulation around the posterior upper neck from right to left. What appears to be drainage from the third injection down the left side of the head is seen.

SUMMARY

The lymph flow rate in the infant cited here appeared very rapid, but may simply reflect a difference between infants and adults. Even what were felt to be ectatic lymphatics showed very rapid flow.

Lymphoscintigraphy is a simple technique, which often yields as much or more information than lymphangiography in the investigation of lymphedema (8,9,10). Lymphoscintigraphy has also been found to be useful in the diagnosis of lymphocele (11) and tumor spread to regional lymph nodes (1). Lymphoscintigraphy now also appears useful in the investigation and planning of treatment for hygromas.

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