structing sickle cell thrombi (7) and as oxygen inhalation therapy prevented further occlusive episodes.

CONCLUSION

Pulmonary scintigraphy can play an important role in diagnosing ACS as well as monitoring the effect of therapy on its evolution, particularly in the more infrequent cases when bigger vessels are involved.

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Splenogonadal Fusion Diagnosed by Spleen Scintigraphy

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Splenogonadal fusion (SGF) is a rare congenital malformation characterized by fusion of the spleen and a gonad (almost always the left one) frequently associated with orofacial and/or limb developmental abnormalities. Only 125 cases were reported between 1883 and 1994. This report concerns a case of SGF in a 20-yr-old woman with an accidental finding of a splenic space-occupying lesion protruding into the lower abdomen in ultrasound and CT. Radiocolloid spleen scintigraphy and SPECT proved to be the best procedure to establish the correct diagnosis of SGF. As SGF is often asymptomatic, more liberal use of splenic scintigraphy is suggested in patients with congenital limb and/or orofacial anomalies. SGF should be included among the differential diagnoses of left abdominal, pelvic or scrotal masses.

Key Words: spleen-congenital-anomalies; radionuclide-imaging; splenogonadal fusion; facial abnormalities

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CASE REPORT

A 20-yr-old female patient of Lebanese Jewish origin underwent medical investigation because of chronic and severe constipation for about 6 yr and diffuse abdominal pains for about 1 yr. Past patient history disclosed dysphagia and recurrent pneumonia during childhood, which ceased after tongue surgery at age 7–8 yr. At age 17, injection of Botulinus was performed in the right hemiface to correct congenital paresis of the left facial nerve. Only temporary partial cosmetic result was achieved. On physical examination, marked asymmetry of the face and jaw with signs of left facial paresis were noticed.

Routine laboratory tests showed only mild microcytic hypochromic anemia. Gastroscopy revealed mild gastritis with a positive test for *Helicobacter pylori*.

Colonoscopy demonstrated a polyp, 1 cm in size, located about 10 cm from the anus. The polyp was resected through the colonoscope, and its histology was compatible with benign juvenile polyp. Barium enema showed a markedly tortuous sigmoid colon and splenic flexure but no other abnormalities.

Abdominal ultrasound revealed a slightly enlarged (12-cm) spleen with homogenous structure and a space-occupying lesion of unclear nature in its medial aspect. Bipartite spleen was suspected.

Abdominal CT (Fig. 1) revealed a 6-cm solid structure in the left upper abdomen possibly linked to the spleen, next to the tail of the pancreas and the ventral surface of the left kidney. The caudal tip of this lesion extended to the pelvic entrance. After contrast injection, the lesion was enhanced in a similar degree to that of the spleen.

A first liver-spleen scintigraphy was performed in a clinic outside the hospital. It reported both normal liver and spleen. The scan itself was not available for review.

The patient was admitted to the department of internal medicine in our hospital for CT-guided needle biopsy of the space-occupying lesion. Cytologic examination of the needle biopsy material revealed lymphoid cells in different stages of maturation compatible with lymphoproliferative process. Bone-marrow biopsy was interpreted as normal.

The unclear clinical picture of anemia with confusing cytological and bone-marrow findings led to another liver-spleen scintigraphy. This time the scan was performed in our department of nuclear medicine.

Planar scintigrams were performed 30 min after intravenous injection of 148 MBq (4 mCi) of ^{99m}Tc-CaNaPhytate (Soreq Radiopharmaceuticals, Israel) on Elscint Apex-410 gamma camera with low-energy general purpose collimator (Fig. 2).

Liver-spleen SPECT was performed on an Elscint SP-4 camera with low-energy high-resolution collimator. Data were acquired in 64 × 64 pixel frame size, 60 frames over a 360° orbit, 30 sec each stop. Uniformity corrected frames were prefiltered by a Metz filter set to FWHM 10 mm and order 5, then reconstructed to standard transaxial, coronal and sagittal slices. For best visualization, modified coronal (oblique) slices of the spleen (Fig. 3) were obtained also.

Both planar scintigram and SPECT revealed a spleen having abnormal shape with a tail-like projection originating from its lower pole extending through the lower abdomen to the pelvic

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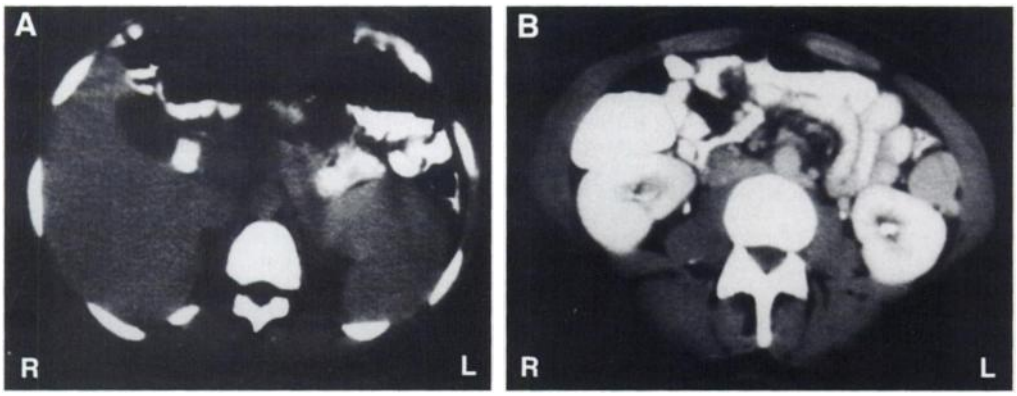

FIGURE 1. Selected slices of abdominal CT scan. (A) Note the widened and distorted lower pole of the spleen mimicking the space-occupying lesion. (B) More caudal slices showed an elongated structure adjacent to the lateral abdominal wall and the anterior surface of the left kidney.

entrance. The scan finding was interpreted as a continuous type of splenogonadal fusion. The suspicion of left abdominal lymphoproliferative process was excluded, and the patient was discharged from the hospital for ambulatory follow-up.

DISCUSSION

SGF is a rare anomaly with a wide spectrum of severity first reported more than 100 yr ago (*1–3*). With the most recently published cases, the number of recognized cases of SGF reported in the medical literature is 125 (*4,5*); only one scintigram of this anomaly was published (*6*). According to Putschar and Manion (*1*), SGF can be of a continuous or discontinuous type.

The continuous form of SGF appears as an uninterrupted cord-like structure between the spleen and the left gonad containing splenic and/or fibrous tissue. It frequently contains a vascular packet originating from the spermatic or ovarian artery or vein. Orofacial and severe limb malformations, including complete absence of limbs, are more frequent in this form of SGF.

The discontinuous form appears as an isolated pelvic or scrotal mass of splenic tissue attached to the testis or the ovary; it is less often associated with skeletal and orofacial malformations. In many case reports, this presentation was labeled as pelvic accessory spleen (*7,8*) or as ectopic accessory spleen (*9*).

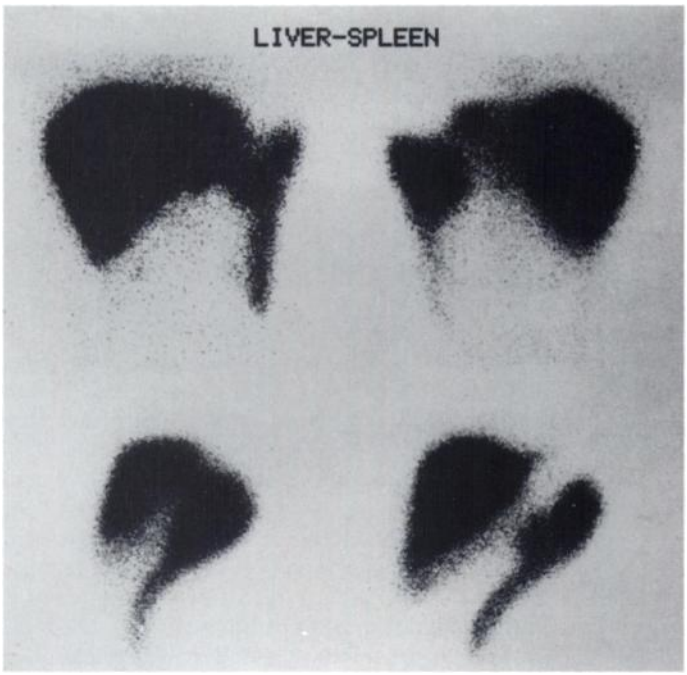

FIGURE 2. Planar radiocolloid scintigram of the liver and spleen. Note the tail-like extension of splenic tissue to the lower abdomen. The film is slightly overexposed to show better the thin cord of splenic tissue.

The etiology of the malformation is obscure. Most probably it arises during the fifth through eighth week of embryonic life when the developing spleen is in intimate contact with the mesonephric-gonadal anlage and when the Meckel's cartilage and the extremity buds are differentiating. A diffuse but nonfatal injury to the fetus occurring at this time and interfering with the development of these three elements could explain the quite odd association of these otherwise rare malformations. For more detailed discussion on SGF etiology, the reader is referred to the extensive study of Putschar and Manion (*1*) and more recently of Pauli and Greenlaw (*3*).

Review of the relevant medical literature raises the possibility that SGF might be overlooked by many physicians in different specialties of medicine. Most of the cases were diagnosed as an incidental finding during various surgical procedures or at autopsy (*7–12*). Awareness of the existence of this rare condition may reduce the number of unnecessary diagnostic procedures and eliminate invasive diagnostic interventions and unneeded surgical procedures. It is important to include SGF among the differential diagnoses of left, lower abdominal, pelvic and scrotal masses (*3,11,12*).

As this case demonstrates, splenic scintigraphy is the modality of choice for diagnosis of SGF. It should be performed whenever an anomaly associated with SGF exists and in any patient with unexplained left abdominal, pelvic or scrotal mass. Since SGF is often asymptomatic and discovered incidentally, Pauli and Greenlaw (*3*) suggested prospective ultrasound and/or scintigraphic screening of all patients with Hanhart syndrome (congenital limb deficiency with mandibular, oral and palatal abnormalities) and Möbius syndrome (congenital facial and abducens diplegia) in order to study the relationship of SGF and these anomalies.

Care should be taken to obtain good scintigraphic visualization of the spleen, the whole abdomen and pelvis. The scrotum should be scanned in case of scrotal mass. As the bridge of splenic tissue connecting the spleen and the left gonad may be thin or discontinuous, it is imperative to obtain scans of impeccable quality. The appropriate use of image processing may be required to enhance the low-count density of thin or small splenic tissue fragments. It is assumed that the initial false-negative spleen scan was caused by high-contrast technique. The use of SPECT technique may further improve lesion detectability and should be performed in cases of doubt.

CONCLUSION

Radiocolloid spleen scintigraphy, preferably enhanced by SPECT, is the modality of choice in diagnosing SGF. The

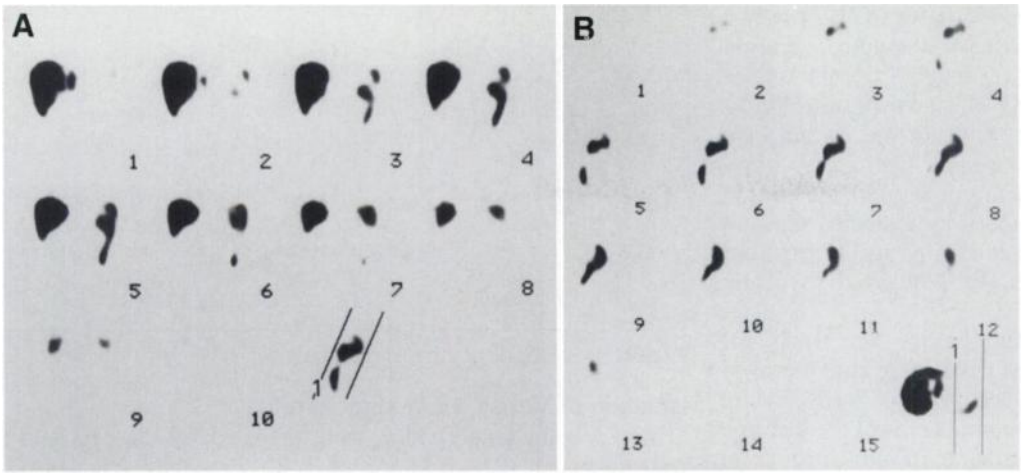

FIGURE 3. Selected (A) oblique and (B) sagittal slices of the liver-spleen SPECT. The original coronal slices were slightly reoriented to display better the origin of the anomalous splenic tissue at the lower pole of the spleen. With SPECT no further image enhancement is necessary.

noninvasiveness, relatively low price, sensitivity and specificity make scintigraphy well suited for screening purposes.

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Copper-62-ATSM: A New Hypoxia Imaging Agent with High Membrane Permeability and Low Redox Potential

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An ideal hypoxia imaging agent should have high membrane permeability for easy access to intracellular mitochondria and low redox potential to confer stability in normal tissue, but it should be able to be reduced by mitochondria with abnormally high electron concentrations in hypoxic cells. In this context, nitroimidazole residues are not considered to be essential. In this study, Cu(II)-diacetyl-bis(N^4-methylthiosemicarbazone) (Cu-ATSM), a ^{62}Cu-bisthiosemicarbazone complex, with high membrane permeability and low redox potential, was evaluated as a possible hypoxia imaging agent, using electron spin resonance spectrometry and the Langendorff isolated perfused rat heart model as well as rat heart left anterior descending occlusion model. **Methods:** Nonradioactive Cu-ATSM was incubated with rat mitochondria, after which reduction of Cu(II) to Cu(I) was measured with electron spin resonance. As a model of hypoxic mitochondria, rotenone (Complex I inhibitor)-treated mitochondria were used. **Results:** In this study, Cu-ATSM was reduced by hypoxic but not by normal mitochondria. **Conclusion:** Thus, retention of ^{62}Cu-ATSM was studied serially in perfused rat hearts under conditions of normoxia (95% O_2 + 5% CO_2), hypoxia (95% N_2 + 5% CO_2) and reoxygenation (95% O_2 + 5% CO_2). In normoxia and reoxygenation, ^{62}Cu-ATSM injected as a single bolus showed low retention (23.77% and 22.80%, respectively) 15 min after injection, but retention was increased markedly under hypoxic conditions (81.10%). Also, in the in vivo left anterior descending occluded rat heart model, ^{62}Cu-ATSM retention was inversely correlated with accumulation of ^{201}Tl, a relative myocardial blood flow marker.

Key Words: copper-62-ATSM; hypoxia; mitochondria

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Hypoxic tissue is important for detection of ischemia in the brain, heart and other tissues, as well as tumors. Nitroimidazole compounds, originally developed as radiosensitizing agents (*1*), are of great interest because of their selective accumulation in

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