Middle meningeal artery embolization for chronic subdural hematoma in a young patient with refractory thrombocytopenia secondary to leukemia: more evidence for a paradigm shift? Illustrative case

Emalee J. Burrows, MBBS,1 Seng Chye Lee, MBBS,1 Omar K. Bangash, MBBS,1 Timothy J. Phillips, MBBS, CCINR,1,2 and Sharon Lee, MBBS1

1Neurosurgical Service of Western Australia, Sir Charles Gairdner Hospital, Perth, Western Australia, Australia; and 2Neurological Intervention and Imaging Service of Western Australia, Perth, Western Australia, Australia

BACKGROUND Chronic subdural hematoma (CSDH) is one of the most common neurosurgical presentations, with an increasing number now also presenting with concurrent thrombocytopenia. Although middle meningeal artery (MMA) embolization has been considered in elderly patients with high comorbidities, it may permit treatment of CSDH in patients who are at high risk for recurrence or deemed unsuitable for surgical management due to thrombocytopenia.

OBSERVATIONS A 35-year-old man who had severe thrombocytopenia due to blast cell crisis with chronic myeloid leukemia developed an atraumatic CSDH. The patient developed severe headaches in the hospital while being treated for febrile neutropenia. He remained neurologically intact. MMA embolization was undertaken due to the morbidity and mortality risks associated with surgery and the high risk of recurrence due to severe thrombocytopenia. At 2 months post-procedure the patient was asymptomatic and there was almost complete resolution of the hematoma.

LESSONS Thrombocytopenia in the presence of a CSDH is becoming increasingly common. This case highlights the particular role of MMA embolization in patients with severe thrombocytopenia where surgery carries high morbidity and increased mortality.

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KEYWORDS middle meningeal artery embolization; chronic subdural hematoma; thrombocytopenia; endovascular embolization

Chronic subdural hematoma (CSDH) is a relatively common neurosurgical condition that mainly affects elderly individuals aged 65 years or older. Studies around the world have found an incidence rate of 1.72 per 100,000 persons to 20.6 per 100,000 persons per year, with most of these in persons aged 65 years or older. It is rare to have a young person with CSDH.1

Middle meningeal artery (MMA) embolization as a treatment for CSDH has been gaining interest as an adjunct and possibly an alternative to surgical evacuation in select patients. Current interest largely centers around use in elderly patients with significant medical comorbidities.2,3 This includes those who are taking anticoagulant and antplatelet medications who are deemed to have a higher risk of recurrence as well as operative morbidity and mortality.2,3 In particular, patients who have recurrent CSDH after surgical evacuation constitute a subgroup that may benefit from MMA embolization.3

Being minimally invasive in nature, it has been shown to be safe even with patients on anticoagulation medication4 and it may be able to help clinicians treat patients who have CSDH but who are not good candidates for surgery due to inherent coagulopathies or who are on anticoagulation medication that is risky to cease because of the patient’s underlying condition; however, more research is still required to further evaluate its safety and efficacy.

This article aims to discuss a case of the use of MMA embolization in a severely thrombocytopenic patient and review the literature.

ABBREVIATIONS CSDH = chronic subdural hematoma; CT = computed tomography; GCS = Glasgow Coma Scale; MMA = middle meningeal artery.

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Illustrative Case

Presentation and Admission

A 35-year-old man with febrile neutropenia postchemotherapy (fludarabine, cytarabine, idarubicin, and granulocyte colony-stimulating factor [FLAG-I1da]) presented with a background of chronic myeloid leukemia (with blast cell crisis) diagnosed 3 months earlier and who developed severe thrombocytopenia secondary to chemotherapy. He was previously active, fit, and well with no other significant past medical or family history.

On day 3 of his admission, he described the onset of mild headaches. His platelet count was noted to be 18 $\times 10^9/\mu L$ (normal: $150–400 \times 10^9/\mu L$). Over the next 2 days, his headache persisted, and he reported photophobia, although he remained neurologically intact. On review, he reported a retro-orbital as well as temporal pulsating character to his headache, with his platelet count dropping to $14 \times 10^9/\mu L$.

Investigations

The next day he had severe worsening of his headache, and an urgent brain computed tomography (CT) scan was arranged. His scan revealed a left convexity CSDH with significant local mass effect and 11 mm of midline shift (Fig. 1 left). The patient did not recall any history of trauma or head injury in the preceding months. His Glasgow Coma Scale (GCS) score remained at 15, with no neurological deficits. He was started on antiepileptic medication, his blood pressure was ensured to be kept within normal parameters, he was given platelets with a target of above 80, and he was monitored closely for neurological deterioration.

One week after discovering his CSDH, he was noted to have worsening headaches, now associated with nausea and vomiting. A repeat brain CT demonstrated an increase in the size of the CSDH with worsening mass effect and an increase in midline shift to 16 mm (Fig. 1 right). On examination, the GCS score remained at 15 with no neurological deficits.

Treatment and Differentials

It was anticipated that the patient would have severe persistent thrombocytopenia, as he was due to have stem cell treatment and would be dependent on platelet transfusions up until several weeks after the stem cell treatment. He was deemed to have a high risk of morbidity and mortality with surgery due to his ongoing severe thrombocytopenia with a high risk of recurrence. The patient was counseled on these risks and the consensus was to trial nonoperative management except in the event of neurological deterioration, when emergency surgery would be performed. A neuro-interventional consult for MMA embolization was arranged after the discussion. The neuro-intervention team explained the procedure as well as the risks involved and emphasized, in particular, that this procedure was a relatively new procedure not usually done for the patient’s condition and that results may be difficult to predict. Despite the caveats, the patient opted to go ahead with MMA embolization.

Uncomplicated MMA embolization was performed with preinterventional platelet supplementation. Access was via the right distal radial artery snuffbox with a 6-Fr-diameter Benchmark 0.071” and Duo 0.013" system. The anterior, posterior, and accessory MMA branches were occluded with 33% n-butyl cyanoacrylate (glue), while the petrous division of the MMA was preserved. Use of n-butyl cyanoacrylate was chosen to allow embolization at more peripheral locations and reduce the risk of recanalization. Pre-embolization, the left MMA supply was found to be hypervascular, and postembolization, there was successful devascularization of the left hemispheric meningeal dura (Fig. 2). The dominant supply was found to be through the superficial temporal and occipital artery scalp-dural anastomoses. Recurrent meningeal branches of the internal carotid artery were not found. There were no postprocedural complications.

Outcome and Follow-Up

Over the next few days, the patient experienced a gradual decline in the frequency and severity of his headaches and nausea following the procedure. Head CT at 1-week postembolization demonstrated a reduction in the size of the CSDH, with the midline shift reduced from 14 mm to 9 mm (Fig. 3 left).

His next follow-up head CT at the 3-week follow-up showed that there was a significant reduction in mass effect with a midline shift of 7 mm. At his follow-up at 2 months postembolization, he was symptom free, and his CT head showed complete resolution of his CSDH (Fig. 3 right).

Discussion

Observations

CSDH has been increasing in incidence and is projected to be the most common neurosurgical diagnosis by 2030. This is due to

![FIG. 1. Preoperative CT images of the brain demonstrating CSDH with 11 mm of midline shift and significant mass effect (left) and an increase in size to 16 mm 1 week later (right).](image-url)

![FIG. 2. Embolization cast. Pre-embolization image (left) demonstrating a hypervascular left MMA territory. Postembolization image (right) demonstrating extensive devascularization of the left hemispheric meningeal dura. The dominant supply is now through the superficial temporal and occipital artery scalp-dural anastomoses.](image-url)
In this situation, MMA embolization acts to devascularize these subdural critical mass and the mass effect causes symptoms in the patient. Inflammation and progression are still considered to be poorly understood. It was once thought that CSDH arises after trauma that causes shearing of taut bridging cortical veins, resulting in slow bleeding into the subdural space that, over time, leads to a CSDH. This hypothesis, however, has been disputed mainly by the observation that there is usually a long delay from trauma to symptoms (4–7 weeks), much longer than one would expect from an actively bleeding vein that would most likely cause symptoms within days.

Our understanding of the pathogenesis of CSDH has evolved. It is now hypothesized that CSDH forms after trauma or microtrauma causes a splitting of the dural border cell layers, which leads to an inflammatory cascade with the release of angiogenic factors. This leads to the formation of inflammatory membranes and neovascularization of these membranes with fragile, leaky vessels that continue to exude fluid and blood into the area. Blood and inflammatory fluid slowly accumulate asymptptomatically until the size of the hematoma reaches critical mass and the mass effect causes symptoms in the patient. In this situation, MMA embolization acts to devascularize these subdural membranes and shift the fluid balance toward reabsorption.

Current use of MMA embolization has largely focused on reducing recurrence in elderly patients with significant medical comorbidities who present with CSDH. Additionally, there is some interest in the patient who is neurologically intact but symptomatic with CSDH but deemed a high risk for surgery. Notably there have not been any prospective or published randomized controlled trials demonstrating efficacy of the technique in treating CSDH. To the best of our knowledge, the present case of a young man with severe thrombocytopenia with an atraumatic large CSDH is the first case of successful treatment of CSDH with MMA embolization in this patient population. The patient made a full recovery with full resolution in 2 months following treatment.

Thrombocytopenia in neurosurgery is a major risk factor. Surgeons are faced with the dilemma of bleeding that requires potential surgery, but surgery can cause further bleeding. In a patient with non–life-threatening CSDH, nonoperative management can be reasonably considered. MMA embolization may be a possible tool in this setting that is of lower risk from a bleeding perspective but can treat the pathology.

The decision to operate is a challenging one due to the inherent risks of intracranial surgery for CSDH evacuation, including, most commonly, recurrence (25%), seizures, infection, and surgery-related acute intracerebral hemorrhage (1.7%). In particular, recurrence is associated with more than double the risk of other surgical complications.

Studies examining the risk of intracranial surgery in thrombocytopenic patients demonstrate a substantially increased risk in hematoma formation (40%) and associated increased morbidity and mortality. This improves in the setting of transfusion-corrected perioperative platelet counts, but refractory thrombocytopenia continues to demonstrate a very poor prognosis. This is true regardless of the type of surgery, where any degree of preoperative thrombocytopenia is associated with higher odds of readmissions, complications, and mortality.

Although not yet reported in the literature, there is a theoretical risk of distal embolic complications in thrombocytopenic patients undergoing MMA embolization. Rates of approximately 1.2% are seen in patients without thrombocytopenia. For this reason, MMA angiography prior to embolization to identify potentially dangerous anastomoses such as the ophthalmic artery, along with the use of particles greater than 150 microns to avoid cranial nerve injury, is emphasized. In large meta-analyses, MMA embolization has been demonstrated to be a safe technique for CSDH and a number of cases have also reported its safe use in the presence of concurrent anticoagulant therapy.

Treatment-related complications show no difference compared with traditional surgical intervention, including in the elderly population. The treatment failure rate is also much less frequent, 1.4% vs 27.5%, with patients having similar measures of independence postoperatively evaluated through the Rankin scale scoring system.

To investigate previous reports of MMA embolization for CSDH in patients, we searched PubMed using the keywords “middle meningeal artery embolization” and “thrombocytopenia” and “subdural hematoma,” which yielded 2 observational studies. In 2021, Lee et al reported 22 cases of MMA embolization in patients with cancer with refractory thrombocytopenia, with 77% having reduced subdural hematoma size and only 1 procedural complication. There has also been a report of 1 patient receiving MMA embolization in the setting of thrombocytopenia, which demonstrated no complications and resolution of the hematoma. Our case agrees with previous observations, which demonstrated this technique to be a safe and effective option for CSDH treatment. Mortality and morbidity in the former study tended to be very high due to other comorbidities, with 73% ultimately dying due to cancer complications.

Lessons
Given this is a single case report, very limited conclusions can be drawn. Although platelet supplementation was performed, it seems plausible that MMA embolization was causative in treating the CSDH. The patient had worsening symptoms and increased size of CSDH despite initial platelet supplementation; however, following MMA embolization, there was demonstrable reduction in the CSDH over the subsequent postoperative CT images of the brain at 1-week postembolization (left) with a reduction in midline shift from 14 mm to 9 mm and at the 1-month follow up (right) with complete resolution of CSDH.

J Neurosurg Case Lessons | Vol 5 | Issue 9 | February 27, 2023 | 3
few weeks. Systematic studies of this technique will provide stronger evidence regarding the efficacy of this technique in this patient population.

CSDH is a common neurological diagnosis and is often observed as a result of or in concurrence with patients experiencing refractory thrombocytopenia. In this situation, MMA embolization can be considered as a stand-alone or adjunct treatment option, due to the increased risks of surgery in thrombocytopenic patients and the growing body of evidence demonstrating the safety and efficacy of MMA embolization in this population. MMA embolization may reduce the likelihood of recurrence and surgical complications, while also improving functional outcomes.

References

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Correspondence
Emalee J. Burrows: Sir Charles Gairdner Hospital, Perth, Western Australia, Australia. emalee.burrows@gmail.com.