Cauda equina syndrome caused by multiple intraspinal hemorrhage following percutaneous coronary intervention: a case report and a review of the literature

Yong-Kuan Lim
Keelung Chang Gung Memorial Hospital of the CGMF

Pin-Yuan Chen
Keelung Chang Gung Memorial Hospital of the CGMF

Wei-Siang Chen
Keelung Chang Gung Memorial Hospital of the CGMF

Yu-Cheng Kao
Keelung Chang Gung Memorial Hospital of the CGMF

Mei-Mei Lin
Hualien Tzu Chi Hospital Buddhist Tzu Chi Medical Foundation

Yun-Cong Zheng (✉ m7630@cgmh.org.tw)
Chang-Gung Memorial Hospital and University  https://orcid.org/0000-0002-1935-527X

Research Article

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Abstract

Background

Percutaneous coronary intervention and dual antiplatelet therapy are common management for patients with coronary artery disease. Multiple spontaneous intraspinal hematomas mixed with epidural hematoma and subdural hematoma following regular percutaneous coronary intervention is an extremely rare complication. We describe our experiences to treat the elderly who presented with spontaneous spinal epidural hematoma and subdural hematoma in different spinal locations after percutaneous coronary intervention. Neurological examination and magnetic resonance imaging were followed to assess the treatment outcomes for more than 2.5 years.

Case presentation:

In this article, we present a 70 years-old male taking dual antiplatelet therapy for 1 year after drug-eluting stents implantation to right coronary artery and left anterior descending artery for non-ST elevation myocardial infarction had a sudden onset of paraplegia then the autonomic dysfunction immediately after another percutaneous coronary intervention. Whole spinal MRI showed mixed spontaneous spinal subdural hematoma and spinal subdural hematoma, included: anterior C5-T3 acute subdural hemorrhage, right lateral T4 to T8 epidural hemorrhage, and L5-S1 intrathecal hematoma. After urgent cauda equina neurolysis and T7-9 laminectomy to evacuate spinal epidural hemorrhage in accordance with the neurological symptoms, the patient regained walking ability immediately. The urination and defecation function recovered then. The surgical results maintained for at least 2.5 years, even after another percutaneous coronary intervention 1 year later.

Conclusions

Intraspinal hematomas in acute coronary syndromes are scarce but critical conditions after percutaneous coronary intervention. Multiple mixed spontaneous spinal subdural hematoma/ epidural hematoma could be fully reversed by circumspect neurolysis and limited laminectomy timely.

Background

A spontaneous spinal epidural hematoma (SSEH), defined by bleeding in the epidural space of the spine related to atraumatic etiology, with an incidence about 0.1 per 100,000 patients per year [1]. It is rare but a catastrophic neurological condition, usually need urgent surgical decompression [2, 3]. A spontaneous spinal subdural hematoma (SSDH), associated with permanent neurological deficits, is much less common than SSEH [4–6]. The clinical symptoms of SSEH or SSDH are sudden onset of neck or back pain initially associated with motor and/or sensory deficits in limbs, indicating spinal cord compression caused by the hematoma [7, 8]. Multiple intraspinal hematomas are more serious clinical conditions. As far as I know, mixed SSEH and SSDH causing CES as a complication of PCI has not been well documented.

Bleeding in the management of acute coronary syndromes (ACS) and percutaneous coronary intervention (PCI) has emerged in recent years, although it is relatively less to ischemic complications [9]. Mixed SSEH and SSDH following PCI lead to CES is true urgency to operation, especially in patients with rapid neurological deterioration. Meanwhile, platelet activation blocked by DAPT raises the difficulties of neurosurgery. We shared our management strategies for the CES caused by SSEH and SSDH. This scarce case highlights the importance of smooth coordination between cardiologist and neurosurgeon after the PCI to cope with possible complications of intraspinal hematomas.

Case Presentation

A 70-years-old male had no major medical history before until 4 years ago when he diagnosed as non-ST elevation myocardial infarction (NSTEMI), Killip 1. PCI was emergently introduced to the patient and coronary angiogram revealed coronary artery disease (CAD) with 2-vessel-occlusion. Successful percutaneous coronary angioplasty (PCA) from both radial arteries with 6F catheters and then drug-eluting stent (DES) for right coronary artery and left anterior descending artery were done. The patient had uneventful recovery following the procedure and he was discharged under DAPT control (aspirin at a dose of 100mg once daily and ticagrelor at a dose of 90mg twice daily). After 1 year of DAPT treatment, the medication was shifted to monotherapy with aspirin at a dose of 100mg once daily for 3 years.

Percutaneous coronary intervention

This time, he presented to a medical clinic with sudden onset retrosternal pain with radiation to back. After the transfer into our emergent room under the suspicion of ACS attack, the electrocardiogram disclosed equivocal ST-elevation with T-wave inversion. Ejection fraction of left ventricular was 61% in 2D-echocardiogram and Troponin-T was as high as 329.4 mg/dl. NSTEMI was impressed, 40mg methylprednisolone and local anesthesia with 2% lidocaine were prescribed as premedication. Cardiac catheterization revealed total occlusion of left circumflex artery majorly. PCI with 6F sheath was introduced from right radial artery and DES of Medtronic Resolute 2.5*38mm was placed to left circumflex artery. Activated clotting times of 290 to 300 seconds were maintained by using a weight-based dosing of 60 to 70 U/kg of heparin. DAPT (a aspirin at a dose of 100mg once daily and ticagrelor at a dose of 90mg twice daily) continued after the procedure.

Cauda equina syndrome
On admission to intensive care unit, vital signs were within normal ranges. Three hours after the PCI, he complained of severe backache from mid-thoracic to lower cervical level and it failed to relieve after acetaminophen 500mg oral intake. The patient stated no traumatic history since the last stent implantation 4 years ago. Then right leg weakness developed. Medical Research Council (MRC) scale down from 5/5 to 2/5, and it progressed to left lower extremity gradually. The strength of upper extremities remained intact. Bilateral pupil size was 3.0+/3.0+ and there was no sensation while urinating and defecating. Brain computed tomography for possible ischemic complication found no corresponding lesions. Neurological examination executed by the consulted neurologist disclosed acute paraplegia within 1 hour (MRC scale of bilateral lower extremities: 1/5) with hypoesthesia below T7 level, nearly anesthesia on lower limbs, and positive Babinski signs. Saddle anesthesia complicated with urine retention and incontinence led to indwelling catheter dependent. Rectal examination showed flaccid anal sphincter and reflex of bilateral Achilles’ tendon decreased.

The patient had no chest pain, dyspnea exacerbation, loss of consciousness, dizziness, upper limbs weakness, facial palsy, disorientation, and blurred speech. The neurologist suggested CES rather than ischemic stroke. The auxiliary data of the blood and coagulation parameters showed mildly decreased platelet count (145 /L) and high Troponin-T (2336 mg/dl) but other coagulation factors were within the normal range (Table 1). Magnetic resonance imaging (MRI) of the spine revealed multiple intraspinal hematomas, include: C5 to T3 segmental subdural hematoma ventral to the spinal cord (Fig. 1AB), T4 to T8 epidural hematoma compressed the spinal cord dorsally (Fig. 1CD), and intrathecal hematoma at L5 to S1 deviated the nerve roots from normal distribution (Fig. 1EF). Interrupted intraspinal hematomas distorted the cord and cauda equina.

**Surgical strategies**

Dexamethasone phosphate 5mg was given via intravenous route to reduce secondary neurological insults and acetaminophen was held due to critical bleeding with increasing Troponin-T. The neurosurgery team discussed the urgent situation about CES and possible treatment options with his daughter. After the hesitation of other family members, surgical decompression carried out immediately within 8 hours after neurological symptoms attacks. L5 to upper S2 wide laminectomy was performed initially for the incontinence (Fig. 2A). A 4.5 cm cyst content with blood compressed underlying eliminated (Fig. 2AB) and further neurolysis to roots of the cauda equina executed cautiously under the microscope (Fig. 2C). No anatomical variations were detected during surgery. Dural closure was accomplished after good hemostasis (Fig. 2D).

Due to incomplete spinal cord impairment below T7 level, we decided to perform limited T7 to T9 laminectomy to evacuate the epidural hematoma. Liquefied hematoma gashed out with pressure and was aspirated to achieve full cord decompression. Two epidural drains guaranteed the relief of epidural pressure then. Total operation time was 6 hours and blood loss was 400cc. 12U platelet transfusion was done following the operation. We took conservative observation to cervical subdural hematoma because no obvious neurological signs over upper limbs and no severe cord compromised by hemATOMA.

**Multidisciplinary care**

After the surgery, we withdrew ticagrelor and adjusted the DAPT as aspirin at a dose of 100mg once daily and clopidogrel at a dose of 90mg once daily. Intravenous dexamethasone phosphate 10mg continued four times daily. Immediately, muscle strength of left lower leg near totally recovered on postoperative day 1 (MRC scale of left lower extremity: 4/5). The neurological deficits kept improving include saddle sensation and he was transferred to neurosurgery ward under lumbar brace protection on postoperative day 2 as gradually declined Troponin-T level. On postoperative day 5, epidural drains were pulled out. Muscle strength of lower extremities continued improving after the hemovac drains removal on postoperative days 5 through 9. The movement became symmetry (MRC scale of bilateral lower extremities: 4+/5) on postoperative day 10 under comprehensive rehabilitation. And he started to aware urination and stool passage but still was unable to control it. He tried the catheter removal but failed due to hematuria and the difficulty in voiding after that. On postoperative day 15, he could sit at a wheelchair stably without numbness over the saddle area. On postoperative day 17, the patient was able to ambulation without assistance. He was discharged home without fecal incontinence on day 30. The outpatient urology follow-up was arranged since Foley catheter dependence.

**Postoperative follow-up**

DAPT therapy backed to aspirin and ticagrelor after the hematuria improved. Benign prostatic hyperplasia was diagnosed at the urology clinics. After medication, self urination achieved one month later (Fig. 3). Two months after the surgery, spinal MRI revealed the resolution of hematomas and the regression of the mass effect over the whole spinal cord but residual seroma over the lumbosacral junction left (Fig. 1G). The patient returned to his normal life independently without obvious neurological sequelae.

Half year later, he received scheduled PCI for the restenosis of previous DES noted by the last coronary angiogram. DES for left main to the proximal anterior descending artery and distal right coronary artery were installed smoothly under proper sedation and analgesics control. There were no bleeding complications of the PCI. The same DAPT continued to prevent stent thrombosis and the benefits of neurosurgical intervention maintained without recurrence till now (Fig. 4). The seroma was replaced by contrast-enhancing granulation at L5 level in MRI follow-up 18 months after the operation leading to mild numbness over right calf laterally which didn't influence the quality of daily life (Fig. 1H).

**Discussion**

Our report of the first documented case of CES caused by segmental SSEH and SSDH as a complication of PCI with DES illustrates several aspects of this emergency event that are still not fully understood. The patient had received aspirin and ticagrelor as DAPT before and after this PCI in recent 5 years. According to the time frame, he suffered from CES immediately after the PCI. It was thought to be the major cause of spontaneous spinal hematomas, rather than DAPT.
From a study reported by Philippe Gabriel Steg, the frequency of bleeding in ACS and PCI underestimated due to the highly selected patients at low risk of bleeding. And the increasing use of combinations of antithrombotic drugs and PCI may increase the risk of bleeding complications. The bleeding rate was higher in the primary PCI than elective PCI and the bleeding site was more common on the arterial access site [9]. Our case had multiple intraspinal bleeding sites was extremely rare.

Older age, lower body weight, and invasive procedures use were powerful predictors of bleeding complications in ACS and PCI. In a study by Moscucc, the adjusted odds of having a major hemorrhage prior to discharge increase by about 30% per decade of age as reported in Global Registry of Acute Coronary Events (GRACE) [10]. The hypothesis is that collagen and amyloid deposits in the tunica media layer of the vascular, increase with aging, may cause brittle vessels that are less inclined to constrict and are more prone to bleed [10].

Until now, the cause of the SSEH is still controversial and it doesn't have a definitive known cause for most of the cases, as well as SSDH, due to its rarity in general populations (Table 2). Some believed that SSEH is caused by the ruptured epidural veins of the weakened venous plexus especially at the dorsal epidural veins of the spinal cord [3]. Meanwhile, SSDH tends to occur on the ventral side of the spinal canal since the dura matter adheres more tightly to the posterior longitudinal ligament than the ligamentum flavum [11, 12]. They hypothesized “a minor trauma or forgotten effort leading to increased intrathoracic and intraluminal pressure of the vessels traversing the subarachnoid space” is the etiology of SSDH [13, 14].

The Valsalva maneuver may increase intra-abdominal and intra-thoracic pressure which transferred to the spinal epidural venous system and makes the valveless nature of the epidural venous plexus ruptured [15]. The consequently elevated intra-thoracic pressure could also damage intraluminal vessels. This vicious circle probably leads to mixed SSEH and SSDH present at the same time. Although we cannot verify the direct etiologies, we suggest that the pre-existing weakened venous wall due to aging, the DAPT therapy for ACS and the Valsalva maneuver due to discomfort during the period of the PCI intervention contributed to the segmental SSED with SSDH and following CES.

When bleeding complications occur, the area of hematoma is hard to self-limited due to concurrent DAPT that inhibits the activity of the platelet. We adjusted the DAPT as aspirin and clopidogrel according to more pronounced platelet inhibition effect and higher rate of non-procedure-related bleeding along with ticagrelor [16]. After the neurosurgery, we shifted the DAPT back to aspirin and ticagrelor to prevent thromboembolic events of the PCI intervention based on the guideline for the management of patients with NSTE-ACS [17, 18].

Conclusion

As more aggressive intervention and anti-thrombotic therapy established for ACS, complications of the spinal hematomas should be taken into consideration. The outcome of SSEH and SSDH could be favorable if it is diagnosed timely and treated early. If the duration of the complete neurological deficits is less than 12 hours, it still could be reversed by the urgent operation [7].

Abbreviations

ACS  acute coronary syndromes
CAD  coronary artery disease
DAPT  dual antiplatelet agents
DES  drug-eluting stent
MRC  Medical Research Council
MRI  magnetic resonance imaging
NSTE-ACS  non-ST elevation acute coronary syndrome
NSTEMI  non-ST elevation myocardial infarction
PCA  percutaneous coronary angioplasty
PCI  percutaneous coronary intervention
SSDH  spontaneous spinal subdural hematoma
SSEH  spontaneous spinal epidural hematoma

Declarations
**Ethical approval:** All procedures performed in studies involving human participants were in accordance with the ethical standards of the institutional and with the 1964 Helsinki declaration and its later amendments.

**Consent to participate:** The need for consent to participate was waived by the above committee. The IRB is organized and operates in accordance with Good Clinical Practice and the applicable laws and regulations (IRB file No 202000270B0). Thus, our study was followed by the World Journal of Emergency Surgery guidelines of retrospective ethics approval.

**Availability of data and material:** The datasets generated during and/or analysed during the current study are available from the corresponding author on reasonable request.

**Competing interests:** All authors certify that they have no affiliations with or involvement in any organization or entity with any financial interest (such as honoraria; educational grants; participation in speakers’ bureaus; membership, employment, consultancies, stock ownership, or other equity interest; and expert testimony or patent-licensing arrangements), or non-financial interest (such as personal or professional relationships, affiliations, knowledge or beliefs) in the subject matter or materials discussed in this manuscript.

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**Authors’ contributions:**
Zheng YC, Lim YK, Chen PY, Chen WS, Kao YC, Lin MM contributed to the data collection and analysis.
Zheng YC, Lim YK, Chen PY contributed to the conception and design of the study.
Zheng YC and Lim YK contributed to drafting the text and preparing the figures.

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

Dr. Yun-Cong Zheng, as a corresponding author as mentioned above had nothing to disclose of the submitted work which title “Treat cauda equina syndrome following percutaneous coronary intervention” (Including grants, data monitoring board, study design, manuscript preparation, statistical analysis, etc.)

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**References**


### Tables

**Table 1** Laboratory data of patient at symptoms presentation

<table>
<thead>
<tr>
<th>Variable</th>
<th>Reference range, adults</th>
<th>On presentation</th>
</tr>
</thead>
<tbody>
<tr>
<td>Hematocrit (%)</td>
<td>41–53</td>
<td>35.3</td>
</tr>
<tr>
<td>Hemoglobin (g/dl)</td>
<td>13.5–17.5</td>
<td>13.0</td>
</tr>
<tr>
<td>White cell count (per mm3)</td>
<td>4500–11,000</td>
<td>8700</td>
</tr>
<tr>
<td>Differential count (%)</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Neutrophils</td>
<td>40–70</td>
<td>80.3</td>
</tr>
<tr>
<td>Lymphocytes</td>
<td>22–44</td>
<td>14.2</td>
</tr>
<tr>
<td>Monocytes</td>
<td>4–11</td>
<td>4.7</td>
</tr>
<tr>
<td>Eosinophils</td>
<td>0–8</td>
<td>0.6</td>
</tr>
<tr>
<td>Basophils</td>
<td>0–3</td>
<td>0.2</td>
</tr>
<tr>
<td>Platelet count (per mm3)</td>
<td>150,000–400,000</td>
<td>145,000</td>
</tr>
<tr>
<td>Red cell count (per mm3)</td>
<td>4,500,000–5,900,000</td>
<td>3,970,000</td>
</tr>
<tr>
<td>Mean corpuscular volume (fl)</td>
<td>80–100</td>
<td>88.9</td>
</tr>
<tr>
<td>Mean corpuscular hemoglobin (pg)</td>
<td>26–34</td>
<td>32.7</td>
</tr>
<tr>
<td>Mean corpuscular hemoglobin level (g/dl)</td>
<td>31–37</td>
<td>36.8</td>
</tr>
<tr>
<td>Red cell distribution width (%)</td>
<td>11.5–14.5</td>
<td>12.3</td>
</tr>
<tr>
<td>Prothrombin time (sec)</td>
<td>11–14</td>
<td>11.4</td>
</tr>
<tr>
<td>Prothrombin time international normalized ratio</td>
<td>0.9–1.1</td>
<td>1.1</td>
</tr>
<tr>
<td>Partial thromboplastin time, activated (sec)</td>
<td>22.1–35.1</td>
<td>31.1</td>
</tr>
</tbody>
</table>

†Reference values are affected by many variables, including the patient population and the laboratory methods used. The ranges used at Chang Gung Memorial Hospital are for adults who are not pregnant and do not have medical conditions that could affect the results. They may therefore not be appropriate for all patients.

**Table 2** Overview of articles involved in spontaneous intraspinal haemorrhages
### Spontaneous spinal hemorrhage types and possible causes

<table>
<thead>
<tr>
<th>Type</th>
<th>Description</th>
<th>Causes</th>
<th>References</th>
</tr>
</thead>
<tbody>
<tr>
<td>SSDH</td>
<td>(Outcome: prognosis is determined by the the neurological status at presentation)</td>
<td>Pereira BJ, de Almeida AN, Muio VM, de Oliveira JG, de Holanda CV. Predictors of Outcome in Nontraumatic Acute Spinal Subdural Hematoma and Literature Review. World Neurosurg 2016;89:574-7 e7.</td>
<td></td>
</tr>
</tbody>
</table>

### Figures

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Serial T2-weighted magnetic resonance images before and after the event. Whole spinal MRI at symptoms onset revealed C5 to T3 segmental subdural hematoma ventral to the spinal cord and presented slight hyperintensity in the sagittal view (A) and the axial view (B). Another high signal epidural hemorrhage extended from T4 to T8 right laterally, causing compressive myelopathy in the sagittal view (C) and the axial view (D). Moreover, the deposit of intrathecal hematoma at the L5-S1 level showed hypointensity in the sagittal view (E) and the axial view (F). The Sagittal MRI that was performed 2 months after the operation showed the complete resolution of cervicothoracic hemorrhage with a 6.8 cm residual seroma at L4-S1 level (G). At one and a half years following the event, the spinal hemorrhage was completely resolved, and L5 granulation tissue remained (H). Green arrows in the sagittal view marked the area of intraspinal hematomas. White arrows in the axial view indicated the boundary between hematomas and spinal cord or roots.
Figure 2

Surgical procedure and intra-operative findings in lumbosacral spine. (A) shows a 4.5 cm cystic lesion with blood content causing tension in cauda equine after L5 to the upper S2 laminectomy and dura mater opening. (B) shows the ventral displacement of cauda equina during the removal of the mass. (C) shows nerve root preservation under the microsurgical neurolysis of cauda equina, thereby further relieving the tension. (D) shows dural closure after good hemostasis.
Figure 3

Summary of the emergent course

- Patient was diagnosed with NSTEMI
- PCI with DES was performed for the right coronary artery and left anterior descending artery
- DAPT (Aspirin at a dose of 100 mg once daily, and Ticagrelor at a dose of 90 mg twice daily) was prescribed after the procedure for 1 year and was then adjusted to mono therapy of Aspirin.

2015

- Patient had NSTEMI again and PCI with DES was introduced to the left circumflex artery
- Heparin was used during the PCI and DAPT was continued after the procedure
- 3 hours after PCI, the patient complained of back pain, then paraplegia, urinary and fecal incontinence
- MRI of the spine revealed multiple intraspinal hematomas
- Laminectomy and removal of hematomas was performed.
- DAPT was adjusted to mono antiplatelet therapy of Ticagrelor after the operation.
- After adequate neurological improvement, DAPT was continued back to Aspirin and Ticagrelor.

2018

- Neurological deficits were fully recovered

2019

- Another PCI with DES was performed to the restenosis of left main anterior descending artery and right distal coronary artery
- No complications occurred this time

2020
Figure 4

Summary of the longterm outcomes