

First Case Report of Louse – Borne Relapsing Fever in Saudi Arabia

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Relapsing fever is endemic in some parts of the world such as Africa, South America, and Far East.¹⁻³ Epidemics have occurred among refugees and war victims in some parts of Africa.^{4,5} There is very little information about this infection in Saudi Arabia. The prevalence or incidence in Saudi Arabia is unknown. Its apparent rarity may be due to under diagnosis and under reporting. A high index of suspicion in appropriate clinical situations will lead to its early recognition and treatment. Relapsing fever is an acute febrile illness caused by blood spirochetes belonging to genus borrelia. This spirochete can be acquired through the bite of an infected soft tick (*Ornithodoros* species) or contact with the hemolymph of an infected human body louse (*Pediculus humanus*) and thus constituting two types of relapsing fever (RF) depending upon the responsible vector i.e. Tick – borne relapsing fever (TBRF) and Louse – borne relapsing fever (LBRF). It is characterized by recurrent cycles of pyrexia which are separated by intervals of apparent recovery. During pyrexia, organisms can be found in blood and that is the only way of reliable diagnosis in many situations.^{6,7} While during afebrile period, organisms hide themselves in spleen, liver and even central nervous system.

Case Report

An Ethiopian of 40 years was brought in unconscious state by Red Crescent society ambulance in emergency of King Abdul Aziz Hospital, Makkah on 17th Sept. 2002. On physical examination he looked jaundiced, dehydrated, his liver and spleen were enlarged. His BP 80 / 60 mm Hg, pulse 100 / min, temperature 39 c. His blood chemistry was urea 169 mg/dl, glucose 150 mg/dl, LDH 650 i.u, AST 87, ALT 47, ALP 137, Bilirubin 2.5 mg/dl, uric acid 10.4 mg/dl, K 3.4 mmol/l; coagulation tests PT, APTT were normal. His CBC showed WBC $10.7 \times 10^9/L$, Hb 10.5 g/dl, Platelet count $35 \times 10^9/l$. DLC was normal with 76% neutrophils. While looking for his low platelet count we found his blood film containing multiple number of spiral shaped organisms – borrelia recurrentis (Fig. 1). Later on requests of malaria parasite were negative and CSF was normal. His HIV, HCV, HBsAg were negative. He was started injectable Penicillin and his later samples did not reveal spirochete. He

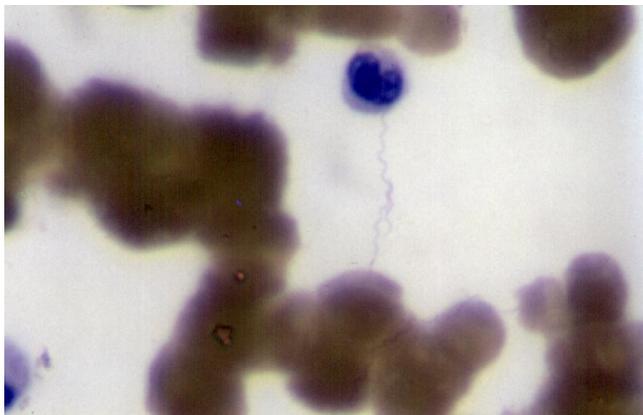


Fig. 1: Two borrelia organisms and a monocyte are seen (x100).

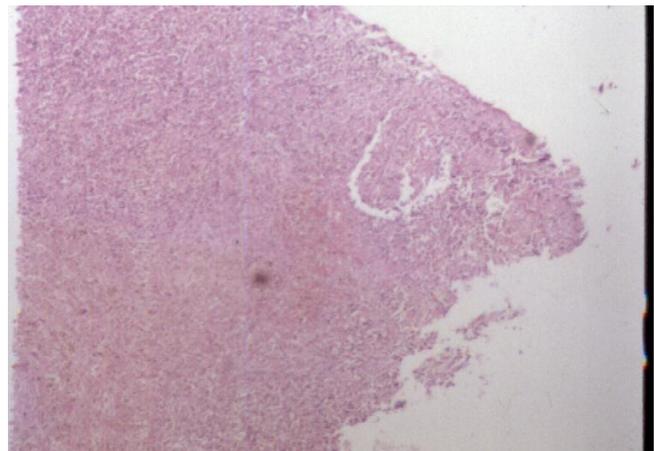


Fig. 2: Splenectomy specimen showing capsular rupture (x4).

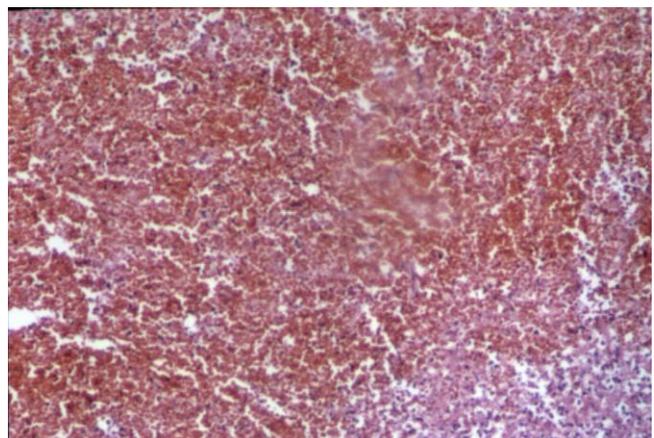


Fig. 3: Splenic tissue showing areas of hemorrhage in red pulp(x40).

had signs of splenic rupture and after ultrasound and surgical advice; he underwent splenectomy. Histopathology of spleen showed splenic rupture with distortion of architecture; wide areas of necrosis, hemorrhage, and hemorrhagic infarcts being infiltrated by lymphoplasmacytic cells, histiocytes, fibroblasts and neutrophils (Fig. 2, 3). Warthin starry stain for spirochetes showed spiral shaped organisms.

After splenectomy he stayed in hospital for 10 days and he was discharged in a stable condition.

Discussion

Tick – borne or louse-borne borreliosis can be a rare cause of febrile illness in areas other than Sudan, Ethiopia, Africa, and North America. It remains an important and tricky cause of recurrent fever in patients of such epidemic and endemic areas. It is responsible for morbidity and mortality especially in young adults and pregnant women.⁸⁻¹⁰ Definitive diagnosis is usually made by the demonstration of *Borrelia* organisms in the peripheral blood smears stained with Romanowsky stains.^{6,7} Among the other essentials of diagnosis is the presentation as fever with rash, headache, shaking chills, myalgias, arthralgias and during the acute phase – hepatosplenomegaly.¹¹ The diagnosis in our case was accidental as we found spirochetes while trying to validate the slide for thrombocytopenia. As patient belonged to Ethiopia where louse – borne relapsing fever is common and one single shot of Penicillin cleared the spirochetes from blood stream and no second sample contained this organism. As for as diagnosis was concerned there was no doubt about LBRF as animal inoculation, bacteriological culture media i.e. Kelly's medium and serological tests were not available. Because it harbours plasmid and during afebrile period *borrelia* changes its antigenic structure and thus serologic tests have not been developed even in epidemic areas of disease. Our patient showed complication of splenic rupture which was removed and biopsy results further confirmed the diagnosis.

As this patient was diagnosed in Makkah; a place of pilgrimage and umrah, so proper precautions exercised by infection control department regarding isolation and thus nobody acquired the disease and any unfavourable outbreak avoided as louse borne disease could have caused a great problem in Makkah due to overcrowding during pilgrimage and umrah rituals.

The incidence or prevalence of RF in Saudi Arabia is not known. However two cases of Tick-borne relapsing fever have been reported from King Khalid University hospital, Riyadh in 1995. Our case appears to be a first case of Louse-borne relapsing fever (LBRF) in Saudi Arabia.

Spirochetes can often be easily detected in Giemsa stained peripheral blood smears during febrile phase in suspected cases but identification of species is difficult as tick – borne RF is concerned.¹² Recent attempts of isolation and culture of spirochetes has been made^{13,14} but these methods do not identify specific type of the spirochete.¹² The applications of relatively new techniques such as DNA hybridi-

zation probe and polymerase chain reaction (PCR) has been shown to be able to analyze molecular and genetic determinants.¹⁵⁻¹⁸ Monoclonal antibodies have not been developed due to changes in antigenic structure of spirochete, induced by plasmid. Thus clinical history, epidemiologic data and detection of spirochetes in blood smear during febrile phase are the mainstay of diagnosis.

It was concluded in Al – Gwaiz et al's study that sporadic cases of *Borrelia* infections occurred in Saudi Arabia with strengthening evidence regarding the isolation of a third – instar nymph *Ornithodoros* from an indigenous goat in Makkah area of Saudi Arabia and suggested that parasite may be widely distributed in Saudi Arabia.¹⁹⁻²⁰ As more than 5 million people visit this holy place for pilgrimage and umrah during the year so physicians of this area should especially investigate and suspect for *Borrelia* while looking for origin of fever and also with special preference for those coming from diseased areas or having history of travel plus normal WBC or slightly leukopenia and thrombocytopenia. A lice infestation of 14% has been reported from pediatric age group 2-10 years so pediatricians need to be on guard about this LBRF although a rare disease.²¹ A prospective study of a larger number of cases with febrile illness may provide a better information regarding the prevalence or incidence in the Saudi population. The findings of a survey in West Africa regarding borreliosis are good example in which common occurrence was seen in Senegal.²²

References

1. Dworkin MS, Shoemaker PC, Fritz CL, et al. The epidemiology of tick – bone relapsing fever in the United States. *AM J Trop Med Hyg* 2002 Jan; 66 (6): 753-8.
2. Paul WS, Maupin G, Scote-Weight AO, et al. Outbreak of tick – bone relapsing fever at the north rim of the Grand Canyon: evidence for effectiveness of preventive measures. *Am J Trop Med Hyg* 2002 Jan; 66 (1): 71-5.
3. Dworkin MS, Schawan TG, Anderson DE Jr. Tick – bone relapsing fever in North America. *Med Clin North Am* 2002 Mar; 86 (2): 417-33, viii-ix.
4. Bryceson ADM, Parry EHO, Perine PL et al. Louse-borne relapsing fever. *Q J Med* 1970; 29: 129-70.
5. Perine PL, Reynolds DF. Relapsing fever epidemic in the Sudan and Ethiopia. *Lancet* 1974; 2: 1324-5.
6. Cobey FC, Goldbarg SH, Levine RA, Patton CL. Short report: Detection of *Borrelia* (relapsing fever) in rural Ethiopia by means of the quantitative buffy coat technique. *Am J Trop Med Hyg* 2001 Aug; 65 (2): 164-5.
7. Beck MD. Laboratory Procedure. In: Relapsing fever. Special Bulletin No.61. California State Department of Public Health, Sacramento 1936: 19-23.
8. Ahmad MAM, Abdul Wahab SM, Abdul Malik MO et al. Louse – borne relapsing fever in the Sudan. A historical review and clinicopathological study. *Trop Geogr Med* 1980; 32: 106-11.

9. Salih SY, Mustapha D, Abdul Wahab MS et al. Louse – borne relapsing fever : a clinical laboratory study of 363 cases in the Sudan. *Trop Med Hyg* 1977; 71: 43-8.
10. Melkert PW. Relapsing fever in pregnancy: analysis of high risk factors. *Br J Obstet Gynecol* 1988; 95: 1070-2.
11. Chaparro S, Montoya JG. *Borrelia* and *Leptospira* species. In: *Current Diagnosis and Treatment in INFECTIOUS DISEASES*. Lange Medical Books / Mc Graw-Hill Medical Publishing Division 2001; 680-81.
12. Schwan TG, Gage KL, Karsters RH et al. Identification of tick – borne relapsing fever spirochete *Borrelia hermsii* by using a species – specific monoclonal antibody. *J Clin Microbiol* 1992; 30: 790-5.
13. Kelly R. Cultivation of *Borrelia hermsii*. *Science* 1971; 173: 443-4.
14. Barbour AG. Isolation and cultivation of Lyme disease spirochetes. *Yale J Biol Med* 1984; 57: 521-5.
15. Picken RN. Polymerase chain reaction primers and probes derived from flagellin gene sequences for specific detection of the agents of Lyme disease and North American relapsing fever. *J Clin Microbiol* 1992; 30: 99-114.
16. Schwan IG, Simpson WJ, Schrumph ME et al. Identification of *Borrelia burgdorferi* and *Borrelia hermsii* using DNA hybridization probes. *J Clin Microbiol* 1989; 27: 1734-8.
17. Vidal V, Cutler S, Scragg IG. Characterization of silent and active genes for a variable large protein of *Borrelia recurrentis*. *BMC Infect Dis* 2002Oct 14; 2 (1): 25.
18. Takahashi Y, Cutler SJ, Fukunaga M. Size conversion of a linear plasmid in the relapsing fever agent *Borrelia duttonii*. *Microbiol Immunol* 2000; 44 (12): 1071-4.
19. Hoogstraal H, Wassef HY, Diab FM et al. *Ornithodoros (Aluronasus) laborensis* in Saudi Arabia; biological, veterinary and medical implications. *Fauna of Saudi Arabia* 1984; 6: 165-9.
20. Al – Gwaiz LA, Al – Mashhadani SA, Ayoola EA et al. Relapsing fever in Saudi Arabia Report of two cases. *Ann Saudi Med* 1995 Mar; 15 (2): 165-7.
21. Boyle P. Pilot study of the prevalence of head lice infestation in a population of Saudi Arabian children. *Fam Prac* 1987 June; 4 (2): 138-42.
22. Trape JF, Duplantier JM, Bouganali H, et al. Tick-borne borreliosis in West Africa. *Lancet* 1991; 337: 473-5.