CASE REPORT

Case Report: Subacute onset of the motor-sensory axonal neuropathy variant of Gullain-Barré syndrome after epidural anesthesia [version 1; referees: 2 approved with reservations]

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Abstract

Guillain-Barré syndrome (GBS) is an acute ascending peripheral neuropathy, caused by autoimmune damage of the peripheral nerves. GBS can be divided into three subtypes: acute inflammatory demyelinating neuropathy, acute motor axonal neuropathy, and the more rare type, acute motor and sensory axonal neuropathy (AMSAN). Reports of AMSAN with onset after epidural anesthesia and spinal surgery are extremely rare, and the linkage between development of GBS and neuroaxial anesthesia remains conclusively unconfirmed. We present a case in which the patient developed subacute motor and predominantly sensory neuropathy following epidural blockade. The case emphasizes the need of including AMSAN in differential diagnostic considerations to changes in motor and sensory function following epidural anesthesia, allowing accelerated rehabilitation and relevant alleviating therapy.

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Background

Guillain-Barré syndrome (GBS) is an acute peripheral neuropathy caused by an autoimmune response against the myelin sheets of peripheral nerves and can be subdivided into motor and mixed motor and sensory types\(^1\). Acute onset of GBS after epidural anesthesia has previously been reported, raising concern of a correlation between GBS and use of neuroaxial anesthesia\(^2\).

In this case report a patient is presented with subacute onset of motor and predominantly sensory polyneuropathy following epidural anesthesia.

Case

A 65-year-old woman without previous neurological disease was admitted to the surgical department due to pain in the sacral bone and one and a half week lasting water-thin stool per rectum and brownish stool per vaginam. The patient had various comorbidities, including previous left-sided nephrectomy, hysterectomy, and cystectomy with an ileum bladder due to stage T4 vesical cancer. The patient had no history of recurrence.

Computed tomography (CT) and magnetic resonance imaging (MRI) confirmed sigmoid diverticulosis with diverticular stenosis and fistulae between colon-rectum and upper vagina. A Hartmann’s procedure was performed with combined general and epidural anesthesia and the epidural infusion was discontinued after 2 days. One day after discontinuation, the patient developed dysesthesia and numbness in both hands and lower extremities up to knee level, accompanied by substantial decrease of fine motor skills. Symptoms were initially interpreted as unintended neural compression during surgery.

Neurological examination 10 days after onset of symptoms showed unaffected cerebral function with preserved function of all cranial nerves. Both hands presented with symmetrical, bilateral atrophy. Motor testing showed grade 4+ muscle strength on a 1–5 muscle strength scale in all extremities, with all deep tendon reflexes preserved, except for the Achilles bilaterally. Sensory testing confirmed dysesthesia ascending up to wrist- and knee level bilaterally with reduced sense of touch, vibration, pain, and thermal changes. Proprioception remained intact but the patient was observed with ataxic walk. The condition was determined as a motor and predominantly sensory polyneuropathy with subacute onset and unknown etiology. A lumbar puncture found inconspicuous cerebrospinal fluid with normal protein level and absent pleocytosis. Electromyography confirmed a sensorimotor polyneuropathy in upper and lower extremities of predominantly axonal type. Cylindrical root compression and multiple segments presenting anterolysis. However, the specialized unit for columnar diseases found neither any plausible cause for the current symptoms, nor indication for decompressive surgery. Positron emission tomography–computed tomography disproved plausible neoplastic cause for the neuropathy. Paraclinically, the patient had a normal biochemical profile except for an increased erythrocyte sedimentation rate and an increased level of light-chain immunoglobulins, subsequently discarded as possible light-chain amyloidosis by negative Congo-red coloration.

The patient was only treated with gabapentin, parenteral nutrition and substitution of vitamins B, D and magnesium during the next ten months after onset of neurological symptoms. Parental nutrition was given for one week shortly after onset of neurological symptoms. Vitamin B was substituted through intramuscular injection of 1 milligram (mg) every third month, vitamin D by two tablets of 25 mg daily and magnesium by one tablet of 360 mg daily. Pain relief was provided by an initial dosage of 300 mg gabapentin daily, and increased intermittently during the following one and a half year to 900 mg three times daily.

The patient was followed every third month at the Neurology clinic for the next one and a half year. Her condition remained unchanged with stationary paresthesia in upper and lower extremities, considerably impaired activity-level, and complicated by neuropathic pain.

Discussion

This case report followed the CARE Guidelines for reporting of case reports\(^5\) and details of surgery, hospitalization and physical examinations were retrieved from the official national registration system used at all Danish hospitals, with consent from the patient.

GBS can be divided into three subtypes: acute inflammatory demyelinating neuropathy, acute motor axonal neuropathy, and the more rarely seen acute motor and sensory axonal neuropathy (AMSAN)\(^6\). The majority of cases with GBS are caused by prior infection of the upper respiratory tract or gastroenteritis and a subsequent abnormal immune response to the infectious agent, typically with onset of symptoms 2–4 weeks after the initial infection\(^7\).

Our case presented a subacute motor and predominantly sensory affection, most consistent with the AMSAN subtype. This was supported by the consistent findings at the neurological examination and electromyography. Differential diagnoses were inflammatory neuropathy or neuropathy due to mineral- or tracer deficiency caused by diarrhea or previous small intestine resection. The patient was substituted with vitamins B, D and magnesium without remission of the current complaints. Cerebrospinal fluid and biochemical parameters showed no signs of cerebral infection and the patient did not present with fever. Although diarrhea was present, this debuted prior to onset of neurological symptoms and whether the diarrhea was on an infectious background was undetermined, but it could most likely be explained by her complicated diverticular disease. Furthermore, diagnostic imaging revealed no relevant anatomical pathology causing the neurological symptoms. Thus, the patient presentation and clinical course were highly suggestive of AMSAN.
Prior cases of acute onset of AMSAN after spinal surgery or epidural anesthesia have been reported\(^4\);\(^5\);\(^6\). The suggested underlying mechanism is probably an intraoperative release of antigens, leading to autoimmunization and a subsequent immune response with attack of neural elements\(^4\);\(^6\). Other theories have suggested how surgery may cause either an immune imbalance with increased number of T-cells, or a transient immunosuppression promoting subclinical infections\(^6\). The AMSAN variant may result in fulminant weakness and sensory loss within 7 days, and recovery is poor\(^8\);\(^9\) in contrast to the typical GBS presentation with motor impairment that typically will recur fully within weeks to months\(^2\). No curative interventions are currently known, but this case emphasizes the importance of aggressively investigating any new onset of motor and sensory loss postoperatively since it may be the AMSAN variant of GBS. This could possibly aid to accelerate correct diagnosis and forward rehabilitation and relevant alleviating therapy with considerable effect on the patients’ quality of life.

This case report presents a case of subacute motor and predominantly sensory loss after epidural anesthesia, highly suggestive of AMSAN. Although rarely reported, AMSAN should be included in the differential diagnostic considerations to changes in motor or sensory function after epidural or spinal anesthesia.

**Consent**

Written informed consent was obtained from the patient for publication of this case report and any accompanying images and/or other details that could potentially reveal the patient’s identity.

**Author contributions**

M.S.: study design, drafting, and final approval; J.R: study design, revision, and final approval; J.B.: study design, revision, and final approval.

**Competing interests**

No competing interests were disclosed.

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### References

Open Peer Review

Current Referee Status: ? ?

Version 1

Gerard Said
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Interesting case of an "axonal GBS" syndrome occurring shortly after epidural anesthesia, in a patient with a number of health problems. I assume that the patient could also have developed a nutritional axonal polyneuropathy due to her poor health condition.

The authors claim that light chain amyloidosis was excluded after negative Congo red staining. Which tissue was studied?

Amyloid neuropathy unlikely because of the rate of onset, and stability of deficit afterwards.

I have read this submission. I believe that I have an appropriate level of expertise to confirm that it is of an acceptable scientific standard, however I have significant reservations, as outlined above.

Competing Interests: No competing interests were disclosed.

Jean-Michel Vallat
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This is an interesting case, but I am not sure that at the present time, we have enough data; if the authors can let us know the followings, it will be all right:

- Why Campylobacter jejuni or any other infection was not looked for, as the diarrhea was present prior to the onset of the neurological symptoms?
- Were anti-glycolipid antibodies tested? In a significant number of acute dysimmune neuropathies, they are considered as useful biomarkers if they are present.
- If AMSAN was discussed, why did the patient not receive at the beginning, any treatment such as immunoglobulins or plasma exchanges?

I am surprised that after 10 days of evolution, when the motor testing was only 4+, a bilateral atrophy of hands was observed.
Actually, the first AMSAN cases have been reported by Feasby et al. (1986).

**References**

I have read this submission. I believe that I have an appropriate level of expertise to confirm that it is of an acceptable scientific standard, however I have significant reservations, as outlined above.

*Competing Interests:* No competing interests were disclosed.