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ABSTRACT

Background. Spontaneous spinal hematoma (SSH) is a rare condition that can result in severe functional disability and even death. But early detection and prompt intervention can substantially reduce the morbidity. We present a series of seven operated cases of SSH.

Methods. All operated cases of SSH between 2017 and 2019 were studied. The demographic and clinical features, risk factors and imaging features were analyzed. The functional outcome at discharge and 6 months were assessed.

Results. Seven operated cases of SSH with mean age 35(SD-20.9) were studied. Six cases were spontaneous spinal extradural hematomas (SSEDH) and one case was spontaneous spinal subdural hematoma (SSSDH). The most common site was cervicothoracic. Risk factors associated with SSH were thrombocytopenia, pregnancy, and necrotising pancreatitis. Two patients had preoperative Frankel's grade A-B, three had grade C and two had D. The mean interval between the onset of symptoms and surgery was 4.7days.

The functional outcome was dependent on the pre-operative functional status of the patient. Patients with SSEDH and thrombocytopenia had a poor outcome.

Conclusion. SSH even though spontaneous may be associated with risk factors. The presence of thrombocytopenia and preoperative functional status predicted outcome. This is the only single institution case series to report thrombocytopenia as a factor predicting poor outcome.

INTRODUCTION

Spontaneous spinal hematomas (SSH) are relatively rare and the majority of spinal hematomas are due to trauma. SSH includes both spinal extradural hematoma as well as subdural hematoma. The incidence of spontaneous spinal extradural hematoma (SSEDH) is around 0.1/100000 and that of spontaneous spinal subdural hematoma (SSSDH) is still rarer (1)(2). These conditions usually present as emergencies with rapidly progressive neurological deficits. Prompt recognition and early intervention is very important for a better outcome. Spinal hematoma was defined spontaneous when there was

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no associated trauma, iatrogenic injury (secondary to procedures like lumbar puncture, epidural catheter insertion), or post-operative spinal hematoma.

MATERIALS AND METHODS

Setting: Department of Neurosurgery, Government Medical College, Kozhikode, Kerala which is a 3000 bedded teaching hospital in south India where around 2000 neurosurgical cases are operated annually.

Cases: All operated cases of spontaneous spinal hematomas during 2017-2019

All operated cases of SSH during the study period were studied. History was taken to assess any risk factors like hypertension, coagulopathies or bleeding diathesis. Duration from the onset of symptoms to surgery was noted. A neurological examination was done and the preoperative functional status was assessed by Frankel's grade. All patients were evaluated with 1.5T MRI with contrast and MR angiography. The site of the bleed was assessed from MRI and the number of spinal segments involved was noted. All patients underwent standard decompressive laminectomy and evacuation of bleed. Laminectomy was done at the level of maximal cord compression. A maximum of two-level decompressive laminectomy was done. After surgery, the functional status was reassessed at discharge and 6 months. All patients with improvement in Frankel's functional scores were taken as a good outcome.

REPRESENTATIVE CASES

Case 1

A 75-year-old male who was on treatment for necrotizing pancreatitis developed sudden onset of neck pain followed by weakness of bilateral upper limbs and lower limbs. The patient had numbness of the body up to the neck. There was also associated urinary retention. On examination, there was grade 3 power of upper limbs and grade 2 power of lower limbs and blunting of all sensations below the clavicles (Frankels grade C). His routine blood workup showed elevated prothrombin time with an INR of 2.3, platelet count and activated partial thromboplastin time were normal. His S Amylase was 4300. MRI of the cervical spine showed T1 and T2 hyperintense signal from C2 to C6 with cord compression (Figure 1) suggestive of SDH. After correcting the coagulopathy with fresh frozen

plasma transfusions, we proceeded with C4C5 decompressive laminectomy. Dura was bulging with underlying hematoma and was opened in the midline. Thick SDH was evacuated. Postoperatively the lower limb power improved to grade 3 (Frankels grade C). At 6 months follow up both lower limbs and upper limbs had grade 4 power (Frankels grade D).

Case 2

24-year-old 34 weeks pregnant lady presented with complaints of sudden onset neck pain and rapidly progressive weakness of both lower limbs. On examination, she had grade 2 power of both lower limbs and sensory loss below the nipple level (Frankels grade C). Her routine blood investigations were within normal limits. MR imaging of the spine showed T2 intermediate signal lesion noted involving left posterolateral epidural space at C7-T1 vertebral level suggestive of hematoma (Figure 2). The management difficulty in view of 34 weeks completed pregnancy with regards to positioning as well as chances of intrauterine death were well explained. She was taken for caesarian section and after the procedure, in the same sitting C7T1 laminectomy was done in the prone position and the hematoma was evacuated. Postoperatively patient showed improvement in motor power. The baby was shifted to the neonatal ICU for further care. At 6 months follow up the patient was able to walk with support and do her day-to-day activities (Frankels grade D).

Case 3

A 25-year-old male patient presented with fever for 10 days and sudden onset of back pain for one day. He developed rapidly progressive weakness of lower limbs with urinary retention. On examination, he had grade 0 power of both lower limbs and there was sensory blunting below the nipple (Frankels grade B). His lower limb reflexes were absent and plantar was mute bilaterally. On routine blood profile, his platelet count was 17000 but markers for dengue were negative. MR imaging of his spine showed cervicothoracic C5-T11 region spinal anterior extradural hematoma with spinal canal compromise and cord compression. After correcting the platelet count with platelet transfusion, we proceeded for surgery. T4 T5 two-level decompressive laminectomy was done (at the level of maximum cord compression). Postoperatively patient remained

with the same preoperative motor power. At six months follow up the lower limbs were spastic with

flexor spasms and no spontaneous power (Frankels grade B). He is still on a urinary catheter.

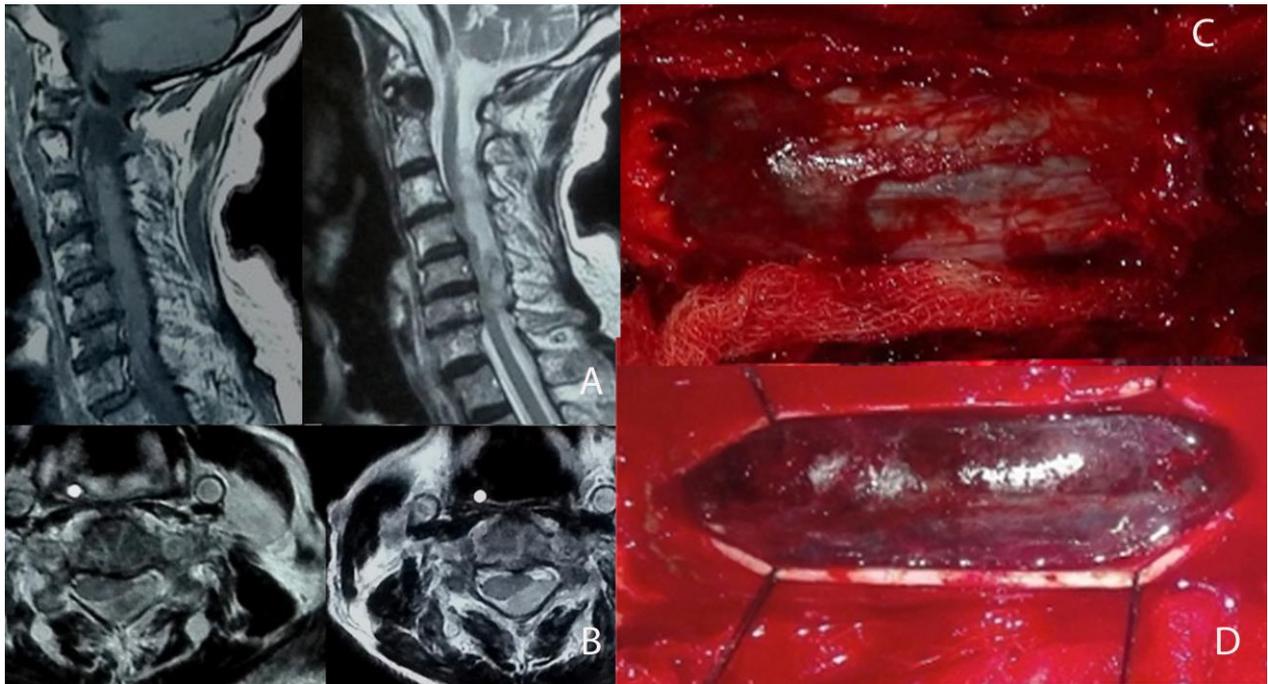


Figure 1. A- T1 T2 sagittal MR section of the cervical spine showing spinal SDH extending from C2-C6 B- T1T2 axial MR sections C- Per operative image showing bluish bulging dura with underlying hematoma D- dura is kept open with thick SDH.

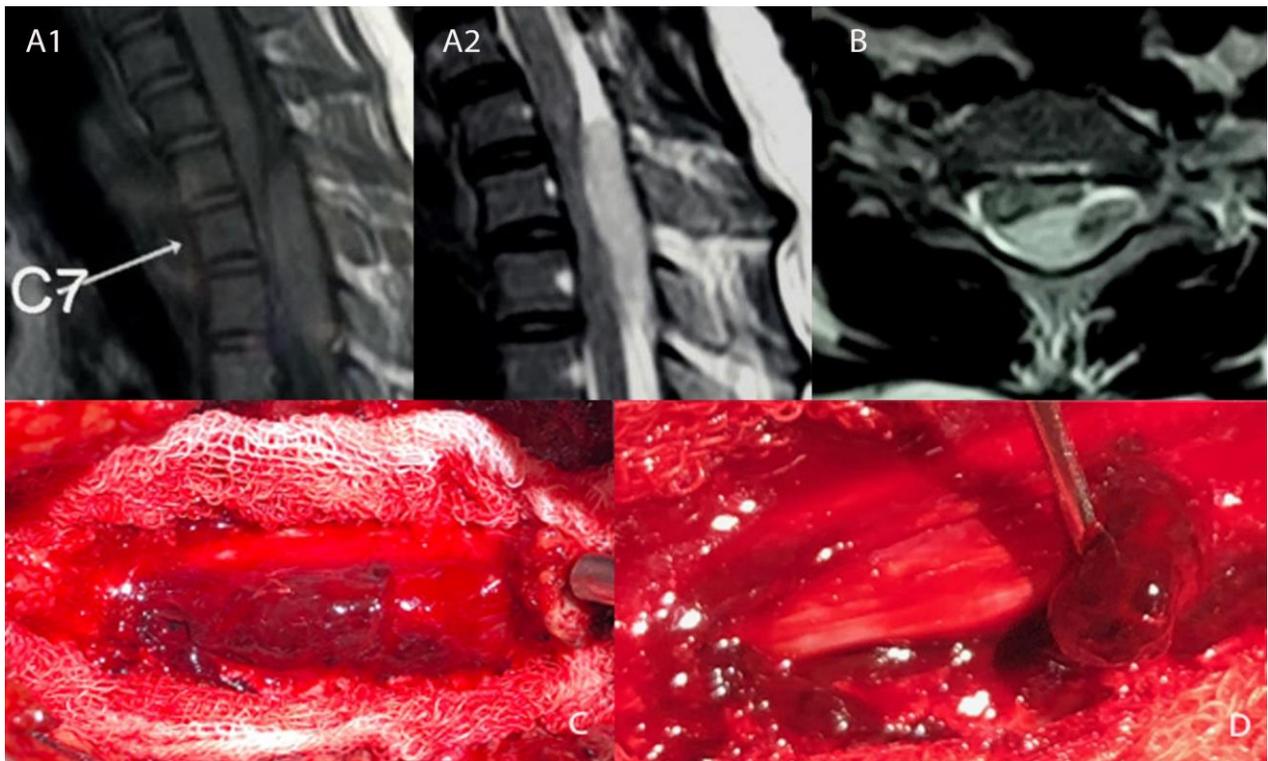


Figure 2. A1, A2- T1 and T2 sagittal MRI of the cervical spine showing SSEDH extending from C6-D2; B- axial MR sections showing SSEDH; C, D – per operative images of decompressive laminectomy and hematoma evacuation.

Cases	Age	Sex	Risk Factor	Platelet count/ μ l	INR	Pre-op Frankel's Grade	Type of SSH	Spinal Level of SSH (segments)	Time from onset to maximal weakness (hours)	Duration till Surgery (Days)	Frankel's Grade at discharge	Frankel's Grade 6 months follow up
1	25	M	Thrombocytopenia	17000	1.1	B	EDH	C7-D11 (13)	6	1	B	B
2	29	M	Nil	4.4 lakhs	1	D	EDH	D1-D2 (2)	72	5	E	E
3	24	F	Pregnancy	4.2 lakhs	0.8	C	EDH	C7-D1(3)	12	2	C	D
4	75	M	Necrotising Pancreatitis	3.3 lakhs	2.3	C	SDH	C2-C6 (5)	24	2	C	D
5	30	M	Thrombocytopenia	15000	1.2	A	EDH	C7-D1(3)	8	2	A	A
6	12	M	Thrombocytopenia	10000	0.9	C	EDH	C7-D3 (5)	10	2	C	C
7	50	F	Nil	2 lakhs	1.1	D	EDH	C5-D4 (8)	12	1	E	E

INR- International normalized ratio

Table 1. Demographic and clinical characteristics of the patients

RESULTS

Of the 7 cases, the mean age was 35 ± 20.9 and 5 (71%) were males (Table 1).

Thrombocytopenia was the most common risk factor and was present in three of our patients. One patient was 34 weeks pregnant and another had necrotising pancreatitis with coagulopathy. Two of them had no known identifiable risk factor. One patient had SSSDH while the rest had SSEDH. The most common level affected for SSEDH was cervicothoracic ($n=5$). The SSSDH was purely cervical. The craniocaudal extension of the hematoma was variable with the longest involvement of 13 spinal segments and the shortest of two segments. Of the SSEDH one patient had anterior spinal EDH and the rest had posterior spinal EDH. All patients had local back pain as the first symptom which heralded the onset of neurological deficits. Five of the patients with Frankel's grade less than 'D' had urinary retention. Five patients had preoperative Frankel's grade A-C and two had 'D'. All patients were operated at the earliest once the general conditions were favourable and the thrombocytopenia/ coagulopathy was corrected. The mean time between the onset of symptoms to maximal weakness was 20.5 hours. But in patients with thrombocytopenia, this was 8 hours. The mean interval between the onset of significant symptoms and surgery in our series was 4.7 days. Of our seven cases, four had improvement in their

Frenkel score and were categorised as a good outcome. At 6 months, all the remaining patients remained in their same preoperative Frenkel score. Those who had thrombocytopenia as risk factor had a poor outcome. Among the patients with thrombocytopenia two patients had fever with thrombocytopenia and for one patient thrombocytopenia was detected on the preoperative blood workup. Also, when the pre-operative functional status was low (Frenkel's A, B, and C) the outcome was poor. The improvement of functional status was significantly dependent on the pre-operative Frenkel's score. We couldn't find any association between the number of segments involved or the time period between the onset of symptoms to surgery with postoperative outcome.

DISCUSSION

SSEDH was first described by Jackson and Bain in the second half of the 19th century while SSSDH was first reported by Schiller (3)(4). SSH is a very rare clinical entity and its presentation can be variable from mild local pain to severe functional impairment in the form of weakness and bowel and bladder involvement. Usually, spinal hematomas are associated with trauma, iatrogenic procedures or spinal surgeries but we studied the patients who had a spontaneous spinal hematoma. The risk factors for SSH include AVMs, coagulation abnormalities,

anticoagulant use, vertebral haemangiomas, hypertension and pregnancy (5). However, in 40-60% of cases demonstrates no identifiable risk factor (6). In two of our patients, we could not identify any risk factors.

Thrombocytopenia was the most common risk factor for SSH in our study which was not reported before. Thrombocytopenia may be associated with a febrile illness which is common in northern Kerala where this study was conducted (7). In our series, two patients with thrombocytopenia had a preceding febrile illness, probably due to viral infection. The most common cause for viral infection with thrombocytopenia is dengue infection which was screened in all patients and was found negative. In a previous study from north Kerala, 26% of cases of thrombocytopenia associated with febrile illness were due to viral infections other than dengue(7). Viral infections causes thrombocytopenia both by decreased production of platelets from bonemarrow, as well as by the increased destruction by antibodies(8).

SSEDH is usually due to bleeding from posterior epidural venous plexus. But Beatty and Winson had postulated an arterial origin for SSEDH particularly in the cervical region since the intrathecal pressure is higher than the venous pressure(9). In all of our cases, we did not find any evidence of vascular malformation with imaging (MR angiography) as well as perioperatively. Bleeding from anterior epidural veins is rarer since they are smaller in caliber and are situated underneath the posterior longitudinal ligament. But anatomical variations are possible where the anterior epidural plexus can have a larger caliber. Among our SSEDHs one was located anterior to the thecal sac. The most common site of SSH in our series was cervicothoracic, probably due to mechanical factors as well as the prominence of epidural veins in the cervical and thoracic regions (6).

The factors associated with poor outcomes were thrombocytopenia and pre-operative functional status. The post-operative recovery depends on the extend of ischemia to the cord which depends on various factors like rate, force, and duration of compression(10). We postulate that in patients with thrombocytopenia as the basic platelet plug formation is not happening to arrest the bleeding there will be a rapid accumulation of blood in the epidural space(11-13). Also, the endothelial supporting function of the platelets is lost which

heralds the bleeding in thrombocytopenia causing endothelial gaps for RBC extravasation(12). Here the rate and force of compression may be very high on the cord and may result in neuronal death. All of our patients with thrombocytopenia had rapid deterioration of their motor power (mean-8hours). This shows the rate and force of compression of the spinal cord are high in these patients which resulted in poor postoperative outcomes. After a thorough literature search, this is the only single-institution case series to report thrombocytopenia as a factor predicting poor outcome. From the literature review, various studies have reported different values of platelet count below which there is a significant risk of major bleeding(14,15). We could not find any association between outcome and time took for surgical intervention from the onset of weakness.

Previous studies have shown that the most important factor determining the long-term outcome is the neurological status of the patient before surgical intervention. Other bad prognostic factors are the onset of severe symptoms in a shorter time frame, the involvement of thoracic cord and lack of sensory sparing. The evacuation of the SSEDH within 12 hours has been reported to predict the outcome(16). We found that the preoperative functional status predicted the outcome in our series.

The presence of SSEDH in pregnancy is even more rare and only 27 cases have been reported^[16,17]. Several theories are attributed to the occurrence of SSEDH during pregnancy. Usually, these hematomas tend to occur during the third trimester as the pressure in the vertebral venous plexus is normally elevated due to the compression by the gravid uterus(20). When there is a sudden change in pressure, as if when the patient sneezes, coughs, or during voiding there can be rupture of these veins resulting in SSEDH.

One of our cases was an SSSDH, which was due to coagulopathy secondary to necrotising pancreatitis of unknown etiology. The pathophysiology of coagulopathy in severe pancreatitis is due to the activation of platelets and various inflammatory cytokines which will result in consumption coagulopathy(21,22). The SSSDH is produced by the bleeding from subarachnoid vessels as the spinal subdural space is devoid of blood vessels unlike the cranial subdural space. The rupture of subarachnoid vessels may be due to trivial

trauma or sudden fluctuations in pressure due to coughing, sneezing, etc. Recently radiculomedullary veins were also postulated to be involved in spinal subdural bleeds (23).

CONCLUSIONS

The most important risk factor which caused SSH in our series was thrombocytopenia. The most common site was cervicothoracic. The outcome was dependent on preoperative functional status. Thrombocytopenia was noted as a bad prognostic factor in our study. The outcome was not dependent on the extent of hematoma or time period between symptom onset and surgery.

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