

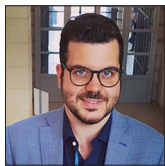
Case Report

Spontaneous anterior cervicothoracic spinal epidural hematoma extending to clivus in SARS-CoV-2 infection

Gianluca Scalia¹, Giuseppe Emmanuele Umama², Salvatore Marrone³, Francesca Graziano¹, Angelo Giuffrida¹, Giancarlo Ponzo¹, Massimiliano Giuffrida¹, Massimo Furnari¹, Gianluca Galvano⁴, Santo Bonanno⁵, Giovanni Federico Nicoletti¹

¹Department of Neurosurgery, Highly Specialized Hospital and of National Importance "Garibaldi"; ²Department of Neurosurgery, Cannizzaro Hospital, Trauma Center, Gamma Knife Center, Catania; ³Department of Experimental Biomedicine and Clinical Neurosciences, School of Medicine, Postgraduate Residency Program in Neurological Surgery, Neurosurgical Clinic, AOUP "Paolo Giaccone", Palermo; ⁴Department of Diagnostic Imaging, Interventional Radiology and Neuroradiology, Highly Specialized Hospital and of National Importance "Garibaldi"; ⁵Department of Emergency, Acute Care and Observation Medicine, Highly Specialized Hospital and of National Importance "Garibaldi", Catania, Italy.

E-mail: *Gianluca Scalia - gianluca.scalia@outlook.it; Giuseppe Emmanuele Umama - umama.nch@gmail.com; Salvatore Marrone - salvo.mr89@gmail.com; Francesca Graziano - fragraziano9@gmail.com; Angelo Giuffrida - dr.angelogiuffrida@gmail.com; Giancarlo Ponzo - giancarlo.ponzo@gmail.com; Massimiliano Giuffrida - mass.giuffrida@tiscalinet.it; Massimo Furnari - massimofurnari@alice.it; Gianluca Galvano - gianlucagalvano@gmail.com; Santo Bonanno - santob72@tiscali.it; Giovanni Federico Nicoletti - gfnicoletti@alice.it



*Corresponding author:

Gianluca Scalia,
Department of Neurosurgery,
Highly Specialized Hospital
of National Importance
"Garibaldi", Catania, Sicily, Italy.
gianluca.scalia@outlook.it

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ABSTRACT

Background: The treatment of spontaneous spinal epidural hematomas (SSEHs), depending on the lesion size and myelofurcular involvement, can be surgical or conservative. Here, we present a 55-year-old patient who sustained a SSEH several months following a systemic SARS-CoV-2 infection.

Case Description: A 55-year-old immunocompromised female (i.e., history 17 years ago of Hodgkin's lymphoma, nodular sclerosis variant) recently developed a SARS-CoV-2 infection treated with nonsteroidal anti-inflammatory agents. She then reported the sudden onset of cervicodorsalgia after a slight cervical flexion/extension maneuver. The brain and cervicothoracic spine MRI studies documented a clival anterior spinal epidural hematoma with maximum spinal compression at the T1-T2 level; it also extended inferiorly to the T6 level. Two weeks later, the follow-up MRI showed a remarkable reduction in the anteroposterior diameter of the hematoma that correlated with significant neurological improvement and almost complete pain regression. She was discharged after a total 15-day hospital stay, with complete symptoms relief.

Conclusion: We present a 55-year-old chronically immunocompromised (i.e., due to the history of Hodgkin's lymphoma) female who, following a SARS-CoV-2 infection, developed an anterior SSEH extending from the clivus to the T6 spinal level that spontaneously regressed without surgical intervention.

Keywords: COVID-19, Epidural, Hematoma, Spinal, Spontaneous

INTRODUCTION

Epidural spinal hematomas have an estimated incidence of 0.1/100,000 patients/year. Most occur in the second and seventh decades, due to hematological, infectious, neoplastic conditions, or the use of anticoagulants. They are more frequently seen in the posterior vertebral canal and can be spontaneous or secondary (e.g., traumatic and iatrogenic) in origin. Treatment of spontaneous spinal epidural hematomas (SSEHs) can be surgical or conservative, depending on

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the lesion size and neurological deficit (i.e., myeloradicular involvement). The COVID-19 infection may result in severe thromboembolisms, cardiovascular/cerebrovascular accidents, or hemorrhagic complications occurring even months following active infections. Here, we report a 55-year-old chronically immunocompromised female who, following a recent COVID-19 infection, spontaneously developed a SSEH extending from the clivus to T6 which spontaneously resolved on successive MR studies without the need for surgery.

CASE DESCRIPTION

History and neurological examination

A 55-year-old female had a history of Hodgkin's lymphoma (nodular sclerosis variant CD15- and CD30+) diagnosed 17 years earlier based on a left hypertrophic/confluent supraclavicular lymph nodes biopsy. She had 4 cycles of chemotherapy (ABVD scheme) and supraclavicular lymph nodes excision. The subsequent repeated PET studies were negative (i.e., without radiopharmaceutical captures).

Recent COVID-19 infection

She recently had a paucisymptomatic SARS-CoV-2 infection treated with nonsteroidal anti-inflammatory agents (NSAIDs). She reported the sudden onset of cervicodorsalgia after a routine cervical flexion/extension maneuver. The pain increased despite the administration of NSAIDs (VAS score 7/10), and she remained neurologically intact. Her laboratory studies, however, demonstrated an elevated white blood cell count, a low mean platelet volume, and elevated erythrocyte sedimentation rate [Table 1].

MR imaging

The patient underwent brain, cervical, and thoracic spine MR studies that documented a clival through T6 anterior spinal epidural hematoma. There was a high signal in the cord opposite the T1-T2 level of maximum compression. Further, T1-weighted Gd-enhanced MRI sequences confirmed peripheral epidural and meningeal contrast enhancement consistent with the diagnosis of a SSEH [Figure 1].

Medical course

The patient underwent a multidisciplinary assessment that required serial MR/other imaging and hematological evaluations. Notably, the cervico/thoracic spine MRI repeated 14 days later showed a progressive SSEH reduction; this also correlated with near-complete pain regression [Figure 2]. The patient was discharged asymptomatic on the 15th hospital day.

Table 1: Laboratory tests.

Marker	Value	Laboratory range
Creatinine	0.41 mg/dL	0.50–0.90
Total proteins	5.8 g/dL	6.6–8.7
Albumin	3.30 g/dL	3.68–5.75
Alpha-2 macroglobulin	1 g/dL	0.51–0.85
Beta-1 microglobulin	0.31 g/dL	0.34–0.52
Gamma-globulin	0.59 g/dL	11.10–18.80
White blood cells	15.71×10 ³ /μl	4–10
Platelets	483×10 ³ /μl	150–450
Hemoglobin	12.60 g/dL	12–16.50
Mean platelet volume	6.22 fl	7.20–11.10
Hematocrit	36.81%	38–48
Neutrophils	80.3%	35–75
Lymphocytes	11%	25–55
Sodium	133 mmol/L	136–145
Prothrombin activity	68%	80–120
International normalized ratio	1.30	0.95–1.15
C-reactive protein	24.7 mg/L	0.1–5
Erythrocyte sedimentation rate	48 mm	0–18

DISCUSSION

The ability to treat SSEH with/without surgery depends on their location, severity of MR-documented cord compression, and neurological deficits. Groen showed that 80% of patients with SSEH could be managed conservatively and fully recovered.^[1] Lim and Wong recently reported a 79-year-old patient who did not require surgery following a SARS-COVID-19-related SSEH extending from the C3 to C6 levels.^[3]

COVID-19 hypocoagulability syndromes

Although the correlation between anticoagulant therapy (i.e., NSAIDs) and SSEH risk has long been known, it is newly documented that COVID-19 infections may lead to disseminated intravascular coagulation and/or other bleeding diatheses (MOD).^[6] In this case, on admission with COVID-19, the patient was placed on enoxaparin (i.e., protocol low dose and in combination with tranexamic acid). Recently, it has been postulated that the early use of tranexamic acid with anticoagulant therapy may reduce the progression of COVID infectious damage, although only in selected cases with normal D-Dimer values.^[5] In a patient such as ours with a history 17 years previously of Hodgkin's lymphoma, there was an increased potential for evolving hyperinflammation and hyperclotting.^[7]

Chronic vasculitis/hyper-hypocoagulation syndromes persist long term after COVID-19

It is likely that a vascular pathologic processes may persist even after regression of COVID-19. The malacia of post-COVID

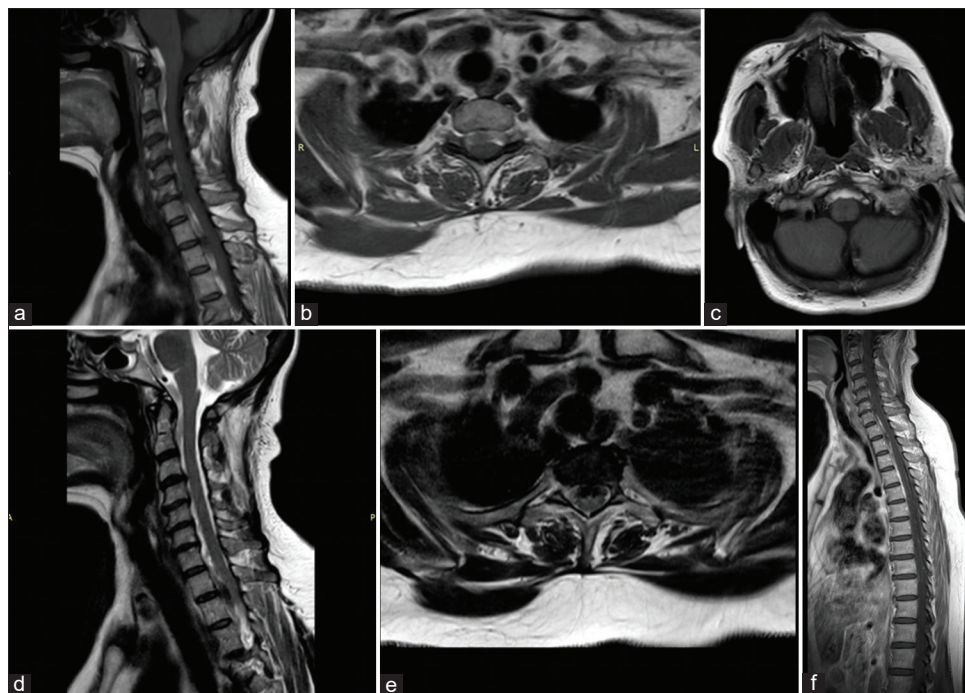


Figure 1: Sagittal (a), axial (b) T1-weighted cervicothoracic spine MRI images and axial brain MRI (c) documented an anterior spinal cervicothoracic and clival epidural hyperintensity and hypointensity in sagittal (d) and axial (e) T2-weighted sequences with maximum spinal compression at Th1-Th2 level and extension to Th6. T1-weighted Gd-enhanced MRI sequences (f) also showed a peripheral epidural and meningeal contrast enhancement.

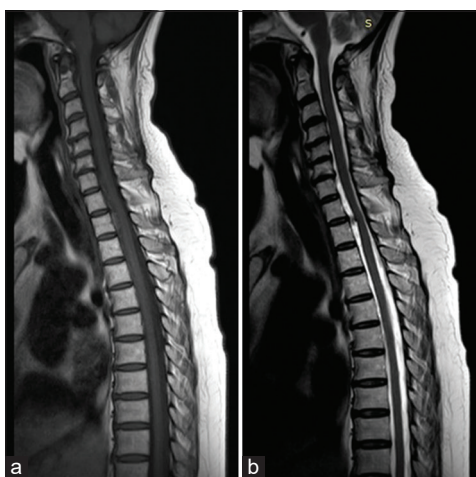


Figure 2: Sagittal T1-weighted (a) and sagittal T2-weighted (b) cervicothoracic spine MRI with extension to the craniovertebral junction showing an almost total reduction of epidural hematoma.

vasculitis would be an essential prerequisite to explain the subsequent SSEH in this patient.

Two pathogenic hypotheses leading to SSEH

There are two enteropathogenic hypotheses attributed to COVID-19 that could have led to the SSEH in this patient. The first related to increasing in intrathoracic pressure, the

second to microtrauma between vessels – pathological structures (i.e., spondylosis-osteophytes or hypertrophic posterior longitudinal ligament).

Evolution of SSEH

Slow progression of bleeding with SSEH is mostly of venous origin, although there are cases of rapid progression as reported by Liao *et al.*^[2] The epidural vascular plexus consists of low-pressure thin-walled veins, without valves that are in communication with the thoracic-abdominal venous system. They can, therefore, “erupt” after sudden intrathoracic pressure increases (i.e., Valsalva maneuvers).^[8] In the 1960s, Markham *et al.* described the “SSEH Syndrome”^[4] variously attributed to sudden bending, rapid turning in the bed, explosive sneezes, intense cough, and even coitus, due to the transient pressure. The more mechanistic hypothesis occurs due to rupture of the anterior venous plexus due to microvascular trauma (blood vessels against an osteophyte as in abrupt flexion-extension maneuvers).

CONCLUSION

The authors described a peculiar case of a 55-year-old female with a recent COVID-19 infection, and a likely persistent postinfection vasculitis that developed an extensive

spontaneous anterior clivus-T6 SSEH, spontaneously resolved on successive MR studies without surgery.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent.

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Nil.

Conflicts of interest

There are no conflicts of interest.

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