The occurrence of aseptic meningitis probably due to ECHO virus 1 disease has been reported together with ECHO virus 1 infection has been implicated in central nervous disease, especially aseptic meningitis, pericarditis, pleurodynia, diarrhea, undifferentiated upper respiratory infection, paralytic disease, exanthema, myalgia and ocular disease. However, a review of the literature would indicate that there have been no reports of isolations of ECHO virus 1 from the cerebrospinal fluid (CSF) of an adult with aseptic meningitis. Further, there have been no reports of ECHO virus 1 aseptic meningitis with simultaneous involvement of the pericardium, myocardium and liver.

Case Report

History: The patient, an 18-year-old white diabetic youth, was originally seen by one of us (L.A.S.) on October 15, 1969. His illness had begun seven days earlier with generalized aching and an oral temperature of 101°F. He became afibrile for several days and on the day of his examination his temperature climbed back to 104°F and he was nauseated, vomited several times and complained of a splitting headache. He had photophobia and severe retrobulbar pain. He did not have a sore throat, chest pain or cough and no diarrhea or urinary frequency.

The patient's diabetes was ushered in by an acute, severe episode of acidotic diabetic coma nine years ago. He had been taking 62 units of NPH insulin daily with a 2800 calorie diabetic diet. He had no interim history of acidosis, acetonuria or hypoglycemia.

The patient's system review was negative for congenital or rheumatic heart disease. He had no previous history of jaundice or infectious mononucleosis. There was no recent history of parenteral skin penetration, shell fish ingestion, or contact with children or infants with any febrile disease or diarrhea. He was admitted to the hospital on the same day at 2PM.

Physical Examination: The patient was an acutely ill white youth. His temperature was 102°F (rectally), his pulse 96 per minute and regular with respiratory rate of 24 per minute and a blood pressure of 100/80 mm Hg. His upper and lower eyelids were swollen and the conjunctivae were suffused, but the sclerae were not icteric. There was adenopathy, particularly one gland in the left anterior cervical triangle was swollen, painful but easily movable. There was similar lymphadenopathy of both axillae. There was no neck vein distention. The lungs were clear to percussion and auscultation. The patient had a grade 1 systolic murmur over the apex and aorta. This same murmur was heard before this acute illness and remained unchanged in character and intensity. It was not transmitted to the axilla but transmitted to the neck. The patient did not have paradoxic pulse. There was evidence of nuchal rigidity. The remainder of the physical and neurologic examination was within normal limits.

Laboratory Data: Urinalysis revealed evidence of occult blood (Hemastix), 2+ sugar (Clinistest tablets), negative test for acetone and protein, specific gravity of 1.013 with a pH of 7.4. Microscopic examination of the urinary sediment revealed two to three white blood cells and one to two red blood cells per high-powered field. He had a hemoglobin of 15.1 gm per 100 ml, hematocrit 43 percent, white blood cells 8600 with 70 percent neutrophils, 10 percent band forms, 13 percent lymphocytes, 2 percent atypical lymphocytes and 5 percent monocytes. A lumbar puncture performed on admission revealed an opening pressure of 180 mm Hg, a closing pressure of 120 mm Hg and normal fluid dynamics. The cerebrospinal fluid (CSF) glucose was 120 mg/100 ml (normal 40 to 80 mg/100 ml), a concomitant blood glucose

Spectrum of ECHO Virus 1 Disease in a Young Diabetic

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The occurrence of aseptic meningitis probably due to ECHO virus 1 disease has been reported together with significant myopericarditis, abnormal liver function tests, conjunctivitis, lymphadenopathy, pyuria and hematuria. Virus isolation and identification was from the spinal fluid. No previous report of such isolation in an adult exists in the literature.

ECHO virus 1 was first isolated from the stools of Egyptian children by Melnick and Aagren in 1952.1 ECHO virus 1 infection has been implicated in central nervous disease, especially aseptic meningitis, pericarditis, pleurodynia, diarrhea, undifferentiated upper respiratory infection, paralytic disease, exanthema, myalgia and ocular disease. However, a review of the literature would indicate that there have been no reports of isolations of ECHO virus 1 from the cerebrospinal fluid (CSF) of an adult with aseptic meningitis. Further, there have been no reports of ECHO virus 1 aseptic meningitis with simultaneous involvement of the pericardium, myocardium and liver.
was 210 mg/100 ml. The CSF protein was 36 mg/100 ml (normal 15 to 45 mg/100 ml). Microscopic examination of the CSF showed 165 white blood cells per cubic millimeter and one red blood cell per cubic millimeter. Of the total 165 white blood cells there were 71 polymorphonuclears (43 percent) and 94 lymphocytes (57 percent). No organisms were found on the Gram stain and examination of the spinal fluid for cryptococci gave negative results. Culture of the spinal fluid was negative for bacteria and fungi.

Other normal admission laboratory tests were serum amylase, CO₂, potassium, serum heterophile agglutination test, febrile agglutinations and VDRL. Staphylococcus aureus was isolated from his pharynx.

The patient had persistent elevation of serum glutamic pyruvic transaminase lasting for five days with elevations up to 85 Karmen units (normal 0 to 45 Karmen units) also the alkaline phosphatase was elevated up to 3.8 Bessy Lowery units (normal 0.8 to 2.3 Bessy Lowery units) and remained elevated for two weeks. A creatinine clearance and Addis count would have been of interest but were not done. A test for serum guanase was performed on October 18, 1969 and showed 19 nanomols per millimeter per minute (normal 0 to 3 nanomols per millimeter per minute). An ornithine carbamyl transferase test (OCT), also performed on the same day was 15.0 nanomols of radioactive CO₂ (upper limits of normal being up to five nanomols of radioactive CO₂).

Serial electrocardiographic tracings from October 15, 1969 until October 21, 1969 were compatible with the pericarditis and myocarditis (Fig 1). The tracing of January 23, 1970 was normal and subsequent tracings have remained normal to date. There was no other occurrence of arrhythmias except the premature atrial contractions as seen on October 16, 1969 (Fig 1). There was no block and no Q wave changes developed.

Clinical Course: The patient continued to complain of headaches, retrobulbar ocular pain and poor appetite. His temperature dropped to 99°F (rectally on the second day of his admission. At no time did the patient complain about chest pain or shortness of breath. He was discharged to home care on October 21, 1969. His outpatient follow-up was characterized by fatigue, lassitude, headaches and constant anorexia. He had several incidences of hypoglycemia in spite of consecutive reductions of insulin dosage. He was advised against returning to work for one month and told to avoid strenuous activity for three months. When he was reexamined in April 10, 1971, his physical examination was within normal limits, except for the systolic murmur which was present before his acute illness. At that time he was exercised on a treadmill and easily achieved a pulse rate of 180 beats per minute without any T or ST abnormalities during the test.

Virologic Studies: ECHO 1 virus was isolated in Rhesus monkey kidney tissue culture inoculated with the cerebrospinal fluid obtained on the ninth day of disease, October 16, 1969 (the isolate was obtained on second passage). No cytopathogenic agents were isolated from pharyngeal secretions or from a rectal swab extract obtained on the same day.

Both sera obtained on the ninth and 77th days after onset contained homologous neutralizing antibodies at a titer of 1:8. A later convalescent serum obtained on June 18, 1970 also contained homologous neutralizing antibodies at a titer of 1:8. Attempts to isolate the virus from the conjunctivae and urine were not made.

FIGURE 1. Representative serial electrocardiograms of a young diabetic patient with ECHO virus 1 disease demonstrating ST elevations and reciprocal depressions on October 16 and October 18, 1969. Tachycardia and relatively low voltage is seen on the electrocardiogram of October 16, 1969. There is also graphic evidence of a premature atrial contraction in AVL on October 16, 1969. Although ST segment elevations were present in V₅ on October 16, 1969, they are not shown in order to conserve space. The ST elevations in V₅ were identical to those in V₆. The standardization was the same in both tracings.

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ANOMALOUS ORIGIN OF LEFT PULMONARY ARTERY

The patient's appetite remained depressed for a month, and others, but not as often as Coxsackie viruses.

Since carditis due to ECHO viruses has been associated with serious consequences, including life-threatening arrhythmias and even death, the diagnosis of asymptomatic or truly benign pericarditis or myocarditis occurring with ECHO viruses is obviously important. Therefore, the so-called "benign" pericarditis associated with enterovirus disease may be a somewhat misleading classification.

The electrocardiographic changes in our patient were thought to be consistent with viral pericarditis and myocarditis. There were no historical or clinical grounds to associate them to coronary artery disease, hypokalemia, alkalosis or severe central nervous system disease.

The elevations of the serum glutamic pyruvic transaminase pointed to liver involvement. There was a sustained rise in the alkaline phosphatase. Confirmation of the hepatic origin of these admittedly small enzyme changes were furnished by the simultaneous elevation of the patient's serum guanase and his ornithine carbamyl transferase. These two latter enzymes, particularly guanase, are specific for parenchymatous liver disease.

The patient's appetite remained depressed for a month, and he was found to be hypoglycemic on several occasions despite reduction of insulin dosage. This anorexia might possibly have been due to hepatic involvement. Hepatitis associated with ECHO 9 virus disease has been reported in a previous communication by us.

A recent editorial in the Journal of the American Medical Association appears pertinent in regard to this case. The editorial writer states that isolation and identification of ECHO virus from the spinal fluid is preferable for definitive diagnosis in patients with this type of meningitis as compared to isolation from the throat and feces. The former was accomplished in our patient.

The pathophysiology of cardiac involvement in enterovirus disease is a subject for a recent editorial by Lerner. He points out the occurrence of an early acute infectious phase and prolonged auto-immune and recovery phase of entero viral myocarditis. Accordingly, our patient was not permitted to return to work until his electrocardiogram had become completely normal and his fatigue had subsided.

REFERENCES

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Anomalous Origin of Left Pulmonary Artery from Ascending Aorta, Right Aortic Arch and Right Patent Ductus Arteriosus*

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A case of anomalous left pulmonary artery (ALPA) arising from the ascending aorta, a right aortic arch and a right patent ductus arteriosus, a combination not previously reported, is described. Current concepts regarding the genesis of an ALPA also predict main pulmonary artery hypoplasia and defects of the ventricular septum. These associated defects were not present in our patient. Their absence suggests that the developmental faults associated with these abnormalities are even more complex than previously suspected. The need for early diagnosis and the efficacy of surgical correction are emphasized.

In 1969 Caudill and co-workers described a case of anomalous origin of the left pulmonary artery arising from the aorta and added the 35th such report to the world literature. Anomalous origin of the right or left pulmonary artery (ALPA, ARPA) from the aorta or its primary divisions (ie innominate) is a rare phenomenon although it was first recognized over 100 years ago. It was not until 1949, however, that Bopp demonstrated an ARPA at a postmortem examination.

The importance of angiographic studies to demonstrate the origin of both pulmonary arteries has been emphasized by several authors, as an ALPA or ARPA is a correctible lesion and successful surgical intervention has been reported. It is, of course, most important to meticulously seek out associated lesions which are so frequently present.

This presentation describes the association of an ALPA arising from the ascending aorta, a right aortic arch and a right patent ductus arteriosus, a combination which has not, to our knowledge, been previously reported. This association of defects is particularly noteworthy as it does not include pulmonary outflow anomalies or defects of the ventricular septum. These latter deficiencies would be anticipated by the current and

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