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# Visceral Leishmaniasis with Breast Involvement in the Pediatric Population

**Saeed Al Hindi[1], Husain Al Aradi[1], Noor Al Hashimi[2*] and Mohamed Mubarak[1]**

*[1]Department of Surgery, Salmaniya Medical Complex, P.O.Box 12, Manama, Kingdom of Bahrain.*
*[2]Department of Pediatrics, Salmaniya Medical Complex, P.O.Box 12, Manama, Kingdom of Bahrain.*

***Authors' contributions***

*This work was carried out in collaboration among all authors. All authors read and approved the final manuscript as per the ICMJE recommendations.*

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***Case Report***

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## ABSTRACT

**Aims:** The aim of the paper is to acknowledge the atypical presentation of Visceral Leishmaniasis (VL) and subsequently tailor the management based on the patient's characteristics and clinical context, while comparing it to standard practice.
**Case Presentation:** We report a case of VL in a nine-year old Bahraini girl, presenting with a painful breast cyst. Ultrasound guided fine need aspiration of the cysts revealed the leishmania amastigotes. The patient was treated with complete cyst aspiration and received no further treatment. The patient did well, with no signs of recurrence or disease in the follow-up period.
**Discussion:** The diagnostic gold standard of VL is direct visualization of the protozoans in specimens acquired from tissue biopsies or aspirations, as is the case with our patient. The first line treatment for VL is intravenous pentavalent antimonials, such as as meglumine antimoniate. However, due to the immunocompetent state of our patient and the asymptomatic presentation, the only treatment was therapeutic aspiration of the cyst.
**Conclusion:** Ultrasound guided aspiration of the cyst, with regular follow up with breast ultrasound, proved to be an effective method of managing visceral leishmaniasis in our patient.

**Corresponding author: E-mail: noor5alhashimi@gmail.com;*

*Keywords: Atypical presentation; benign breast mass; parasitic infection; therapeutic aspiration.*

## 1. INTRODUCTION

Leishmaniasis is a parasitic infection caused by the intracellular protozoan parasite Leishmania. The female sand-fly, is the only vector responsible for mediating transmission of the parasite [1].

The clinical features of Leishmaniasis can be broadly divided into three categories: cutaneous leishmaniasis (CL), mucocutaneous leishmaniasis (ML), and visceral leischmaniasis (VL).

The worldwide prevalence of these manifestations varies geographically, depending on the type of Leishmania species, the reservoir, and the vector. [Table 1] Kumar [2] Generally, the majority of the disease burden falls on poorer countries in tropical and subtropical regions, with an annual incidence of 1.5 to 2 million cases [‘1]. However, due to under-reporting in poorer countries, as a result of deficient epidemiological surveillance systems and diagnostic methods, these numbers are merely an estimate [3].

The focus of this paper is visceral leishmaniasis (VL), also known as Kala – Azar “Black fever”.

The manifestation of VL ranges from asymptomatic infection, to subclinical, to clinically overt and potentially life-threatening disease.

**Table 1. Epidemiology of Leishmania species [2]**

| Organism | Infection | Geography | Reservoir | Vector |
|---|---|---|---|---|
| L. donovani | Visceral leishmaniasis | North-east India, Bangladesh, Burma | Humans | Phebotomus argentipes |
| L. infantum | Visceral leishmaniasis | Mediterranean basin, Middle east, China, Asia | Dogs, foxes, jackals | P. pcrniciosufi, P. arias |
| L. donovani | Visceral leishmaniasis | Sudan, Kenya, Horn of Africa | Rodents in Sudan, Canines, Humans | P. orinntalis, P. martini |
| L. major | Cutaneous leishmaniasis | Semideserts in Middle East, North India, Pakistan | Gerbils | P. papatassi |
| L. major | Cutaneous leishmaniasis | Sub-Sahara, Sudan | Rodents | P. duboscqi |
| L. tropica | Cutaneous leishmaniasis | Middle East, Mediterranean basin, central Asia | Humans | P. sergenti |
| L. aethiopica | Cutaneous leishmaniasis | Highlands of Kenya, Ethiopia | Hyraxes | P. longipes, P. pedifer |
| L. chagasi | Visceral leishmaniasis | Central, Northern South America, Brazil, Venezuela | Foxes, dogs, opossums | Luizomyia longipalpis |
| L. mexicana | Cutaneous leishmaniasis | Yucatan, Guatemala | Forest rodents | L. olmeca |
| L. amazonensis | Cutaneous leishmaniasis | Tropical forests of South America | Forest rodents | L. flaviscutellata |
| L. braziliensis | Mucocutaneous leishmaniasis | Tropical forests of South and Central America | Forest rodents, peridomestic animals | Psychodopygus Lutzomyia spp. |
| L. guyanemis | Mucocutaneous leishmaniasis | Guyana, Surinam | Sloths arboreal anteaters | L. umbratilis |
| L. panamensis | Mucocutaneous leishmaniasis | Panama, Costa Rica, Colombia | Sloths | L. trapidoictal |
| L. peruviana | Mucocutaneous leishmaniasis | West Andes of Peru, Argentine highlands | Dogs | L. verrucarurn, L. pvmenis |

Classic features of VL include persistent fever, anorexia, weight loss, hepatosplenomegaly, lymphadenopathy, and features of pancytopenia [2]. The typically involved organs guide the diagnostic methods which include liver biopsy and aspiration biopsies from the spleen, lymph node, and bone marrow. Atypical presentation of VL, such as portal hypertension or gastrointestinal symptoms, has been reported in the literature [4]. However, to the best of our knowledge, there have only been two other reported cases of breast involvement in VL, both of which occurred in the adult population [5]. We report a case of VL with breast involvement in the pediatric population.

## 2. CASE PRESENTATION

A nine-year-old Bahraini girl, not a known case of any medical illness, presented to the pediatric surgery clinic with complaints of a painful right breast mass. Clinical evaluation revealed the following:

A one week history of a painful mass in the right breast. The pain was described as being mild, dull in nature, continuous, and no alleviating or exacerbating factors were noted.

The mass was not associated with any overlying skin changes, and was constant in size and character. There was a history of recent travel to Iran within the last 4 months. No history of fever, recent illness, constitutional symptoms or trauma. No family history of breast conditions or malignancies. Systems review was negative for any symptoms.

On systematic examination of the right breast, the mass was tender, and there was no overlying skin or nipple changes. A small mobile mass, measuring approximately 2 x 2 cm, with regular borders and a smooth surface, was noted in the lower outer quadrant of the right breast. On examining the left breast, a similar sized mobile mass was noted in the upper inner quadrant of the breast. Similarly, there was no overlying skin or nipple changes. A small lymph node measuring around 1-2 cm was noted in the right axilla. There was no axillary lymphadenopathy noted on the left side. A systematic examination of the cardiovascular, respiratory, abdominal, genitourinary and neurological system was unremarkable.

Imaging of the breasts was done using ultrasound and reported the masses to be cysts with calcifications. As this finding is not usual of normal breast development, it necessitated the use of fine needle aspiration, under ultrasound guidance, to determine the nature of the mass.

The fine needle aspiration smear revealed the leishmania amastigotes, thus reaching the diagnosis of VL. [Fig. 1]

Post aspiration of the cyst in both the right and the left breast, another ultrasound scan was performed and reported the following: “Bilateral breast ultrasound was performed. There is evidence of altered nodular echotexture of the right breast with appearance of a focal area of low echogenicity seen at the 7 to 8 o’clock position which incidentally is the site of the previous cysts aspiration. No evidence of residual cyst seen. However, the hypodense areas may be related to edema or probable residual changes. Evidence of small lymph-node is seen at the right axilla, this measures about 0.6 x 0.5 cm.” Note that the size of the lymph node is small and insignificant, and disappeared on follow up.

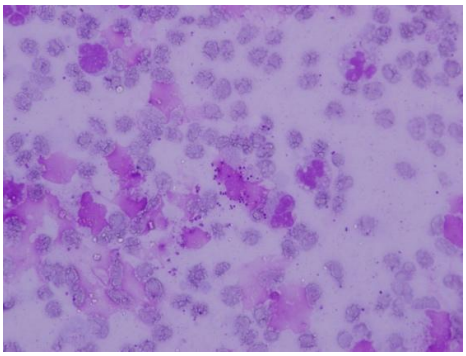

**Fig. 1. Aspiration smear showing Leishmania amastigotes**

At the left breast, similar nodular appearance is seen however there is a bi-septated fluid filled cysts seen at 9 o’clock to 12 o’clock at the left breast in zone B to C. This fluid filled cystic lesion measures about 2 x 1 cm and shows a thin walled with evidence of tiny specks of calcification at its floor. Otherwise, no evidence of debris seen within the cyst.

Routine blood investigations, including liver function tests, renal functional tests, and coagulation profile were normal. No serological tests for leishmaniasis were performed.

The patient was treated by aspirating the cysts in both breasts, under ultrasound guidance, without

receiving further treatment for the leishmaniasis. The patient was followed up for a period of two years, with regular ultrasound scans of the breast, with no signs of recurrence or clinical features indicative of VL.

## 3. DISCUSSION

Clinical features of VL depend on the interplay between the leishmania species and the immune system of the host. In the case of our immunocompetent patient, the infection was asymptomatic with an atypical presentation of a breast lump. Breast complaints in the pediatric population are, for the most part, of benign etiology. With the majority of findings reflecting physiological changes. Hence, the importance of understanding the different stages of normal breast development in children and adolescence. If clinical evaluation is not sufficient, further investigations should include a breast ultrasound. In our patient, ultrasound of the breast lesion revealed a cystic mass with calcification, which deviated from the picture of physiological breast development. Thus, necessitating fine needle aspiration of the cyst to determine its nature, which led to the diagnosis [6].

The diagnostic gold standard is microscopic visualization of the protozoans in clinical specimens acquired from tissue aspirations or biopsies. The most frequently acquired tissue sample is that of the bone marrow, via sternum aspiration, with a sensitivity of 65–80%. This is in comparison to splenic aspirations which provide the most sensitive sample (95%) but carry the risk of splenic rupture and hemorrhage, especially in children with thrombocytopenia [2,3]. If tissue aspiration or biopsy is not feasible, laboratory based serological assays, such as the direct agglutination tests, for serum anti-*Leishmania* IgG and anti-K39 antibodies can also be used. These serological tests have a high sensitivity and specificity, but they do not differentiate between current and past infection. If the above two mentioned methods are inconclusive, or the parasitic load is low such as in HIV co-infection, the use of molecular techniques such as PCR targeting sequences within the Leishmania gene, might be diagnostic [2]. The prevalence of leishmaniasis in poorly resourced areas necessitates the need for a diagnostic method that is reliable and cost effective. A new diagnostic approach relies on the detection of leishmanial antigens in urine samples [3]. This method is rapid, non-invasive, and has a high specificity (95%), making it a good option for poorly resourced areas. However, its low sensitivity (57%) may require the need for additional diagnostic tests [7].

Treatment of leishmaniasis should be tailored to the characteristics of the patient (e.g. immunological status), the infecting leishmania species (e.g. virulence) and the clinical manifestations [8].

The first line treatment is intravenous pentavalent antimonials, such as meglumine antimoniate and sodium stibogluconate. Antimonials achieve satisfactory clinical and microbiological outcomes within 2 weeks [1]. However, their use is limited by their side effects and the resistance patterns of different infecting species. Antimonial side-effects include injection site reaction, nausea, vomiting, myalgia, arthralgia, derangements in liver and renal function tests, and a variety of electrocardiographic (ECG) alterations. In the instances where resistance is encountered, Amphotericin B, a more potent yet toxic drug can be used [9].

Treatment outcomes depend heavily on the immunological status of the patients. Immunocompromised patients, with a low CD4+ count, such as those with HIV co-infection, exhibit a lower response to treatment and higher chances of recurrences or relapse within 6-12 months of treatment [1]. In the case of our patient, the treatment was ultrasound guided aspiration of the breast mass, without the use of medications to treat the infection. The rationale behind this individualized approach was based on the fact that the patient was immunocompetent and the infection was asymptomatic with normal laboratory findings. A similar management approach concerning an atypical presentation of leishmaniasis as an adrenal cystic mass, was reported in literature. Where an immunocompetent patient had surgical removal of the leishmaniasis cyst, with no further treatment, and was reported to have recovered well after the surgery, with no signs of VL at follow up [10].

## 4. CONCLUSION

In conclusion, VL in our patient atypically manifested as breast cysts with calcification, with no other clinical features. Due to the clinical context and immunological status of our patient, the tailored treatment approach was ultrasound guidance aspiration of the cysts, with regular

follow-up to monitor recurrences and complications.

## CONSENT

All authors declare that informed consent was obtained from the patient and the parents for publication of this case report and that maximal patient anonymity was ensured and maintained during the writing of the case report.

## ETHICAL APPROVAL

As per international standard or university standard written ethical approval has been collected and preserved by the author(s).

## COMPETING INTERESTS

Authors have declared that no competing interests exist.

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