Bilateral pulmonary emboli after bilateral mastectomy in a 15-year-old boy with hypogonadism: A case report

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Pulmonary emboli are rare, yet serious, complications of body contouring surgery. When they occur, they more often follow as complications of long, invasive procedures in adults. The present report details a case of bilateral pulmonary emboli in an obese 15-year-old boy with hypogonadism undergoing bilateral mastectomy for gynecomastia. The diagnosis of bilateral pulmonary emboli was made on the basis of clinical presentation and positive ventilation/perfusion scan. The patient responded well to heparin anticoagulation treatment. The relevance of pediatric obesity, pediatric body contouring surgery and the risk of thromboembolic events in pediatric patients are discussed.

Key Words: *Bilateral mastectomy; Gynecomastia; Hypogonadism; Obesity; Pulmonary emboli*

Une embolie pulmonaire bilatérale après une mastectomie bilatérale chez un garçon de 15 ans ayant un hypogonadisme : Rapport de cas

Les embolies pulmonaires sont des complications rares, mais graves, des chirurgies de la silhouette. Lorsqu'elles se produisent, elles constituent souvent des complications de longues interventions effractives effectuées chez des adultes. Le présent rapport détaille un cas d'embolie pulmonaire chez un garçon obèse de 15 ans ayant un hypogonadisme et qui a subi une mastectomie bilatérale en raison d'une gynécomastie. Le diagnostic d'embolie pulmonaire bilatérale a été posé d'après la présentation clinique et une scintigraphie pulmonaire de ventilation. Le patient a bien réagi au traitement anticoagulant à l'héparine. Les auteurs abordent la pertinence de l'obésité juvénile, la chirurgie de la silhouette en pédiatrie et le risque d'événements thromboemboliques chez des patients pédiatriques.

The prevalence of adolescent obesity is on the rise, as are its associated complications and comorbidities (1-3). In 2004, nearly one-third of Canadian adolescents between 12 and 17 years of age were reported as being either overweight or obese – a figure that has nearly doubled since 1978 (4). Since the recognition of this ongoing increase, the health effects related to obesity in children and adolescents have been widely examined. Early-onset type 2 diabetes, breathing difficulties, hypertension, hyperlipidemia and nonalcoholic fatty liver disease have all been well established as health concerns for which the obese pediatric patient is at risk (2,5,6).

It remains a question whether increased morbidity and mortality following surgical procedures is yet another health concern that is associated with adolescent obesity. In adults, obesity has been linked to increased perioperative and postoperative complications in a number of recent studies. High body mass index (BMI) has been associated with significant increases in complication rates following prostatectomy, kidney transplantation, colorectal surgery, hip arthroplasty and body contouring surgery, among others (7-14). Despite this accepted association in adults, the relationship between pediatric obesity and complications following surgical procedures is only recently being considered.

It is not uncommon for obese adolescent males to exhibit some degree of breast development. In recent literature (15), an incidence of up to 65% was reported for gynecomastia in adolescent boys between 14 and 15 years of age. Although a majority of these cases in adolescents are considered to be either physiological gynecomastia (which often resolves as puberty continues) or pseudogynecomastia secondary to obesity, some cases of adolescent gynecomastia, particularly those in male adolescents with endocrine disorders, persist well into adulthood (16). Currently, the standard of treatment for unresolving gynecomastia is surgery (17,18); bilateral mastectomy has been performed in adolescents as young as 13 years of age (19).

Although the risks of body contouring surgery have been well established in adults, there is little literature on the rates of these complications in adolescents (18,20-22). As rates of adolescent obesity continue to increase, understanding the risks of these complications in obese adolescents and how to best manage them, prevent them and identify pediatric patients at high risk for them is becoming of greater importance.

We report a case of bilateral pulmonary embolism in an obese 15-year-old boy with hypogonadism following bilateral mastectomy as treatment for gynecomastia.

CASE PRESENTATION

A 15-year-old boy with partial central hypogonadism and an incidental pituitary adenoma presented with a two-year history of unresolving Rohrich grade 1B gynecomastia. His BMI was 35 kg/m^2, and he had a history of severe childhood asthma, attention-deficit hyperactivity disorder, depression and smoking. His medications included testosterone injections (1 mg intramuscularly once a month), methylphenidate (40 mg once a day), risperidone (3 mg at night) and Ventolin (GlaxoSmithKline Canada) (when necessary). Previous treatment for his gynecomastia included monthly testosterone injections; however, after six months of treatment, no change in breast size was noted. The

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patient underwent bilateral subcutaneous mastectomies through a periareolar approach; surgical time was 30 min. No compression stockings, heparin or other prophylactic anticoagulation treatments were given. The operation proceeded without complication; the patient's heart rate remained stable between 70 beats/min and 100 beats/min throughout the procedure. The patient recovered from anesthesia, and when alert, showed no signs of respiratory distress and was communicating comfortably. It was noted that the patient's oxygen saturations were decreasing slightly; a fraction of inspired oxygen of 40% was required to maintain an oxygen saturation of 94%. As a result of his increasing oxygen requirements postoperatively, a ventilation/perfusion (V/Q) scan was obtained, and he was diagnosed as having a high probability for bilateral pulmonary emboli. He was started on intravenous heparin.

A coagulopathy screen was performed postoperatively to assess for hematological abnormalities; the international normalized ratio and the partial thromboplastin time were both in the normal range (1.1 and 27 s, respectively), and no other measured values suggested any hematological abnormalities. A chest x-ray performed the evening of the patient's surgery was normal. Bilateral Doppler ultrasound of his lower limbs was performed three days following his operation to assess for deep venous thromboses, which showed none. A cardiac assessment found no increase in right-sided pressures or right-sided strain pattern on electrocardiogram. The final diagnosis was bilateral pulmonary embolism without known embolic source.

DISCUSSION

Pulmonary emboli are rare, yet recognized, complications of body contouring surgery. In children and adolescents, the frequency of thromboembolic events, including pulmonary embolism, is much lower than in adults (23). A 15-year retrospective study by Bernstein et al (24) reported an incidence of 78 cases of thromboembolic complications per 100,000 hospitalized adolescents, compared with a reported incidence of between 119 and 900 cases per 100,000 hospitalized adults (25). As a consequence, pulmonary emboli are underdiagnosed in this population due, in large part, to their rarity and due to lack of clinical suspicion (23). Pulmonary embolism can be fatal if unrecognized, and thus the first step in management is rapid identification, followed by prompt anticoagulation treatment. Patients typically present with an acute decrease in the partial pressure of oxygen and symptoms of hemoptysis, dyspnea and chest pain, although asymptomatic and atypical presentations are quite common (24). A diagnosis of pulmonary embolism is made based on the patient's clinical pretest probability and on their results from further investigation: V/Q scan, D-dimer testing and chest x-ray may all help in diagnosis (25). Our patient presented with a decrease in the partial pressure of oxygen and a positive V/Q scan for pulmonary embolism, and was quickly diagnosed and treated appropriately, avoiding any devastating outcomes. The most common source of clinically relevant pulmonary embolism is deep venous thrombosis, arising most frequently from the iliac, popliteal or femoral veins, and thus a lower extremity venous ultrasound is commonly performed to check for deep venous thrombosis (25). In the case of our patient, whose ultrasound was negative for deep venous thrombosis, the source of his pulmonary emboli could not be identified. Other less common sources of deep venous thrombosis are the right heart and the renal and pelvic veins but, in children, it is common for the source to remain unknown (24). No reports are available on pulmonary emboli accompanying bilateral mastectomy in a pediatric patient.

As a specific complication of body contouring procedures, pulmonary emboli are often seen after long procedures such as abdominoplasty or liposuction (26-28). They are less commonly seen in mastectomy, yet are still recognized as known complications and have been reported (26). A retrospective study by Shermak et al (7) examining thromboembolic complications of body contouring surgery reported one case of pulmonary embolism in 188 patients. A BMI of greater than 35 kg/m^2 was the main risk factor for a thromboembolic event identified in patients undergoing body contouring procedures.

The frequency of body contouring procedures in the pediatric population is still relatively small and, thus, rates of pulmonary embolism as a complication of bilateral mastectomy in children are currently undocumented. What makes a patient susceptible to pulmonary embolism has been widely researched, but in children, it is still not well understood due to the rarity of this complication. Victoria et al (23) reported a higher incidence of recent surgery, blood dyscrasias and oral contraceptive use in pediatric patients presenting with pulmonary emboli. The general risk factors for thromboembolic events in adults, identified frequently in the literature as venostasis, hypercoagulability, vessel wall inflammation (Virchow's triad), recent surgery or trauma, and malignancy, can be applied to children as well. Other risk factors such as obesity, smoking, estrogen therapy, prolonged bed rest, inherited disorders of coagulation and previous pulmonary embolism are also well accepted in the literature as known risk factors for thromboembolic complications. In addition, hypogonadism, risperidone and testosterone injections have also been shown to correlate with the incidence of deep venous thrombosis and pulmonary embolism in a small number of studies (7,21,24,25,27,29-31). Our patient was subject to many of these risk factors. It is possible that his combination of risk factors contributed to his complication.

Most notable of our patient's risk factors was his morbid obesity. Shermak et al (7) reported that 100% of the thromboembolic events occurring as complications of various body contouring procedures occurred in patients with a BMI of more than 35 kg/m^2. A chart review by Hatef et al (27) examining thromboembolic risk assessment in body contouring surgery procedures also noted BMI greater than 30 kg/m^2 to be a significant risk factor for thromboembolic complications. In most recent literature, when stratifying patients based on their level of risk, many studies have adopted this correlation between BMI and thromboembolic complications and include BMI as a predisposing factor in their scoring (32). Although no studies to date highlight a similar correlation in the pediatric population, it can be speculated that our patient's morbid obesity contributed to his complication. Shermak et al (7) recommended that patients at higher risk for thromboembolic complications be treated with a high dose of heparin postoperatively, and continue on low-molecular weight heparin for two weeks following surgery.

In addition to obesity, our patient had a history of smoking – a habit currently considered to be an independent risk factor for thromboembolic events (29). Several studies have indicated a

connection between hypogonadism in men and thromboembolic tendencies. A study by Bennet et al (30) suggested a correlation among hypogonadism, defective fibrinolysis and deep venous thrombosis, and found that fibrinolytic activity in response to venous occlusion was abnormal in men with hypogonadism compared with controls. A separate set of studies (21,31) have also linked risperidone treatment to thromboembolic events. Cases of pulmonary emboli and thromboembolic events have been reported in patients treated with risperidone who possess no other risk factors (21,31). A study by Kamijo et al (31) proposed that the affinity of risperidone for the serotonin receptor may increase coagulability in these patients. Our patient was receiving risperidone treatment for his depression and disruptive behaviour up until the date of his surgery; thus, risperidone may also have played a role in his complication.

Recent recommendations from several studies support the treatment of patients at high risk for thromboembolism with prophylactic anticoagulation therapy. A large study by Miszkiewicz et al (26), which examined recommendations in the literature for thromboembolism prophylaxis in plastic surgery, found that a considerable majority (24 of 27 studies) were in favour of prophylactic procedures. A study by Seruya et al (32) suggested the use of both mechanical prophylaxis (graduated compression stockings) and low-molecular weight heparin for high-risk patients undergoing planned body contouring surgery to prevent deep vein thrombosis and pulmonary embolism. These strategies are in agreement with the prophylactic algorithm suggested by Abs (33) who agreed that, along with early mobilization, low-molecular weight heparin and compression stockings should be used. Surveillance investigation of the lower limbs for deep vein thrombosis in high-risk patients is also recommended to prevent progression to pulmonary embolism (33).

CONCLUSION

We encountered an unexpected complication of bilateral pulmonary emboli in an obese adolescent patient following bilateral mastectomy, which may have been due to a variety of factors that increased this patient's thrombotic tendency. The ongoing increase in pediatric obesity, when considered with both the high rate of mortality associated with pulmonary embolism and the multitude of risk factors that may present in an adolescent, emphasizes the need for further understanding of the risks of body contouring surgery in adolescents.

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