patient’s pulmonary mass remained non–small-cell lung cancer, small cell lung cancer, carcinoma, and pulmonary sarcoma.

This patient’s tumor was amenable to complete surgical resection because (1) the preoperative workup, including magnetic resonance imaging of the brain, computed tomography of the chest and abdomen, and positron emission tomography, did not reveal other sites of neoplastic disease and (2) the mass was extending into but not invading the left atrium as seen by cardiac magnetic resonance imaging. The decision was therefore made to proceed with surgical resection regardless of the tumor pathologic features to minimize the patient’s risk of sudden cardiac death from left ventricular inflow obstruction caused by tumor growth and systemic tumor embolization [1, 2]. Although this patient was asymptomatic, the majority of patients with extension of pulmonary metastasis into the left atrium present with or experience symptoms secondary to the intraatrial component of the tumor [6]. Symptoms include palpitation, stroke, syncope, and chest pain [6].

Given its rarity, there are no established adjuvant treatment protocols for extracranial metastatic meningiomas. Although there are reports of curative pulmonary metastasectomies for meningiomas, the long-term prognosis in this rare patient population is not well defined.

This is the first reported case of metastatic meningioma extending into the left atrium through the pulmonary vein. This case highlights the aggressive potential of meningiomas, which are typically considered benign tumors, and emphasizes the need to consider metastatic meningioma in the workup of pulmonary masses in patients with a history of meningiomas.

References


Intravenous Immunoglobulin-Induced Hemolytic Anemia After Thoracoscopic Thymectomy for Myasthenia Gravis

Hisashi Tsukada, MD, Rajitha Sunkara, MD, Dorcas Doja Chi, MD, Deirdre Keogh, NP, and Henning Gaisser et, MD

Department of Surgery, Division of General Thoracic Surgery, Department of Hematology and Oncology, St. Elizabeth’s Medical Center, Tufts University School of Medicine, and Harvard Medical School, Boston, Massachusetts

A 24-year-old woman underwent video-assisted thoracoscopic thymectomy for Osserman IIB myasthenia gravis (MG). In preparation for thymectomy, high-dose intravenous immunoglobulin (IVIG) was administered 1 week before the surgical procedure. After uneventful thoracoscopic thymectomy, the postoperative hemoglobin value decreased from 12.1 mg/dL to 8.2 mg/dL. A diagnosis of IVIG-associated hemolytic anemia was made based on a peripheral smear with numerous spherocytes, a positive direct antiglobulin test result, and increased reticulocyte count. Hemoglobin levels after IVIG administration should be monitored closely before and after elective surgical procedures to identify severe anemia. Transfusion of type-matched blood should be avoided and risk factors understood.

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Intravenous immunoglobulin (IVIG) is an “off-label” therapy for myasthenia gravis (MG) to reduce the risk of myasthenic crisis after thymectomy [1]; it is considered relatively safe, with mild and self-limited adverse effects that are less severe than those seen with plasma exchange. Common adverse events are fever and chills (13.8%), headaches (17.4%), nausea (6.9%), and allergic reaction (1.3%) [1].

The incidence of severe hemolysis after high-dose (> 2 g/kg) IVIG administration is reported at 1.6% [2], Stangel and associates [3] reported that 38 of 59 (64%) patients with neurologic autoimmune disease who received IVIG had statistically significant decreases in erythrocyte count, hematocrit values, and hemoglobin levels, although none was clinically relevant [3]. The true incidence of hemolytic anemia is unknown because mild presentations may be easily missed. Immunoglobulin products are prepared from blood plasma pooled from at least 3,000 to 10,000 donors, and hemolysis may occur from exposure to a broad range of antibodies against red blood cell antigen . We report here a case of clinically

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Address correspondence to Dr Gaisser, Department of Surgery, Division of General Thoracic Surgery, St. Elizabeth’s Medical Center, 736 Cambridge St, Boston MA 02135; e-mail: hgaisser@partners.org.
relevant but asymptomatic hemolytic anemia after IVIG administration before thymectomy.

A 24-year-old woman presented with a 3-year history of generalized weakness, including bilateral ptosis, episodic slurred speech, and difficulty swallowing. At diagnosis, test results determined that a serum acetylcholine receptor antibody was strongly positive. Electromyography showed a typical waning phenomenon. A Tensilon test 1 year before operation was strongly positive. Her symptoms were controlled with pyridostigmine bromide. Chest computed tomography showed an enlarged thymus consistent with thymic hyperplasia. She was referred for thymectomy to improve long-term control of symptoms. There was no surgical or medical history and no family history of autoimmune disease. Three weeks before operation, her red blood cell count was 4.25 M/µL, the hemoglobin level was 12.1 g/dL, and the hematocrit value was 37.6%. Her blood type was A positive. Intraoperative immunoglobulin was administered to a total dose of 160 g between days 7 and 4 before thymectomy. Using total intravenous anesthesia, thoracoscopic thymectomy proceeded with minimal blood loss. After immediate total intravenous anesthesia, thoracoscopic thymectomy proceeded with minimal blood loss. After immediate extubation, the hematocrit value was 23% and the hemoglobin level was 8.2 mg/dL on the first postoperative day. Vital signs remained normal; there was no chest drainage or radiographic evidence of hemothorax. The sclerae were anicteric. The reticulocyte was increased to 4.3%. The patient recalled dark urine after receiving IVIG but denied spontaneous bleeding. Further evaluation included a positive direct antiglobulin test result for IgG, a low-normal haptoglobin level, and moderate spherocytosis on peripheral blood smear (Fig 1); serum lactate dehydrogenase and bilirubin levels were normal. A diagnosis of IVIG-associated hemolytic anemia was made. To characterize the antibody coating, the patient’s red blood cells, the elution of postinfusion red blood cells, showed anti-A activity. The patient remained asymptomatic and was discharged on postoperative day 4. Her anemia resolved spontaneously with no intervention 2 weeks after the operation.

**Comment**

Thymectomy is considered for MG to improve long-term control of symptoms and reduce or eliminate the need for medication. Myasthenic crisis or acute respiratory insufficiency caused by deteriorating chest wall function is a potentially life-threatening complication after thymectomy for MG. Recent advanced perioperative anesthetic management for patients with MG allow these patients to be anesthetized safely without using neuromuscular agents. The rate of postoperative myasthenic crisis through transternal\(^4\), thoracoscopic\(^5\), robotic\(^6\), and transcervical\(^7\) approaches are reported to be 11%, 4.2%, 1.0%, and 0.7%, respectively. A less invasive approach may reduce the risk of crisis and may be considered to avoid administration of preoperative IVIG. In retrospective studies, the risk factors for postoperative myasthenic crisis are bulbar symptoms, history of myasthenic crisis, and serum levels of anti-acetylcholine receptor antibody greater than 100 nmol/L\(^4\). The optimal preparation in symptomatic MG before thymectomy to reduce the risk of crisis often consists of either plasma exchange or IVIG administration. The preferred management for preoperative stabilization of patients with MG is still controversial, because in evidence-based guidelines of the American Academy of Neurology, IVIG is recommended in the treatment of moderate or severe MG based on level B evidence\(^8\). Therefore, the decision between plasmapheresis, IVIG, and no preparation should be guided by the risk of any strategy. A prospective study of plasmapheresis in 1,727 procedures found complications in 36%\(^9\). The most common complications were fever (7.7%), urticaria (7.4%), and hypocalcemic symptoms (7.3%). IVIG-related complications receive less attention but may be equally severe; in addition to hemolytic anemia, these include anaphylactic reactions, pulmonary edema, acute renal injury, and aseptic meningitis. A comparative analysis of IVIG and plasma exchange for MG using the Nationwide Inpatient Sample database found no significant difference in risk-adjusted mortality or complications in 1,606 patients\(^10\). IVIG is favored because of the ease of administration and its lower cost.

Clinically significant hemolytic anemia after IVIG administration is sporadically reported. Kahwaji and colleagues\(^11\) reported a 5.8% incidence of high-dose IVIG-induced hemolytic anemia in renal transplantation, in which it is used for desensitization and treatment of antibody-mediated rejection. Hemolysis after IVIG infusion is almost exclusively observed in patients with a non-O blood group and results from passive acquisition of A or B isohemagglutinins in the infused IVIG product. Red blood cells coated with isohemagglutinin antibodies lead to either intravascular or extravascular hemolysis depending on the extent of complement fixation. Isosmolar liquid IVIG products with higher anti-A/B titer, especially anti-A titers greater than 1:16, and high-dose administration are a more likely cause of a clinically significant risk of hemolysis\(^2\). No

![Peripheral smear showing spherocytes (arrows) resulting from partial phagocytosis of antibody-bound red blood cells in the spleen.](image-url)
Dramatic Improvement After Bilateral Diaphragmatic Plication in Charcot-Marie-Tooth disease

Vivek Srivastava, MCh, MRCSEd, Tina Pasha, FRCA, Andrew Knowles, FRCA, Bernard Boothman, FRCP, Mohammed Paracha, FRCP, Maninder Kalkat, FRCS (CTh), and Joseph Zacharias, FRCS (CTh)

Departments of Cardiothoracic Surgery, Cardiothoracic Anesthesia, Neurology, and Respiratory Medicine, Victoria Hospital, Blackpool; and Department of Thoracic Surgery, Birmingham Heartlands Hospital, Birmingham, United Kingdom

A 52-year-old woman with Charcot-Marie-Tooth disease presented with severe dyspnea due to bilateral diaphragmatic paralysis severely compromising respiratory function. There was little in the available literature to guide us regarding management of this unusual condition, and after deliberation, we decided to treat her with a staged plication of bilateral hemidiaphragms. Postoperatively, she demonstrated very good symptomatic relief supported by objective evidence, including improvement in lung function tests. We describe our management of this difficult condition, including the surgical and anesthetic considerations, and would recommend bilateral diaphragmatic plication as an effective option in patients with this unfortunate disease.


Charcot-Marie-Tooth disease is named after the 3 clinics who first described it in 1886. It is also known as hereditary motor and sensory neuropathy. The management of this condition improved after 1991 when it became possible to use genetic markers for diagnosis. However, the multifactorial respiratory problems that occur with this disease are still very challenging to deal with. We describe bilateral diaphragmatic palsy in one such patient who responded very well to staged bilateral diaphragmatic plication.

A 52-year-old woman with Charcot-Marie-Tooth disease presented with worsening difficulty in breathing over 4 to 5 years. She was unable to lie backwards and used intermittent oxygen at home. She had a history of Charcot-Marie-Tooth disease diagnosed about 30 years earlier. She already suffered from muscular dystrophy in the lower limbs, with bilateral foot drop, leaving her severely disabled, and she needed walking aids for mobility. Other comorbidities included bronchial asthma, chronic back pain, and gastroesophageal reflux.

She was examined, and a chest roentgenogram revealed raised hemidiaphragms bilaterally (Fig 1).

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Address correspondence to Mr Srivastava, Department of Cardiothoracic Surgery, Victoria Hospital, FY3 8NR Blackpool, UK e-mail: srivivek500@hotmail.com.

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