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A rare case of non-traumatic rupture of spleen secondary to Plasmodium ovale malaria

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ABSTRACT

Objective: Plasmodium ovale malaria occurs mainly from mild form of malaria. We present a rare case of a splenic rupture secondary to complication of Plasmodium ovale malaria.
Clinical presentation: A 41-year-old female from Mali admitted to intensive care unit with hemorrhagic shock secondary to splenic rupture. A laparoscopic exploration was performed and patient received a massive blood transfusion for a hemodynamic stabilization. The diagnosis of malaria was confirmed by a blood smear test indicating the presence of P. ovale. A treatment of injectable Quinine was initiated with a positive outcome.
Conclusion: Although usually considered as a cause of a mild form of malaria, P. ovale may be responsible for a ruptured spleen which can lead to a state of life- threatening hemorrhagic shock.

Clinical presentation

This is the case of a 41 year old female. Patient is from Mali and has been living in France for 5 years. She has no significant past medical history. She sought medical care for intermittent nocturnal fever, generalized abdominal pain, vomiting and profuse diarrhea for almost one month prior to admission.

Upon arrival to the emergency room, patient was slightly pale and hypotensive with blood pressure of 90/54 mmHg, tachycardic with heart rate of 100 beats/min, febrile with temperature of 38.3 °C and the oxygen saturation level was 92 % on room air.

Findings on physical examination: The abdominal palpation revealed diffuse tenderness over the left hypochondrium and epigastrium. Patient had an episode of bilious vomiting and profuse diarrhea.

The laboratory tests yielded the following results: Microcytic anemia of 9.2 g/dl, thrombocytopenia of 93 g/l, TP level of 59 % and a CRP of 138 mg/l. The electrolyte panel, kidney and liver function tests were within normal limits.

The patient's condition deteriorated rapidly with the onset of hemodynamic instability followed by a shock requiring a vascular filling with 2 L of crystalloids. Patient's hemoglobin level dropped to 5.8 g/dl, her platelet count to 57 G/L and TP level to 59 %. The patient was transfused with 6 CGR and 5 PFC and received vasopressor amine (Noradrenaline). An abdominal and pelvic CT scan showed a massive hemoperitoneum with splenic rupture and active bleeding (Figs. 1 and 2).

A hemostasis splenectomy (with drainage of 2.5 L of hemoperitoneum) was quickly performed to stabilize the hemodynamic state. The patient was then transferred to the post-operative intensive care unit.

An anatomopathological examination of the spleen revealed a main part of 348 g measuring 19 × 9 × 5 cm associated with another hemorrhagic fragment of 15 × 10 × 4 cm with a lymphoid and histiocytic infiltration.

In post-operative unit, we had a positive outcome that lead to the discontinuation of the mechanical ventilation and amines starting from day 1. During interaction with patient, patient stated she travelled four months ago to Mali and Guinea Conakry and was not compliant with the anti-malaria prophylaxic treatment. The diagnosis of ruptured spleen as a complication of malaria is then considered.

The presence of Plasmodium ovale (Fig. 3) was confirmed on the blood smear tests (MGG staining) and (GIEMSA staining). A plasmodial antigenemia test (optiMAL) was positive for a non

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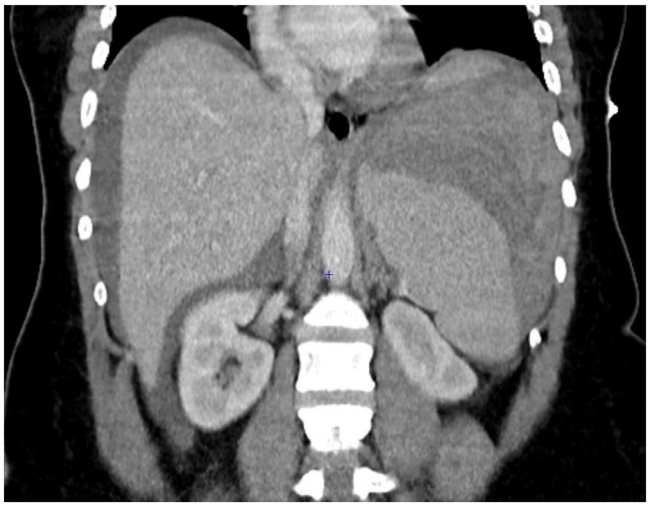

Fig. 1. Front section of abdominal scanner.

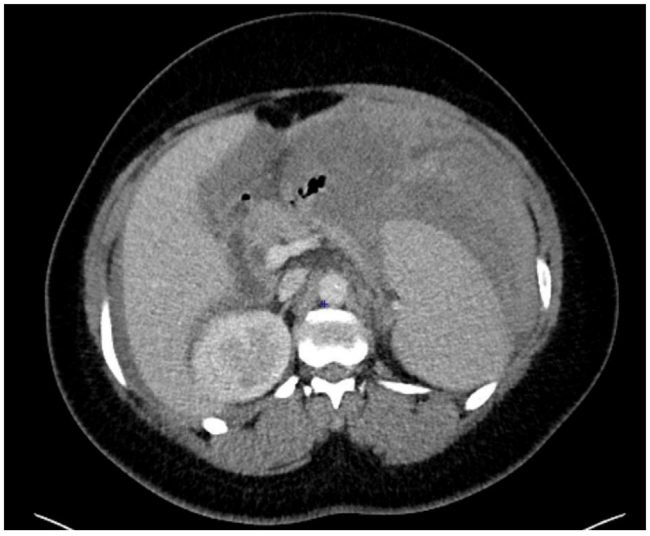

Fig. 2. Axial section of abdominal scanner.

falciparum Plasmodium. A PCR (TaqMan FAST 7500 real-time PCR) was also done and the result confirmed the Plasmodium ovale infestation. The findings of a parasitaemia prior to transfusion indicated a parasitaemia of 3%.

An initial parenteral antimalarial treatment with Quinine was started for 5 days (loading dose of 16 mg / kg; then 8 mg / kg every 8 h). The treatment was continued with Nivaquine per os for total treatment duration of 7 days, (ending on 11/19). No metabolic or cardiac complications were observed during treatment.

With a favorable outcome, the patient was transferred from post operative unit to internal medicine and infectious disease unit on Day 5. During her stay in the infectious diseases unit, the patient benefited from a schizonticidal tissue treatment with PRIMAQUINE for 15 days (from 20/11/15 to 03/12/2015) after a pre-therapeutic assessment eliminating a G6PD deficiency (G6PD dosage at 21.1U/gH).

Regarding the splenectomy, the patient received treatment with ORACILLIN 2 million Units started on 15/11/16 for a period of 2 years, with the end of treatment scheduled for 15/11/17.

On D15 following the splenectomy On 11/26/15), patient received vaccines against pneumococcus (Prevenar 13 then Pneumo 23 at 2 months), meningococcus (Meningo ACW135 conjugate vaccine) and Hemophilus B.

The physical assessment and laboratory tests during follow up care showed a cure of malaria with absence of parasites in the blood on Day 3, 7 and 28.

Similarly, the follow-up care for up to 8 months for acute episode did not show any signs of splenectomy-related immunosuppression infection.

It should also be noted that during hospital stay in infectious disease unit in November 2015, patient developed an acute respiratory failure with shunt-like effect. A chest CT with contrast showed a bilateral interstitial syndrome with no evidence of pulmonary embolism. Thereafter the patient's condition progressed to a chronic respiratory failure requiring frequent follow ups with pulmonologists. A comprehensive assessment and diagnostic testings were performed to identify the cause. (Including Autoimmune assessment, several CT scans with contrast, several LBA fibroscopy with anatomopathological, microbiological analysis; Lung biopsy and anatomo-pathological analysis). The results confirmed the diagnosis of interstitial lymphocytic pneumonitis type LIP.

This rare case of pulmonary pathology is sometimes associated with immunosuppression and autoimmunity, but up to date; no link has been demonstrated between LIP and malaria, specifically with Plasmodium ovale, even if there is a relationship between malaria and autoimmunity.

Discussion

Spontaneous rupture of the spleen is a rare but potentially fatal. Among the causes of this disease, the infectious (30 %) and haematological (27 %) causes represent more than half. The infectious causes are dominated by infectious mononucleosis [8,9].

The second most infectious causes of spontaneous spleen rupture is malaria with a prevalence between 1 / 50,000 and 1 / 100,000 cases [7,8].

In the literature, the vast majority of cases of spleen rupture due to malaria are reported in patients infected with Plasmodium Falciparum and Plasmodium vivax while other species are less likely to be associated [5].

Considered close to Plasmodium vivax malaria, of which it shares the relatively benign characteristic [2], Plasmodium ovale malaria is characterized by a low parasitaemia (<1%). However, the low parasitaemia is not systematically predictive of a benign outcome [1] .

During the P. ovale infestation, the hepatocyte incubation phase is typically asymptomatic and varies from 15 days to a maximum of 4 years. The erythrocyte schizogony lasts 48 h, causing an onset of benign fever with a peak on D1, D3, D5.

The presentation includes a phase of violent chills for 1 h with an increase in the spleen volume followed by a phase of high fever for 4 h and finally a phase of profuse sweats for 4 h. At a distance, only P. vivax and P. ovale can have reviviscence attacks linked to the persistence of plasmodium in the form of hypnozoite (quiescent hepatic parasitic stages), which can thus lead to late relapses up to 5 years.

The spleen is an essential organ of defense against malaria. Different reports of splenomegaly can be found in malaria: splenomegaly during a primary invasion, during a bout of reviviscence, during evolving visceral malaria (PVE), or in the context of a splenomegaly with hyper-IgM.

The mechanisms involved in splenic rupture are varied: sudden splenic congestion due to engorgement of the sinusoids by red blood cells in the presence of young trophozoites, subcapsular hematoma of the spleen, torsion of the pedicle (more common in evolving visceral malaria), intraparenchymal hypertension

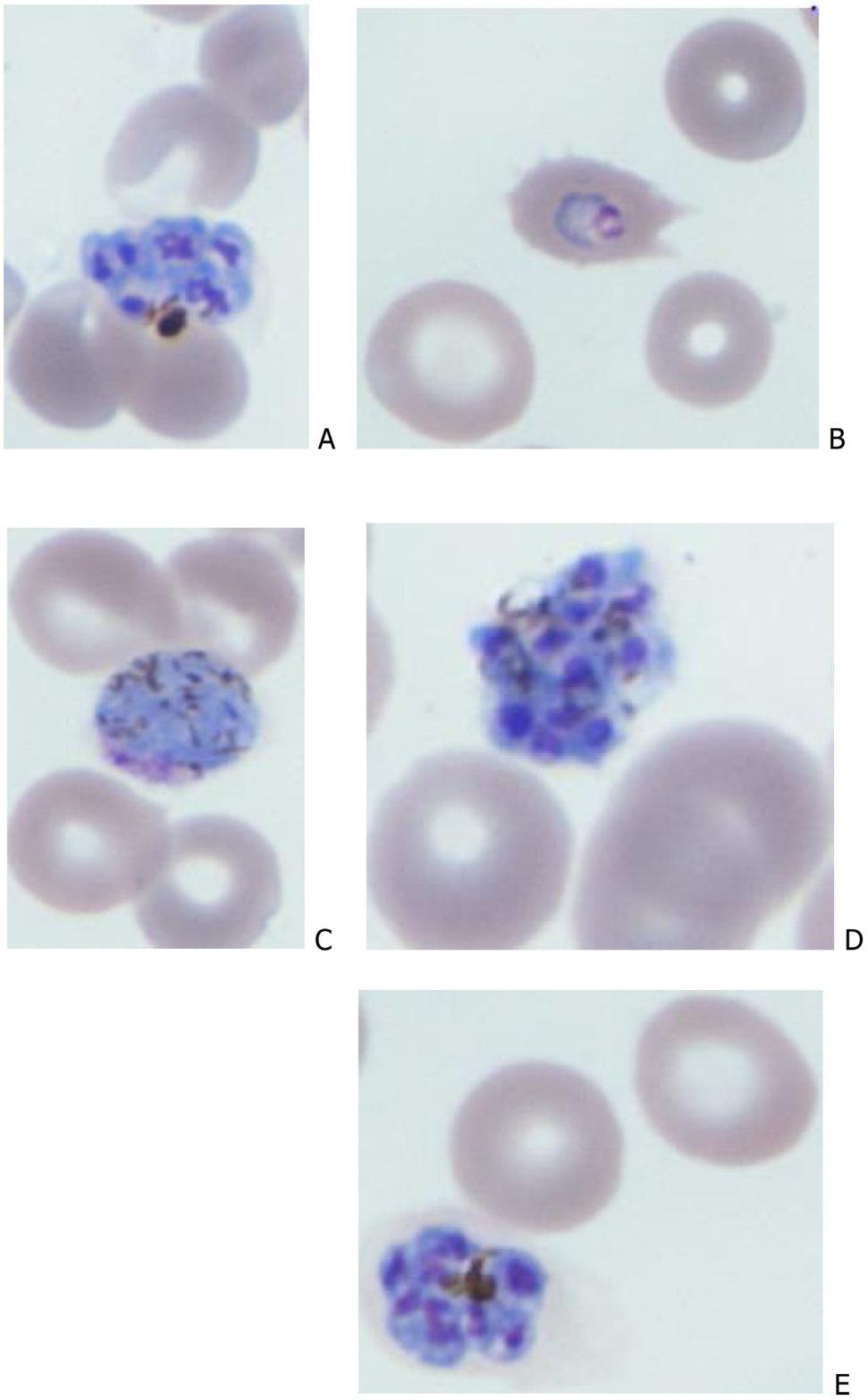

Fig. 3. Different stages of plasmodium: Trophozoites (B), Schizonts (A, D, E), gametocytes (C).

associated with fibrosis phenomena, microcirculation disorders by parasitized red blood cells responsible for local disseminated intravascular coagulation [3,9].

The histological aspect of the patient's spleen suggests for an evolving visceral malaria with dark fibro-congestive splenomegaly, lymphoid and histiocytic hyperplasia and rare parasites. A serology finds an IgG / IgM rate allowing to eliminate a splenomegaly with hyper-IgM.

The first case of Plasmodium ovale splenic rupture described in the literature dates back to October 1991 described by Facer [4]; Complicated cases of splenic rupture malaria are mainly described with Plasmodium Falciparum and Plasmodium Vivax species [5].

The main differential infectious diagnoses were investigated and eliminated with negative CMV, HIV1-2, HBV, HCV, syphilis, leptospirosis serology, a negative EBV serology indicating an old infection (EBNA positive, VCA IgM and IgM negative), negative, co-cultures (salmonella, shigella, campylobacter, Yersinia, SAMS), 5-day sterile blood cultures, a Legionella pneumophila antigenuria negative antigen.

On the therapeutic, treatment with injectable Artesunate (MALACEF® 60 mg) is frequently used; the artesunate is a hemisynthetic derivative of artemisinin. It is a fast-acting drug that is more effective and less toxic than intravenous quinine for the treatment of severe malaria. However, it has a Marketing Authorization (MA) in China only. In France, nominal tAus (temporary Authorization for use) are intended for the treatment of patients with severe P. falciparum malaria [6].

In addition, P. Ovale remains sensitive to all anti-malarial drugs, in particular chloroquine.

Conclusion

The severity of malaria is often secondary to the acute form caused by Plasmodium falciparum. However, chronic forms with splenomegaly can lead to serious hemorrhagic complications with a very life threatening condition during acute phase.

Author agreement statement

We the undersigned declare that this manuscript is original, has not been published before and is not currently being considered for publication elsewhere.

We confirm that the manuscript has been read and approved by all named authors and that there are no other persons who satisfied the criteria for authorship but are not listed. We further confirm that the order of authors listed in the manuscript has been approved by all of us.

We understand that the Corresponding Author is the sole contact for the Editorial process. He/she is responsible for communicating with the other authors about progress, submissions of revisions and final approval of proofs.

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Declaration of Competing Interest

No reported conflicts for all authors.

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