

A Rare Presentation of Postpump Hemichorea

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Postoperative complications after cardiac surgery are worrisome problems that occur in nearly half the operated pediatric patients, mostly when a cardiopulmonary bypass is needed [1]. Up to 5% of open-heart surgeries are associated with neurological complications. Movement disorders occur less frequently. The most common movement disorder in this setting is generalized choreoathetosis, also termed postpump chorea syndrome. The underlying mechanism is unknown.

The aim of the present study was to describe a rare case of hemichorea after open-heart surgery in a young patient.

PATIENT DESCRIPTION

The patient was a 9-year-old girl with a past medical history of patent ductus arteriosus that was closed by catheterization at 18 months of age. During the procedure, mild subaortic stenosis due to a fibrous membrane was found; therefore, the patient's condition continued to be followed by a cardiologist.

After 8 years, severe aortic stenosis developed, with signs of ischemia on a cardiac stress test. The patient was referred for subaortic membrane resection under cardiopulmonary bypass. The duration of the cardiac bypass surgery was 1 hour and 20 minutes. The minimal temperature was 30.1°C. No significant change in pH level occurred and circulatory arrest was not induced. The sur-

gery was successful, but several days later the patient began to exhibit involuntary movements of her left hand, which subsequently spread to the left leg. Her parents also noted a mild behavioral change manifested by angry outbursts.

Neurological evaluation revealed overt chorea of the left arm and left leg. There were no abnormalities in strength, sensory function, deep tendon reflexes, or cranial nerve function. The diagnosis of left-side postpump chorea was confirmed by normal findings on magnetic resonance imaging of the head and by laboratory tests that excluded other possible etiologies such as lupus or Sydenham chorea. Lupus anticoagulant, anti-cardiolipin, anti-dsDNA, and anti-beta2-glycoprotein antibodies were all within the normal range. Although the anti-streptolysin O antibody (ASLO) level was borderline (400 IU/ml, considered low positive), the patient had not complained of a sore throat and throat culture was negative.

Treatment with carbamazepine was initiated, leading to a resolution of the symptoms within a few days. Neurological examination at the 6-month follow-up yielded no abnormalities and no evidence of chorea.

COMMENT

We describe a rare case of hemichorea limited to the left side of the body in a child after cardiac surgery. Symptoms resolved after a few days of treatment with carbamazepine, and no long-term sequelae were noted.

Postpump chorea is a complication of open-heart surgery that was first described in the medical literature in the 1960s. The reported incidence of up to 18% of cardiac surgeries in the early studies has dropped considerably to 0.6% in recent studies pub-

lished by large tertiary medical centers [2]. This complication is rarely seen in adults [3]. The disorder usually appears a few days after surgery [2,3], as in our patient, although there are some reports of later onset of up to 2–3 weeks after surgery.

Symptoms usually resolve completely, although the interval from onset to improvement may vary from a few days to weeks, and in some persistent and severe cases, even for years [2,3].

Our case is unique in that the majority of reported cases of postpump choreoathetosis involved generalized choreoathetotic movements on both sides of the body whereas our patient manifested hemichorea involving the left arm and leg. Our patient had no additional clinically significant neurological deficits typically noted with postpump chorea. Findings on magnetic resonance imaging of the head were normal.

The mechanism underlying the development of chorea after cardiac surgery is unknown. Of the number of theories proposed to date, the most popular attributes this phenomenon to an ischemic-hypoxic injury to the basal ganglia secondary to the reduced blood flow during cardiopulmonary bypass surgery. This result is supported by findings that such hypoperfusion can be exacerbated by the induction of deep and prolonged hypothermia [2,3]. Another recent hypothesis claims that neuroapoptosis is the main cause of the neural damage that occurs [2].

Besides profound hypothermia, other risk factors found to be associated with post-cardiac surgery chorea include the induction of circulatory arrest during surgery, use of the alpha-stat method of pH management (an approach that keeps arterial blood at pH 7.40 and PaCO₂ 40 mmHg when corrected to 37°C during

hypothermia making the blood alkalotic and hypocapnic), presence of a cyanotic congenital heart defect, and patient age (most vulnerable period being 6–9 months to 5–6 years of age) [2,3].

In our case, during the bypass procedure we used the alpha-stat method; however, no significant change in pH level occurred. Profound hypothermia was not induced and there was no circulatory arrest. The child was not in the vulnerable age range and she had no cyanotic congenital heart defect.

CONCLUSIONS

We describe a rare clinical presentation of hemichorea after cardiac surgery with no significant risk factors. This case should prompt further investigations to clarify the mechanism underlying this phenomenon so that preventive measures can be taken.

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Capsule

Association of changes in effusion-synovitis with progression of cartilage damage over 18 months in patients with osteoarthritis and meniscal tear

Synovitis is a feature of knee osteoarthritis (OA) and meniscal tear and has been associated with articular cartilage damage. **MacFarlane** and colleagues examined the associations of baseline effusion-synovitis and changes in effusion-synovitis with changes in cartilage damage in a cohort with OA and meniscal tear. They analyzed data from the Meniscal Tear in Osteoarthritis Research (MeTeOR). The study was comprised of 221 participants. Over 18 months, effusion-synovitis was persistently minimal in 45.3% and persistently extensive in 21.3% of the patients. The remaining 33.5% of the patients had minimal synovitis on one occasion and extensive synovitis on the other. In adjusted analyses, patients with extensive effusion-synovitis at baseline had a relative risk (RR) of pro-

gression of cartilage damage depth of 1.7 (95% confidence interval [95%CI] 1.0–2.7). Compared to those with persistently minimal effusion-synovitis, those with persistently extensive effusion-synovitis had a significantly increased risk of progression of cartilage damage depth (RR 2.0, 95%CI 1.1–3.4). These findings indicate that the presence of extensive effusion-synovitis is associated with subsequent progression of cartilage damage over 18 months. The persistence of extensive effusion-synovitis over time is associated with the greatest risk of concurrent cartilage damage progression.

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 Eitan Israeli

Capsule

Obesity is independently associated with worse patient-reported outcomes in women with systemic lupus erythematosus

Patterson and colleagues investigated whether obesity in women with systemic lupus erythematosus (SLE) is independently associated with worse patient-reported outcomes (PROs). Data were derived from a prospective study of adult women with a diagnosis of SLE that was verified by medical record review. Two established definitions for obesity were used: fat mass index (FMI) ≥ 13 kg/m² and body mass index (BMI) ≥ 30 kg/m². Dependent variables included four validated PROs: disease activity as assessed by the Systemic Lupus Activity Questionnaire (SLAQ), depressive symptoms as assessed by the Center for Epidemiologic Studies Depression Scale (CES-D), pain as assessed by the Short Form 36 (SF-36) pain subscale, and fatigue as assessed by the SF-36 vitality subscale. The authors used multivariable linear regression to evaluate the associations of obesity with PROs, while controlling for potential confounders (age, race, education,

income, smoking, disease duration, disease damage, and prednisone use). The analysis included 148 participants, 32% of whom were obese. In the multivariate regression model, obesity was associated with worse scores for each PRO. Mean adjusted scores for the SLAQ and CES-D comparing obese versus non-obese participants were 14.8 versus 11.5 ($P = 0.01$) and 19.8 versus 13.1 ($P < 0.01$), respectively. The obese group also reported worse mean adjusted scores for pain (38.7 vs. 44.2; $P < 0.01$) and fatigue (39.6 vs. 45.2; $P = 0.01$). In a representative sample of women with SLE, obesity (as defined by both FMI and BMI) was independently associated with worse PROs, including disease activity, depressive symptoms, and symptoms of pain and fatigue. Obesity may represent a modifiable target for improving outcomes among obese women with SLE.

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 Eitan Israeli