

CASE REPORT

Guillain-Barré syndrome after aortic, tricuspid, and mitral valve surgery

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Abstract

Guillain-Barré syndrome incidence within 8 weeks of a surgical procedure appears to be more common than previously thought. GBS following open-heart surgery is exceedingly rare, perhaps underdiagnosed or underreported given surveillance data incidence. Clinicians should be keenly aware of this association and quickly consider a GBS diagnosis.

KEYWORDS

aortic, Guillain-Barré syndrome, mitral valve surgery, tricuspid

1 | INTRODUCTION

Guillain-Barré syndrome (GBS) is an acute autoimmune, inflammatory polyneuropathy of either demyelinating, axonal, or mixed phenotypes. GBS typically follows an upper respiratory infection or gastrointestinal infection with diarrhea; *Campylobacter jejuni* represents the best-known etiology. GBS has also been associated with other viral illnesses and vaccinations. Surgery associated with GBS may be more common than previously recognized. However, GBS associated with open-heart surgery is exceedingly rare. We describe a rare case of GBS occurring after aortic, tricuspid, and mitral valve surgery and review the world's literature.

Guillain-Barré syndrome is a classic example of a neuropathy secondary to disordered immunity. Aberrant B-cell response to glycolipids and related conjugates results in demyelinating, axonal, or mixed nerve damage that manifests as an acute inflammatory immune-mediated polyneuropathy.¹ Clinical manifestations include tingling, progressive weakness, pain, and diminished reflexes. Although classic GBS is that of a demyelinating neuropathy with progressive ascending weakness, several clinical variants exist including Miller Fisher syndrome, characterized by ophthalmoplegia and ataxia.²

Two-thirds of GBS cases are preceded by an upper respiratory tract infection or diarrhea, with *Campylobacter jejuni*

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the most common etiology.² Vaccinations and various infectious vectors have also been associated with GBS, including Epstein-Barr virus, cytomegalovirus, varicella-zoster virus, mycoplasma, and most recently, Zika virus.^{2,3}

Guillain-Barré syndrome incidence following surgical procedures is unknown. Previous national surveillance suggests that 5% of GBS patients had undergone surgery within an 8-week interval before onset.⁴ However, 45% of those patients reported an antecedent illness within that same time period. Most recently, two retrospective case series of patients with GBS found a 9.5 and 15 percent incidence of post-surgical GBS.^{5,6} Thus, it appears that the incidence of post-surgical GBS may be higher than previously reported. Taken together, the most common surgical procedures in these case series were gastrointestinal and orthopedic; rarely patients develop GBS following coronary artery bypass surgery. We present a rare case of GBS associated with open-heart surgery and review the extant world's literature.

2 | CASE

A 53-year-old African-American man presented to the emergency room with acute dyspnea, anxiety, muscle spasms, and severe pain in his mid-thoracic spine. He also reported paresthesia of his arms and legs. Past medical history was notable for recent open-heart surgery with aortic valve replacement, mitral and tricuspid valve repair, and left atrial appendage exclusion 12 days prior to admission. Cardiovascular history was significant for a bicuspid aortic valve with resultant severe aortic insufficiency, severe mitral regurgitation, and mild tricuspid regurgitation with severe pulmonary hypertension and a markedly dilated tricuspid annulus. He had no significant coronary atherosclerosis. Additional medical history included end-stage renal disease on hemodialysis, hypertension, chronic tophaceous gout, and hyperlipidemia. Physical examination revealed BP 176/94 mmHg, pulse 80 beats/min, temperature 97.9°F, respiratory rate 20, and BMI 23.89 kg/m².

Cardiovascular examination revealed a healing sternotomy scar without evidence of infection and artificial aortic valve closure on cardiac auscultation. Cardiac echocardiography showed preserved ejection fraction with a well-functioning bioprosthetic aortic valve. There was no aortic insufficiency, no mitral regurgitation, and trace to mild tricuspid regurgitation. Mean gradient for the aortic prosthesis was 10, mean mitral gradient was 2, and mean tricuspid gradient was 1. There was no pericardial effusion.

Breath sounds were diminished in the left lung base. He had preserved motor strength in the upper and lower extremities. Initial CXR revealed an elevated left hemidiaphragm despite no intraoperative topical hypothermia or internal mammary harvest. Initial laboratory studies demonstrated sodium

133 mmol/L, potassium 4.4 mmol/L, bicarb 24 mmol/L, chloride 94 mmol/L, BUN 28 mg/dl, Cr 5.9 mg/dl, and blood glucose 93 mg/dl. Complete blood count showed WBC 11.6 with neutrophilia and lymphopenia, Hb 7.0, Hct 20.8, and platelet count 317,000.

On hospital day two he subsequently developed progressive weakness and numbness of both lower extremities. Physical examination revealed symmetric bilateral lower extremity weakness, areflexia, and a sensory level around T 10. Computed tomography scan of cervical, thoracic, and lumbar spine demonstrated degenerative changes and osteophytes with no evidence of compression fracture, hematoma, abscess, or transverse myelitis. Lumbar puncture revealed normal opening pressure with clear fluid, protein 315 mg/dl, WBC 5 cells/mm³, RBC 0 cells/mm³, and glucose 62 mg/dl. Cerebrospinal fluid albumin was 133 mg/dl (0–35), and oligoclonal bands were negative. Acetylcholine receptor antibodies were 0.0 nmol/L, and a nasopharyngeal swab for respiratory viruses was negative.

Electrodiagnostic studies showed diffuse sensory-motor peripheral neuropathy with axonal degeneration and demyelinating changes consistent with acute inflammatory demyelinating polyneuropathy, the hallmark for Guillain-Barré Syndrome. Neurology was consulted and recommended transfer to ICU and initiation of IVIG at 0.4 mg/kg. Stat IgA level was normal 228 mg/dl (70–400). On hospital day six the patient's respiratory status decompensated requiring intubation and mechanical ventilation. Plasmapheresis was initiated, and IVIG was discontinued. The patient's hospital stay was complicated by pneumonia, but he was extubated after 7 days. Motor strength gradually improved, he was subsequently transferred to acute rehab, and he was discharged home and ultimately regained full motor strength. Several months later the patient was able to receive a cadaveric heterotopic renal transplant without complication. His Hughes functional grade score, previously 5 indicating severe GBS, was now 0, indicating good prognosis.

3 | DISCUSSION

In 1987, Renlund et al⁷ reported the first case of CABG-associated GBS in a 65-year-old man who developed symptoms 3 days after surgery, successfully treated with plasmapheresis. Since then, nine additional cases have been reported including the current case (Table 1).^{8–13} While there are other extant reports of GBS following cardiothoracic surgery, the temporal precedence between surgery and GBS of 12 and 48 months seems an unlikely etiology.^{14,15} In nine of the ten cases, patients were male, and all patients developed symptoms within 15 days after surgery. All patients were successfully treated with either IVIG, plasmapheresis or plasma exchange and demonstrated either significant

TABLE 1 Reported GBS following cardiothoracic surgery

Author	Year	Age	Sex	Surgical procedure	Onset of symptoms	Treatment	Outcome
Renlud ⁷	1987	65	Male	On pump CABG	Day 3	Plasmapheresis/mechanical ventilation	Rapid improvement
Hogan ⁸	1992	60	Male	Aortic and mitral valve replacement	Day 15	Plasmapheresis	Rapid improvement
Campbell ⁹	2008	53	Male	CABG	Day 14	Plasma exchange/mechanical ventilation	Discharged 1 month, fully mobile
					Day 1	Intravenous immunoglobulin for 5 days/mechanical ventilation and tracheostomy	Discharged after 23 days independently mobile
Algahtani ¹⁰	2009	71	Female	Two vessel CABG	Day 4	Plasmapheresis/mechanical ventilation	Prolonged ICU stay, discharged to rehab facility
Punith ¹¹	2011	65	Male	Emergency aortic valve replacement	Day 11	Intravenous immunoglobulin	Moderate improvement, transferred to rehab facility
					Day 12	Intravenous immunoglobulin	Full neurologic recovery within 12 weeks
					Day 2	Plasmapheresis	Discharged without sequela on the 10th post operative day
Cingoz ¹²	2012	67	Male	Off pump CABG	Day 2	Plasmapheresis/mechanical ventilation	Discharged day 28 stable
Raut ¹³	2018	32	Male	Hypothermic bypass for ruptured Valsalva sinus and aortic valve	Day 2	Plasmapheresis/mechanical ventilation	Discharged day 28 stable
Aldamouk, present case	2018	53	Male	Aortic valve mitral and replacement, tricuspid repair	Day 12	Intravenous immunoglobulin for 2 days followed by plasmapheresis	ICU mechanical ventilation and complete recovery within 6 weeks

improvement or complete recovery. Six patients required mechanical ventilation, as in the current case. Average age of the patients was 61.4 years; average time between the surgical procedure and symptom onset was 7.6 days (range 1–15, median 7.5 days). Only four patients in the series underwent valvular repair, one of which was replacement, as is our case. Table data suggest either increasing incidence or reporting of cardiothoracic surgery associated GBS.

National surveillance data from the Centers for Disease Control and Prevention have documented a 5% incidence of GBS within 8 weeks post-surgery.⁴ However, 45% of those patients reported an antecedent illness within that same time period. There was a direct correlation between increasing age and the incidence of GBS, as well as a male preponderance. Our patient denied any preceding upper respiratory or gastrointestinal symptoms.

In a series published by Gensicke et al.⁵ the risk of developing GBS during 6 weeks following surgery was 13.1 times higher than the risk in the general population. None of the patients in their study had prior open-heart surgery. In a very recent retrospective review of 208 cases of GBS, Nagarajan et al⁶ reported that 15% of patients developed post-surgical GBS within 8 weeks of surgery. Median duration from the surgical procedure to the onset of first GBS symptoms was 19 days. Interestingly, 61% of patients had a known diagnosis of malignancy and 29% had an underlying autoimmune condition. Multivariate analysis demonstrated a statistically significant association of post-surgical GBS with age, malignancy, and the presence of an autoimmune disorder.⁶ In Nagarajan's series only one patient underwent CABG.

The mechanism and pathogenesis of GBS after cardiac surgery are unknown. Surgery may cause exposure of nerve roots leading to oncoantigen-mediated misdirection of autoimmune responses to epitopes within the peripheral nervous system. Immune dysregulation may be secondary to lipid-soluble anesthetic agents.⁶ Additionally, cardiopulmonary bypass has been associated with activation of complement, secretion of both pro- and anti-inflammatory cytokines (IL-8, IL-10), tumor necrosis factor (TNF- α), and activation of neutrophils.¹⁶

Guillain-Barré syndrome incidence within 8 weeks of a surgical procedure appears to be more common than previously thought. GBS following open-heart surgery is exceedingly rare, perhaps underdiagnosed or underreported given surveillance data incidence. Clinicians should be keenly aware of this association and quickly consider the diagnosis in any patient who develops progressive weakness, pain, and diminished reflexes post-operatively.

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CONFLICT OF INTEREST

None declared.

AUTHOR CONTRIBUTIONS

Authors verify that this manuscript is not under review by other journals. All authors, AA, HS, LWH, and TM, have contributed to the writing, editing, and preparation of this manuscript and have reviewed it prior to submission.

ETHICAL APPROVAL

Published with the written consent of the patient. The IRB has determined that case reports are exempt from oversight.

DATA AVAILABILITY STATEMENT

Data sharing not applicable to this article as no datasets were generated or analyzed during the current study.

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