Unilateral limb enlargement in an infant is a diagnostic problem compounded by the patient's size. It may not be possible or feasible to perform special radiographic procedures because of the small size of the structures involved and the need for general anesthesia. Lymphedema in the lower extremity of an infant was studied by isotope lymphangiography—a safe, relatively easy method of examining lymphatic circulation, which does not require general anesthesia, skin incision, or lymphatic dissection.

CASE REPORT

A white female infant had a swollen right foot noted soon after birth. By her fourteenth week the foot and leg were grossly edematous; the circumferences in different regions on the right foot and leg were 2 cm greater than corresponding regions on the left (Fig. 1A). The right thigh was involved to a lesser degree. Both legs were of equal length and temperature. There was no venous dilatation or arterial insufficiency. Inguinal lymph nodes were not palpable. There was no history of trauma, infection, nor familial occurrence of lower-limb asymmetry.

A radiogram made when the infant was 15 weeks old (Fig. 1B) showed a normally sharp muscle-fat interface on the left, but this was hazy and obscure.

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**FIG. 1.** A shows lower extremities of 15-week-old female with edematous right leg and foot. B is radiogram demonstrating obscure muscle-fat interface on right.
on the right side, indicating edema. The bones were of equal length.

Because conventional lymphangiography is impractical in a 15-week-old child, isotope lymphangiography was performed, and 25 μCi of 198Au-colloid gold in a 0.2-ml volume was injected subcutaneously into the first web space of each foot. Scans were performed at 24, 48, and 96 hr postinjection on a single-probe 5-in. rectilinear scanner. The scan area extended from the foot to the liver (Fig. 2). Although all scans failed to show right inguinal and femoral lymph nodes, the nodes were visualized well on the left side. Sufficient radiogold was picked up by the Kupffer cells of the liver to image that organ. Radiogold remained in the right foot while decreasing in the left. Primary, congenital, nonfamilial lymphedema was the diagnosis, and further studies were not indicated.

DISCUSSION

Primary lymphedema is subdivided into familial and nonfamilial types (1). Foremost among the familial types is Milroy's disease which is nearly always present at birth, involves one or both extremities, and occurs in both sexes (2).

Lymphedema is also classified according to the time of onset (3–6). Congenital lymphedema comprises 10–25% of the cases and may occur as nonfamilial lymphedema, Milroy's disease, or gonadal dysgenesis (Turner's syndrome). Lymphedema praecox presents before age 35 and includes 60–90% of the group. The remaining approximately 10% are referred to as lymphedema tarda. Nearly all cases (97%) appear before age 40 and occur ten times more frequently in females. Half involve both lower extremities.

Lymphangiography shows three types of abnormalities (1,3,6–9): (A) Hypoplastic channels are demonstrated in 55–70% of adult patients. (B) Individuals with an early onset of symptoms usually have aplastic lymphatic channels. (C) Hyperplastic lymphatic vessels characterized by pooling of contrast media in dilated channels due to inefficient valves occur almost as frequently as does the aplastic lymphatic variety. Bilateral lymphangiograms in 50% of patients with unilateral edema demonstrate an abnormality in the clinically uninvolved side. It may be impossible to perform lymphangiography in patients with aplastic or hypoplastic lymphatic vessels (10,11). A deterrent to lymphangiography in children under 5 is the requirement for general anesthesia (12).

Colloidal gold (198Au) is the preferred radiopharmaceutical for isotope lymphangiography (13–15). The 198Au leaving a subcutaneous injection site enters the lymphatic system to be deposited in lymph nodes along the route. After a node is saturated, subsequent colloidal gold passes onto further nodes. The 198Au trapped in a given node remains there. Lymph nodes along a chain filter out most of the nuclide but some will bypass all nodes to enter the thoracic duct and then the blood from which it is cleared by the reticuloendothelial cells of the liver and spleen.

Our technique involves the subcutaneous injection of 15–100 μCi of 198Au into the first web space of each foot. Scanning is performed at 24-hr intervals for the next 3 or 4 days. The ileofemoral lymph nodes have maximum concentration of 198Au but paraaortic lymph nodes and liver pick up sufficient amounts to be visualized. Blockage of lymph flow is reflected by decreased or absent radioactivity in the involved region. Absent radioactivity in the liver at 24 hr indicates lymphatic obstruction.

Isotope lymphangiography was the only feasible way of studying this 15-week-old child's aplastic lymphatic system. In Gasquet's series (12) of conventional lymphangiography in children, the youngest successful study was in a 17-month-old child; none was attempted at less than 6 months of age.

The lymph scan presented here showed absence of 198Au in the right inguinal and femoral lymph node groups at both 24 and 96 hr after injection. The abnormalities could be explained adequately only by aplasia of lymphatic channels. This correlates with the increased diameter of the right leg and foot and haziness of the normal muscle-fat interface on the radiogram. Since there was no family history of

FIG. 2. Colloidal gold (198Au) lymphangiogram shows no uptake in right inguinal or femoral nodes over 96 hr. Left side is visualized normally as is liver (tip of which is above letters "A" and "B").
lymphedema, the diagnosis was primary congenital, nonfamilial lymphedema.

SUMMARY

A 15-week-old infant with primary congenital lymphedema was evaluated by isotope lymphangiography. The scan showed absence of $^{198}$Au in the right inguinal and femoral lymph node groups at both 24 and 96 hr after injection. This is a safe, relatively easy, nontraumatic method for studying the lymphatic system of the infant.

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