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Case Report

Hidden treasures: Incidental findings in two cases of chronic subdural hematoma

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ABSTRACT

Background: Extramedullary hematopoiesis (EMH) and plasmacytomas occurring within the cranium are rare entities.

Case Description: We review two cases in which patients presented with subdural hematoma and underwent evacuation. On routine histopathologic examination of their membranes, both patients were subsequently found to have focal EMH, as well as a clonal plasma cell proliferation in one case.

Conclusion: EMH is rare and usually found in individuals with profound and chronic anemia. However, this entity may be more common in chronic subdural hematomas. Solitary extraosseous plasmacytoma is exceedingly rare in the cranium, and its presence in chronic subdural hematoma membranes is of uncertain significance. The cytokine milieu that promotes organization of chronic subdural hematomas may play a role in the establishment of both of entities in this location.

Keywords: Case report, Extramedullary hematopoiesis, Plasmacytoma, Subdural hematoma

INTRODUCTION

The routine microscopic examination of subdural hematoma membranes is typically unexciting beyond offering some information on the chronicity of the lesion. Here, we present two cases of subdural hematoma which were remarkable for focal clusters of ectopic cells, prompting further workup.

CASE PRESENTATIONS

Case 1

An 86-year-old female with a history of atrial fibrillation on anticoagulation was taken to an outside hospital emergency department after being found down in her home with encephalopathy. A CT scan of the head was performed which showed a 13 mm thick left-sided subdural hematoma with a rightward midline shift of 6 mm. She was then transferred to our institution for further management. Her anticoagulation was discontinued and she underwent reversal with an anti-inhibitor coagulant complex. Repeat imaging on admission showed no interval change. During

her hospitalization, the patient's mental status improved, and she was discharged to an inpatient rehabilitation facility.

Approximately 10 days later, follow-up CT head showed evidence of rebleeding with an interval increase in the size of the left subdural hematoma to 15 mm and an increase in the rightward midline shift to 8 mm [Figure 1]. The patient was readmitted and noted to have new-onset right arm weakness. She underwent craniotomy and evacuation of the left subdural hematoma, and the membranes were submitted to pathology for examination.

The specimen consisted of fragments of brown-red tissue measuring 6.7 × 4.5 × 1.6 cm in aggregate and was grossly consistent with a chronic and organized subdural hematoma. On microscopic examination, there were several small clusters of cells with the appearance of erythroid islands and immature mononuclear cells noted within the membranes [Figures 2a and b]. Immunohistochemical (IHC) staining for E-cadherin and CD61 was performed which highlighted erythroid precursors and rare possible megakaryocytes, respectively, supporting the presence of multiple foci of extramedullary hematopoiesis (EMH) within the membranes of this subdural hematoma.

In addition, in one tissue block, there was a cluster of cells morphologically resembling plasma cells [Figure 3a]. IHC staining for CD138 and cytoplasmic kappa and lambda light chain [Figures 3b-d] revealed this to be a cluster of kappa-restricted plasma cells suggestive of a small extraosseous plasmacytoma. The patient had no history of hematologic malignancy and no clinical or laboratory evidence of end-organ damage related to myeloma.

Case 2

A 66-year-old male with diabetes presented with an 8-week history of chronic occipital headaches and tinnitus. The patient's primary care physician obtained imaging of the brain which showed bilateral subacute-on-chronic subdural hematomas with 6 mm of the leftward midline shift [Figure 4a]. There was no known precipitating trauma or fall. He underwent right craniotomy and evacuation of the right subdural hematoma, and the membranes were submitted to pathology for examination.

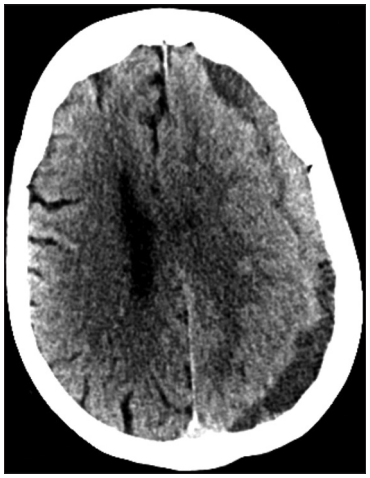

Figure 1: Noncontrast CT showing 15 mm thick left-sided subdural hematoma with the rightward midline shift.

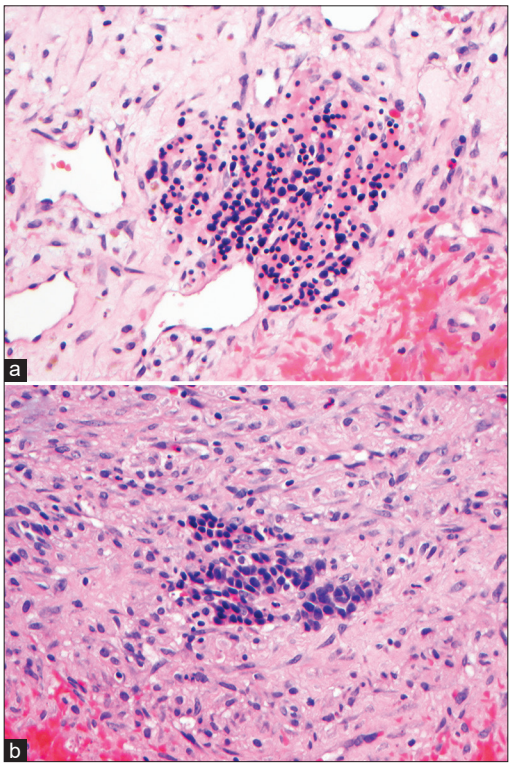

Figure 2: H&E ×100, EMH with erythroid islands (a) and immature mononuclear cells (b).

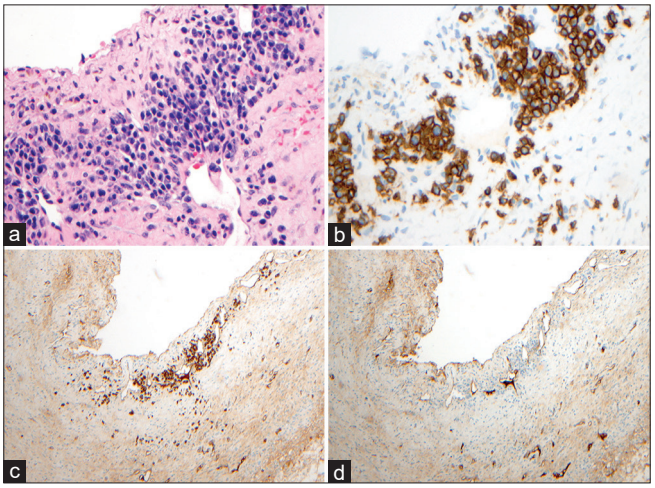

Figure 3: (a) H&E ×400: Large cluster of plasma cells, (b) CD138 ×400: Cells are positive for CD138, (c) Kappa ×100: Cells are positive for kappa IHC staining, and (d) lambda ×100: Cells are negative for lambda IHC staining.

The specimen consisted of fragments of tan-brown, hemorrhagic, soft and membranous tissue measuring 2.7 × 2.0 × 0.4 cm in aggregate grossly consistent with a chronic and organized subdural hematoma. On microscopic examination, there were several erythroid islands noted [Figure 4b] which were highlighted by IHC staining for E-cadherin [Figures 4c]; in addition, there were rare megakaryocytes [Figure 5a] which were highlighted by CD61 [Figure 5b], respectively, supporting the presence of multiple foci of EMH within the subdural hematoma.

DISCUSSION

These routine subdural hematoma specimens both revealed ectopic clusters of cells that were striking enough to prompt further workup. Both EMH and plasmacytomas are most commonly encountered in the bone or marrow elements, and their presence in the cranium is rare. Below is a brief review of the phenomena of intracranial EMH and intracranial plasmacytomas occurring in chronic subdural hematomas.

Intracranial EMH arising in the dura is very uncommon. It has been described in individuals with chronic anemia, including patients with thalassemia and myeloproliferative neoplasms.[3,7] However, EMH occurring in chronic subdural hematomas may not be so uncommon and has been described in several case reports [5,6,10] and case series.[4,9] These case series found evidence of EMH in 41 out of 130 (32%) and 13 out of 38 (33%) of chronic subdural hematomas examined. The case series were both single center studies, had unclear case selection criteria, and variable sampling methodology. However, they do suggest that incidental EMH in subdural hematomas is not uncommon.

Several hypotheses have been put forward to explain this phenomenon. In one case, the patient's subdural hematoma occurred in proximity to a skull fracture and the authors suggested that local extravasation of marrow elements into the hematoma might account for EMH.[6] The authors of one case series noted that EMH was found in proximity to granulation tissue and postulated its development from stem cells found in granulation tissue.[4] This latter thought highlights the role of inflammation in the formation of subdural hematomas and suggests a role for cytokine mediated homing of hematopoietic progenitors to the hematoma. Although EMH in subdural hematoma is likely of little clinical significance, for the pathologist, awareness of this entity is important to avoid mistaking the hematopoietic elements for a metastasis or hematologic malignancy.

Intracranial solitary extraosseous plasmacytomas (SEP) are very rare. They have occasionally been reported to arise in the dura or brain parenchyma, coming to clinical attention by detection on imaging or by causing mass effect.[1,13] The rarity of the entity and the microscopic nature of the monoclonal plasma cell cluster here found incidentally in a subdural

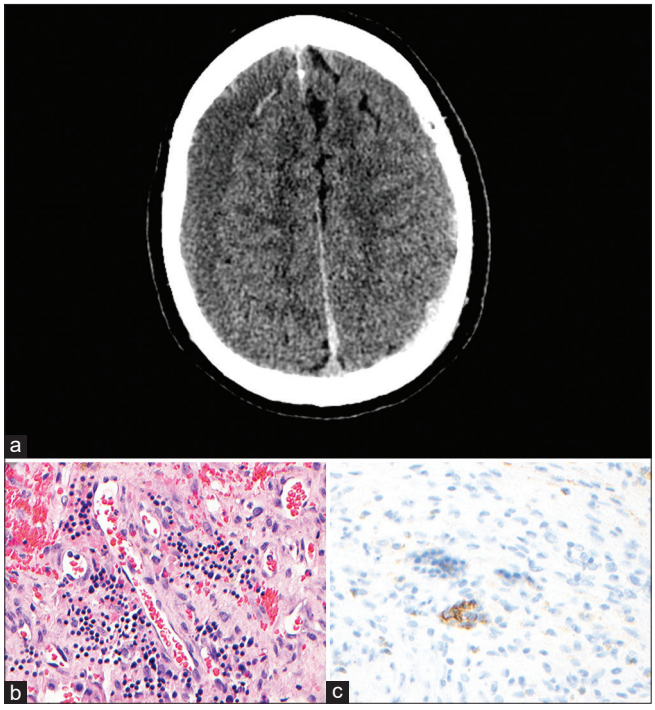

Figure 4: (a) Noncontrast CT showing bilateral subdural hematomas with leftward midline shift, (b) H&E ×400: Perivascular erythroid islands, and (c) E-cadherin ×400: e-cadherin stains immature erythroid precursors.

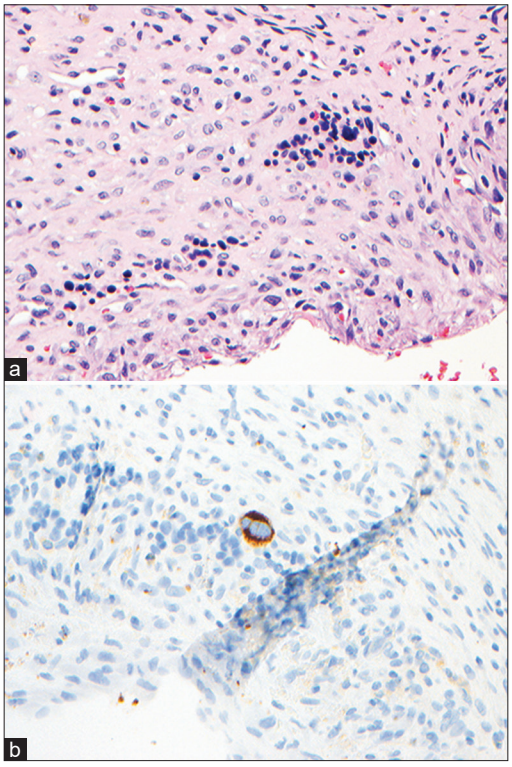

Figure 5: (a) H&E ×400: Erythroid islands and a possible megakaryocyte and (b) CD61 ×400: Highlights a megakaryocyte.

hematoma cast doubt on whether this lesion represents a true SEP. One case report of an intracranial plasmacytic lesion suggested that these lesions are on a spectrum and used the term "atypical monoclonal plasma cell hyperplasia" to describe a similar lesion which the authors postulate may represent a preneoplastic precursor of plasmacytoma.[13] Because SEPs in general present a modest rate of recurrence (25%) and occasionally progress to plasma cell myeloma (15%),[12] the finding in our case was discussed with the clinical team. Ultimately, continued surveillance was chosen instead of further testing for an underlying hematologic malignancy given the uncertain clinical significance of the finding and the patient's goals of care.

It is also worth reflecting on whether these foci of EMH and the possible SEP within this patient's subdural hematoma have a common origin. The formation of a chronic subdural hematoma is driven by inflammation mediated by an array of cytokines.[11] Inflammation in chronic subdural hematoma leads to membrane formation, angiogenesis, and can induce a vicious cycle of repeated inflammation as newly formed vessels leak and rebleed.[2] It is possible that this cytokine milieu could also induce chemotaxis of hematopoietic progenitor cells to the site of an organizing hematoma. Similarly, the generation of plasma cells is driven by a complex array of cytokine-mediated cell signaling.[8] Ongoing inflammation provides an opportunity for tissue B-lymphocytes to undergo activation and terminal differentiation, with the potential to acquire pathogenic genetic alterations on the way. Ordinary SEPs most commonly form in the upper respiratory tract and the GI tract, sites prone to antigen exposure and inflammation.

CONCLUSION

Intracranial EMH is rare and usually found in individuals with profound and chronic anemia. However, this phenomenon may be more common in chronic subdural hematomas; despite their intracranial location, the cytokine milieu that promotes organization of chronic subdural hematomas may play a role in the establishment of foci of EMH. SEP is exceedingly rare in the cranium; the incidental finding of a minute, clonal plasma cell proliferation in the chronic subdural hematoma of Case 1 is of unknown significance and deserves further study because of the known risk of recurrence and progression in SEP of other sites, ultimately, to plasma cell myeloma.

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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