Man-in-the-barrel syndrome after thoracoilium fusion

Case report

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The authors report a case of man-in-the-barrel (MIB) syndrome occurring after an extensive revision involving thoracoilium instrumentation and fusion for iatrogenic and degenerative scoliosis, progressive kyphosis, and sagittal imbalance. Isolated brachial diplegia is a rare neurological finding often attributed to cerebral ischemia. It has not been previously reported in patients undergoing complex spine surgery. This 70-year-old woman, who had previously undergone T11–S1 fusion for lumbar stenosis and scoliosis, presented with increased difficulty walking and with back pain. She had junctional kyphosis and L5–S1 pseudarthrosis and required revision fusion extending from T-3 to the ilium. In the early postoperative period, she experienced a 30-minute episode of substantial hypotension. She developed delirium and isolated brachial diplegia, consistent with MIB syndrome. Multiple studies were performed to assess the origin of this brachial diplegia. There was no definitive radiological evidence of any causative lesion. After a few days, her cognitive function returned to normal and she regained the ability to move her arms. After several weeks of rehabilitation, she recovered completely. Man-in-the-barrel syndrome is a rare neurological entity. It can result from various mechanisms but most commonly seems to be related to ischemia and is potentially reversible. (DOI: 10.3171/SPI.2008.8.08485)

KEY WORDS • complex spine surgery • cruciate paralysis • man-in-the-barrel syndrome

Isolated brachial diplegia was first described in 1917 by Dide.8 In 1969, Mohr13 coined the term MIB syndrome to describe the clinical picture of brachial diplegia following systemic hypotension resulting in watershed infarction of the anterior and middle cerebral artery territories.

Man-in-the-barrel syndrome is a rare entity in the cranial literature and is even more rarely reported in the context of the spine. We present the case of a 70-year-old woman who had previously undergone T11–S1 fusion for lumbar stenosis and scoliosis, presented with increased difficulty walking and with back pain. She had junctional kyphosis and L5–S1 pseudarthrosis and required revision fusion extending from T-3 to the ilium. In the early postoperative period, she experienced a 30-minute episode of substantial hypotension. She developed delirium and isolated brachial diplegia, consistent with MIB syndrome. Multiple studies were performed to assess the origin of this brachial diplegia. There was no definitive radiological evidence of any causative lesion. After a few days, her cognitive function returned to normal and she regained the ability to move her arms. After several weeks of rehabilitation, she recovered completely. Man-in-the-barrel syndrome is a rare neurological entity. It can result from various mechanisms but most commonly seems to be related to ischemia and is potentially reversible.

Operation. Our revision included removal of prior instrumentation with insertion screws from T-3 to the ilium, an L-2 pedicle subtraction osteotomy, and revision L-1, L-2, and L-3 laminectomies (Fig. 1). The duration of the operation was 510 minutes and the estimated blood loss was 2500 ml. The patient received 8 U of packed red

Abbreviation used in this paper: MIB = man-in-the-barrel.
blood cells, 3 U of fresh-frozen plasma, and 1500 ml of cell saver blood during the procedure. The procedure itself was uncomplicated except for a 2-hour intraoperative period in which the patient experienced mild hypotension (mean arterial pressure 50–60 mm Hg) that was treated intermittently with a phenylephrine drip at 0.1–0.2 mcg/kg/min.

Postoperative Course. On the evening following surgery, she was intubated but briskly followed commands with normal strength in all four limbs. Six hours after the completion of the surgery, she suffered a 30-minute episode of substantial hypotension (mean arterial pressure 35–50 mm Hg) treated with fluid resuscitation and a phenylephrine drip at 0.1–0.4 µg/kg/min. Her hemoglobin was 9.9 gm/dl (hematocrit 29.6%). Almost immediately following this episode, the patient became less responsive and could no longer move any of the muscle groups of her upper extremities (manual motor test Grade 0/5) in response to either command or painful stimuli while she was actively moving her lower extremities (Grade 5/5). Responsiveness to painful stimuli appeared to be fully preserved in the upper extremities.

The patient underwent brain and cervical spine MR imaging, which included STIR, diffusion weighted, FLAIR, and contrast-enhanced images. These studies revealed a small area of acute cerebellar infarction, but the findings did not explain the patient’s symptoms. There was hypertrophic degeneration of the right olivary nucleus with T2 signal hyperintensity. We observed diffuse cerebral volume loss with scattered white matter disease that was unchanged compared with MR imaging findings seen at evaluation of the transient ischemic attack 1 year earlier (Fig. 2). Computed tomography and MR images of the cervical spine revealed negative findings. An electroencephalogram demonstrated generalized cortical slowing. Because the patient developed transient hypotension, cardiology and vascular surgery consultations were performed. The results of electrocardiography and cardiac monitoring were normal except for a transient episode of atrial fibrillation. An echocardiogram showed no evidence of heart failure or ventricular dysfunction.

On the 4th postoperative day, the patient became much more responsive and was able to actively follow commands with her lower extremities. She was extubated and exhibited normal sensory function in all areas but had persistent Grade 0/5 strength in all groups of both upper extremities. Over the next 24 hours she gradually regained nearly all upper-extremity function (Grade 4+/5). She regained normal cognitive and neurological function within 10 days of surgery. Six months postoperatively, the results of neurological examination were normal, her pain was limited (visual analog scale score of 3/10), and her function was good (Oswestry Disability Index score of 28).

Discussion

Since the time of Dr. Mohr’s description of MIB syndrome to characterize brachial diplegia,13 several clinicians have reported similar findings in their cases. An MIB syndrome has been associated with many other clinical phenomena. These cases include brain metastasis,14 central pontine lesions,7,16 hemorrhagic shock,5 hypotension after cardiac surgery,10,11 traumatic head injury,6 cardiorespiratory arrest,18 and glioblastoma multiforme.4 Some authors have attributed MIB syndrome to cervical spine disease such as anterior cervical artery infarction,3
cervical metastasis,\textsuperscript{20} and motor neuron disease.\textsuperscript{12} Hence, brachial diplegia can result from either supra- or infratentorial disease. Supratentorial pathology is usually associated with hypotension that may result in either ischemia or infarction of the watershed zone between the anterior and middle cerebral arteries. This watershed zone corresponds to the motor and sensory cortex of the bilateral upper extremities in the temporoparietal region adjacent to the posterior operculum.\textsuperscript{2}

For infratentorial disease, Bell,\textsuperscript{1} in 1970, created the term “cruciate paralysis” to describe brachial diplegia resulting from injury of the upper portion of the pyramidal decussation. The topographic and anatomical attributes of the corticospinal tracts demonstrate how upper-extremity weakness can occur in isolation from lower-extremity weakness with lesions of the craniocervical junction. The corticospinal tracts of the upper extremities descend more medially and anteriorly relative to the lower-extremity corticospinal tracts. Furthermore, the pyramidal decussation of the corticospinal tract of the upper extremity is located more rostrally relative to the lower extremities. Bell described 3 clinical scenarios in which midline upper pyramidal decussation fibers could have been damaged: 1) an odontoid fracture, 2) a cervical hyperextension injury in a patient with basilar impression, and 3) acute hydrocephalus following suboccipital craniectomy for excision of a posterior fossa meningioma. As a result it may be more appropriate to use the term MIB syndrome for supratentorial lesions and cruciate paralysis for infratentorial lesions, as suggested by Geordiadis and Schulte-Mattler.\textsuperscript{10}

The pathophysiology of MIB syndrome, according to some authors, is associated with an acute cerebral event secondary to brain ischemia.\textsuperscript{9,19} During surgery, arterial hypotension (with systolic pressure < 50 mm Hg for > 5 minutes) has been thought to be the ischemic event that caused MIB syndrome in certain cases.\textsuperscript{17} Hurley and Wood\textsuperscript{11} have suggested that the rapidity of the fall in blood pressure was responsible for brain damage. The duration and intensity of hypotension necessary to cause the syndrome are not known, but most probably are influenced by other factors such as carotid artery stenosis and history of hypertension. The utility of steroid agents in this setting has not been documented.

Since our patient had normal cervical MR and CT imaging findings, we excluded cervical and pyramidal decussation lesions. The MR images of the brain did not elucidate the cause of the patient’s condition either. Sometimes, frontoparietal ischemic lesions can be detected using MR imaging in patients with MIB syndrome after arterial hypotension\textsuperscript{9} and can exclude focal lesions.\textsuperscript{4,14} Although we did not find any causative lesion on imaging, we attributed our patient’s symptoms to intra- and postoperative hypotension.

The results of neurological examination immediately after surgery in our patient were normal, and the symptoms started to develop after completion of the procedure. Clerget et al.\textsuperscript{5} have described similar timing in the onset of symptoms. The outcome of MIB syndrome is generally good when it occurs in noncomatose patients or when short-duration ischemia is effectively reversed. Our present patient had a near-complete recovery in 10 days, and a complete recovery was observed after several weeks. Cases of survival with total recovery\textsuperscript{2,11,17,19} or only minor deficiency\textsuperscript{15} suggest that the prognosis depends on the severity and the duration of the systemic hypoperfusion and the effectiveness of the treatment for the low blood flow.

**Conclusions**

Man-in-the-barrel syndrome is a rare neurological entity. It can result from various mechanisms but most
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commonly seems to be related to ischemia and is potentially reversible. Brachial diplegia due to cervical or pyramidal lesions may be better described as cruciate paralysis. Man-in-the-barrel syndrome can be a potential complication of spine surgery and it is important for spine surgeons to be aware of its presence and pathophysiology.

Disclaimer

The authors report no conflict of interest concerning the materials or methods used in this study or the findings specified in this paper.

References


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