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Case Report

Spinal subdural hematoma associated with phenytoin use

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A rare case of spinal subdural hematoma (SSDH) is reported in a patient who was receiving phentoin. The hematoma occurred because of combined effects of trauma and underlying haematological disturbance induced by phenytoin. A young male who was a known case of seizure disorder and was receiving phenytoin for last 10 years presented with history of seizure followed by fall from one storey of his house. This was followed by paraplegia. MRI revealed presence of spinal subdural hematoma in dorsolumbar spine. His haematological parameters revealed deranged coagulogram and anemia with thrombocytopenia. The patient underwent laminectomy and evacuation of hematoma and intrathecal drain was kept in for three days till drainage was clear. However the patient did not improve clinically. His anemia and thrombocytopenia responded rapidly to administration of vitamin B12 and stoppage of phenytoin. SSDH should be kept a possibility in posttraumatic paraplegia. The patients receiving phenytoin must be monitored for change in hematological parameters after chronic use.

Key words: Spinal, subdural, hematoma.

INTRODUCTION

Spinal subdural hematoma (SSDH) is a rare entity usually associated with trauma, iatrogenic manoeuvres, coagulation disorders, anticoagulant therapy, underlying neoplasms, arteriovenous malformations (Boutobza et al., 2001; Chau and Tiu, 2008; Guthikonda et al., 1979; Hausmann et al., 2001; Solomon et al., 1977). SSDH is very rare in absence of these conditions (Kyriakides et al., 2007). So far only 19 cases of SSDH have been reported in literature, the majority with a bleeding diathesis and after a lumbar puncture. Most common site is thoracic region, while one case has been reported in lumbosacral spine associated with anticoagulant therapy (Guthikonda et al., 1979).

It usually presents with sudden pain radiating to limbs or trunk and may be associated with varying degree of motor, sensory and autonomic disturbances (Kyriakides et al., 2007). Headache or neck stiffness may also be presenting symptom due to meningeal irritation, this headache is thought to arise from spinal subarachnoid hemorrhage co existing with SSDH (Russel and Benout, 1978).

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CASE REPORT

A 25 years male who was a known case of seizure disorder and was receiving phenytoin for last 10 years presented with history of seizure followed by fall from one storey of his house. Patient had normal neurological status except for complete flaccid paraplegia in both lower limbs and sensory level was T10. MRI was done which revealed SSDH extending from D9 to L3 (Figures 1 and 2). Base line investigations were done which revealed platelets of 36,000/cul.mm, Hemoglobin (Hb) was 6.6 g, Prothrombin time (PT) was 20 s (control 14 s), Prothrombin time (PTI) OF 70%, INR of 1.56, total leucocytes of 13,000 cubic mm. Patient was prepared for urgent surgery and both platelet rich plasma and fresh frozen plasma (20 ml/kg body weight) was administered. Laminectomy was done and findings revealed dura was tense, blue in colour, (Figure 3) acute SDH was present and clots pouted through the incision. Hematoma was evacuated completely with copious irrigation with saline and an intrathecal drain was kept in which was removed after 4 days when CSF was clear. The patient was not a vegetarian, nor had he any history suggestive of malabsorption. The facility for serum folate and B 12 levels is not available here so it was not done however,

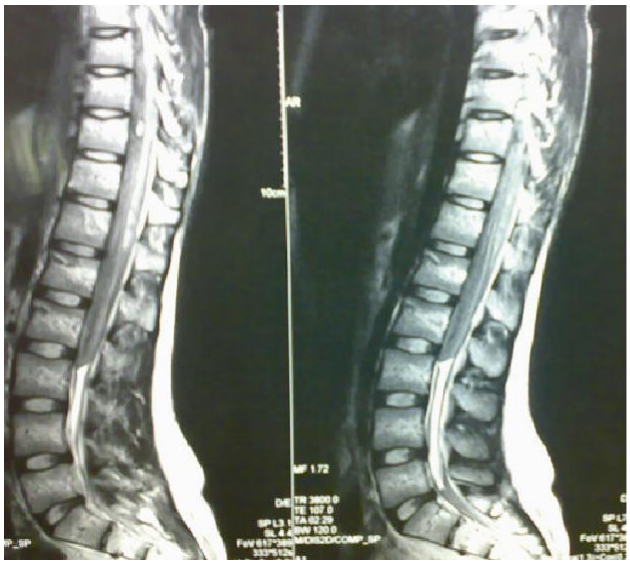

Figure 1. Sagittal T2W and MRI showing presence of subdural spinal hematoma on posterior aspect of cord. Note is especially made of meniscus sign on lower end of haematoma in Figure 1.

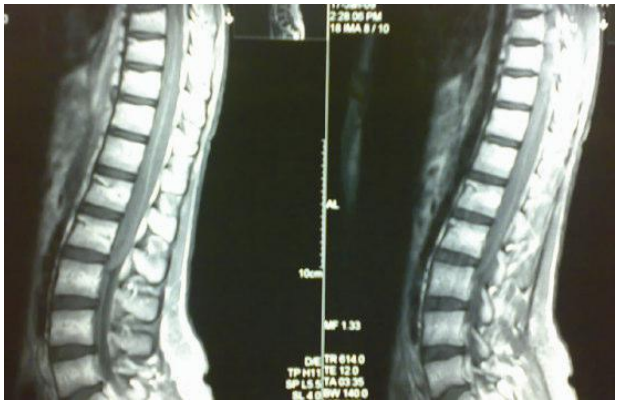

Figure 2. Sagittal T1W and MRI showing presence of subdural spinal hematoma.

bone marrow examination was done which did not show any evidence of aplastic anemia. Consultation was sought from haematologist who after detailed peripheral blood smear and bone marrow examination made an impression that thrombocytopenia and anaemia were due to phenytoin induced megaloblastic anemia. Phenytoin was stopped and he was put on valproic acid. He was administered multivitamin injections including that of vitamin B12. Repeat investigations were done after 4 days on which showed Hb of 8.9 g and platelets of 43,000/cu.mm, PT 16 s (control 14 s), PTI 87.5%, INR 1.18. After three more days patient had Hb 11.8 g platelets of 100,000/cu.mm. He had no improvement in power at the time of discharge (on Sixth day). Unfortunately the patient did not make any improvement on follow up.

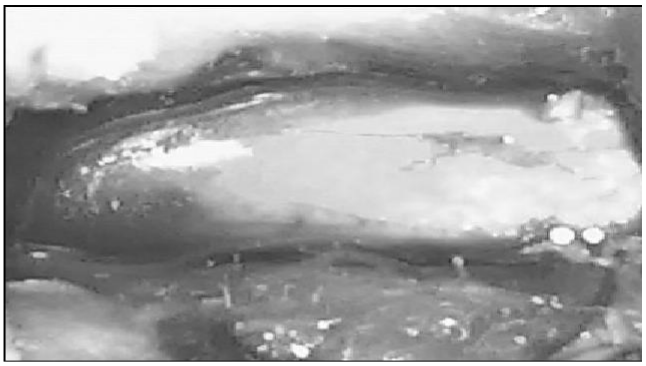

Figure 3. Dura tense, bulging and discoloured due to underlying hematoma.

DISCUSSION

The origin of bleeding in SSDH is controversial. It may arise from SAH that dissects through the arachnoid membrane into subdural space (Kang et al., 2000) or a sudden increase in abdominal or thoracic pressure may lead to rupture of intraspinal vessels and resulting in hematoma formation (Russel and Benout, 1978), or increase in pressure is transmitted intravascularly along the lateral spinal segments of vessel, such a rapid increase cannot be neutralised by simultaneous increase in spinal fluid pressure because of shielding effect of spinal column and ligament (Ryota et al., 2006). This also may be a contributing factor to hematoma in our case as he bled after a seizure which is known to increase intraspinal pressure.

Montano et al. (2008) reported a case of SSDH in a patient with autosomal dominant polycystic disease who received transplant 10 years back and presented with acute low backache with fecal and urinary incontinence and had T7 level of hypoesthesia with paraparesis, this patient had SSDH at T6-T8. This condition with polycystic disease has been associated with increased vascular fragility. On reviewing literature only a handful cases of SSDH secondary to underlying hematological disorder or an iatrogenic coagulopathy have been reported (Gerson et al., 1983; Montano et al., 2008; Chau and Tiu, 2008). In the absence of haematological disorder the entity is even rarer.

SSDH has been seen in 14 years boy who did not have any coagulation defect, the child had an incidental tectal mass, bleeding was suspected to be due to bleed in that mass but post operative studies revealed no change in size or character of mass or no evidence of hemorrhage, thus bleeding may have originated from subarachnoid space (Michael and Frederick, 1991). One case has been associated with intra spinal neurilemmoma, since intra spinal neurilemmoma is subarachnoid in location so bleeding may originate from that space (Michael and Frederick, 1991). SSDH has been seen in patient who had history of recent atrial fibrillation and received vitamin k therapy (Uri et al., 2009). Association of phenytoin with

bleeding tendencies is not common. Targan et al. (1975) reported a case of disseminated intra vascular coagulation with purpura fulminans in a patient who was receiving phenytoin for seizure disorder.

In one of the series, two cases of anticonvulsant induced megaloblastic anaemia have been seen, both of these were deficient in folic acid and had extremely low levels of serum B12 levels apparently associated with defective B12 absorption due to deficiency of intrinsic factor. Both showed impaired intestinal absorption of D-xylose and absorptive defects produced by drugs may play part in initiating anticonvulsant induced megaloblastic anaemia and that once deficiencies of hemopoetic factors are established, a vicious cycle may be set up owing to the effects of these deficiencies on gastrointestinal tract (Reynolds et al., 1965). In our case features of megaloblastic anemia was there. Besides marrow suppression can occur due to phenytoin. Gerson et al. (1983) reported a 53 year old man who developed aplastic anaemia from phenytoin and carbamazepine. Both these drugs undergo metabolism to potentially toxic arene oxide intermediates, these arene oxide intermediates have a role in causing aplastic anaemia. In humans with anticonvulsant induced aplastic anaemia there is increased susceptibility to toxicity which is due to inherited abnormality in metabolite detoxification. Toxicity of electrophile metabolite may result from covalent bonding of intermediates to cell macro molecules. Covalent bonding of metabolites could lead to bone marrow toxicity through variety of mechanisms. Direct stem cell toxicity or mutation or immunological processes involving formation of haptens or damage to lymphocytes with critical functions in hematopoesis could occur.

In our case anemia can be explained by the occurrence of megaloblastic anaemia and thrombocytopenia is due to ineffective erythropoiesis. Long term use of phenytoin has been associated with biochemical evidence of folic acid deficiency and rarely with megaloblastic anaemia. The mechanism is uncertain but is thought to result from phenytoin induced alteration in intestinal absorption of conjugated and/or free dietary folate (Nelson et al., 1978). Folate deficiency resulting from long term phenytoin therapy is a common occurrence but progression to a megaloblastic anaemia is rare. The supplementation of folic acid to folate deficient patient taking phenytoin has been shown to result in lowered serum concentration of phenytoin and possibly loss of
control of seizure disorder (Rivey et al., 1984).

MRI is the investigation of choice. SSDH is seen as a space occupying lesion usually ventral and contained within the duramater and MRI can demonstrate variable T1 and T2 signals depending on age of hematoma.

Subdural hematomas appears as concave on sagittal and irregular on axial on MR imaging. Subdural hematomas are located within thecal sac, so are separate from adjacent extradural fat (Rivey et al., 1984).

Surgery is the ideal management modality and decompression should be done as early as possible. The functional outcome is dependent on timing of surgery and is significantly better when done within 36 h in patients with complete sensorimotor loss and within 48 h in patients with incomplete sensorimotor loss (Chau and Tiu, 2008). Recent reports have pointed out possibility of spontaneous resolution of SSDH and conservative treatment can be recommended for patients with stable neurological status (Rivey et al., 1984).

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