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Testicular schistosomiasis: a clinico-pathological presentation of three pediatric cases and review of literature

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Abstract

Schistosomiasis is an important communicable disease in developing countries and an important cause of morbidity in the tropics and subtropics. It is a very rare cause of painless testicular swelling that tend to mimic malignancy and cause a diagnostic dilemma. Though histopathologic analysis confirms diagnosis, a high index of suspicion is required especially for patients presenting from endemic areas. We aim to present the clinicopathological features of three pediatric cases of Testicular Schistosomiasis (TS) and to highlight the role of high index of suspicion, incisional biopsy and histopathologic diagnosis in prevention of unwarranted orchiectomy and preservation of testicular function. We report three cases of TS in patients aged 5 years, 12 years and 14 years, who presented with progressive testicular swellings. The diagnoses were made by histopathological analysis and the patients were treated with oral praziquantel. TS should always be considered an important differential in all male children and adults presenting with testicular masses especially when they live in endemic regions. Incisional biopsy, proper histopathologic evaluation and treatment could prevent unwarranted orchiectomy and thus preserve testicular function.

Introduction

Schistosomiasis is an important communicable disease in developing countries[1,2] and an important cause of morbidity in the tropics and subtropics,[3] occurring mainly in poor communities without access to safe drinking water and adequate sanitation.[2,4] Sub-Saharan Africa (SSA) is estimated to contribute 93% of the world's 207 million cases of human schistosomiasis, with the largest numbers occurring in Nigeria (29 million) followed by Tanzania (19 million), Democratic Republic of Congo and Ghana (15 million each).[4] It is also a very rare cause of testicular swelling, affecting the scrotum and seminal vesicles, resulting in scrotal pain, dermatitis, and hydrocele, and simulating testicular neoplasm.[2,3] The most common parasites infecting men are *Schistosoma mansoni*, *Schistosoma japonicum* and *Schistosoma hematobium*.[3,5] While *S. mansoni* commonly causes fibrosis in the liver and lungs, leading to portal and pulmonary hypertensions, respectively, *S. hematobium* mainly cause chronic cystitis, squamous metaplasia, and subsequently squamous cell carcinoma.[6] The involvement of unusual sites such as the appendix, ovary, prostate, and cervix have also been reported.[6] However, a prudent search of literature shows only few reported cases of Testicular Schistosomiasis (TS) in Nigeria. We report our experience with three pediatric cases of TS, diagnosed and managed at our center and also review relevant literature.

Case 1

A 7-year-old male presented with an 18 months history of gradual progressive painless swelling of left scrotum. There was associated intermittent hematuria of 1 year, but no associated gastrointestinal symptoms. The hematuria was terminal and intermittent, but did not warrant blood transfusion. There was no associated weight loss, no history of undescended testis nor history of hematuria amongst the

siblings and the parents, and his source of water was from the stream. The patient was referred to our center from a peripheral hospital following a presentation of a left testicular swelling.

On examination, patient was a fully conscious child, mildly pale, anicteric and with respiratory rate 22 breaths/minute, and pulse rate of 72 beats/minute. The left hemiscrotum contained a non-tender, 4.5×2.5 cm firm, non-reducible left hemiscrotal swelling. The left testis was not distinct from the mass and the inguinal nodes were not palpable. The right hemiscrotum was unremarkable. The abdomen was full on examination, with no palpable abdominal organs, masses nor ascites. A clinical impression of testicular tumor was made during surgery (Table 1).

Laboratory investigations were: Hemoglobin (Hb) estimation, 8.5g/dL; Packed Cell Volume (PCV), 26%; Erythrocyte Sedimentation Rate (ESR), 90mm 1st hour; platelet count, 108000/mm^3; total White Cell Count (WBC), 4.5x109/L (N-60%, L-38%, E-2%); negative HIV I and 2 antibodies; Mantoux negative. The Chest X-ray (CXR) findings and Serum Electrolytes, Urea And Creatinine (SEUCr) were within normal limits and there was absence of ova of Schistosoma in urine and stool specimen (Table 2).

Patient had exploration of the left scrotal mass and had left orchidectomy. Gross examination of the orchidectomy specimen show a nodular greyish white firm to hard tissue measuring 4.5x2.5x1.5 cm. The specimen cut with gritty sensation to reveal greyish white solid and cystic surfaces. Microscopic examination shows total replacement of the normal testicular architecture by numerous non-caseating granulomas around viable and degenerating Schistosoma eggs, some of which are calcified. There is also intense eosinophilic infiltration and stromal fibrosis (Figures 1-3). A histopathological diagnosis of TS was made and the patient was treated with oral praziquantel 400 mg start. The patient had normal hemoglobin levels and conservation of the remaining testis after treatment but he was lost to follow-up after 3 months (Table 1).

Case 2

12-year-old male presented at our Pediatric Surgical Clinic [PSC] with a 3-year history of gradual, progressive left testicular multinodular swelling. There was associated intermittent hematuria of 2 years duration, with mild weight loss, but no associated history of undescended testis nor history of hematuria amongst the siblings or the parents. Patient was domiciled at Ohaozara Local Government Area of Ebony State, Nigeria, and his source of water was stream water.

Clinical examination revealed a young, conscious and alert male, afebrile, mildly pale, anicteric and moderately dehydrated, with no pedal edema. The respiratory rate was 28 breaths/minute, and pulse rate 90 beats/minute. Physical examination demonstrated non-tender multinodular left testicular swelling and a provisional clinical diagnosis of testicular tumor was made (Table 1).

The laboratory hemoglobin estimation and PCV were 9.6 g/dL and 29%, respectively, and the erythrocyte sedimentation rate (ESR) 80 mm 1st hour. The White Blood Cell Count (WBC), Serum Electrolyte Urea and Creatinine (SEUCr), urine and stool Microscopy, Culture and Sensitivity (M/C/S) examinations were unremarkable (Table 2). The chest was clinically clear and there were normal heart sounds with no murmurs. The patient had exploration of the left testicular masses and biopsy tissues were taken from the testicular masses and sent for histopathologic evaluation. Gross examination of the biopsy showed three pieces of greyish white tissues aggregating to 1.2 cm. Microscopic examination of sections from the left testis show several granulomas containing Langhans Type Multinucleate Giant Cells surrounding ova of Schistosoma worms, with destruction of adjacent seminiferous tubules (Figures 4-6). A diagnosis of TS was made and the patient was treated with praziquantel which, on commencement, led to significant improvement and thus orchidectomy was avoided and testicular function preserved (Table 1).

Case 3

A 14-year-old male presented with a 1-year history of progressive left testicular swelling. The swelling was non-tender and there was associated history of intermittent of hematuria of 6 months duration. He had no past medical or family history of a similar illness and was fully immunized at his community health center. On examination was a fully conscious male teenager, afebrile (37.20°C), not pale, anicteric, not cyanosed nor dehydrated. The respiratory rate was 22 breaths/minute, pulse rate 82 beats/minute, and Blood Pressure (BP) 92/60 mmHg. Examination of the groin demonstrated a non-tender multinodular left testicular swelling. Examination of other systems was unremarkable. A clinical impression of testicular tumor R/o orchitis was made (Table 1). Laboratory investigations were: Hb, 10.3 g/dL; PCV, 31%; total WBC, 8.3x109/L (N-62%, L-36%, E-2%); ESR, 100 mm 1st hour; negative to HIV I and 2 antibodies; negative Mantoux test and the SEUCr were within normal limits (Table 2). Left orchidectomy was done and sample was sent for histopathology. Gross examination of the biopsy specimen shows a nodular tissue mass measuring 6.0x5.0x2.0 cm. Microscopic examination of the testicular biopsy tissue show testicular tissue with several granulomas around Schistosoma ova, some viable and some degenerating and calcifying. Within the blood vessels are adult pairs wrapped in the copulatory embrace with the slender female adult lying within the gynaecophoric groove of the male with surrounding ova of Schistosoma worms (Figures 7-8). A histopathological diagnosis of TS was made and the patient was subsequently treated with praziquantel 400 mg start. On a follow-up visit 2 months later, patient was stable and the previously palpable inguinal nodes were no longer palpable (Table 1).

Discussion

Schistosomiasis is an important communicable disease occurring mainly in tropical and subtropical regions, and especially in poor communities without access to safe drinking water and adequate sanitation.[1-3] It is estimated that at least 93% of the world's disease burden and those that require treatment, live in SSA[4] Testicular involvement by this disease is rare and only few cases have been reported in our environment,[2] making it pertinent for us to report these three cases discovered over the last eight years.

All the three cases we present in this report are within the age range of 7-14 years, and presented with painless unilateral testicular nodules. These findings are in tandem with reports from previous studies in our environment,[2] other regions in Nigeria,[3,7,8] and other countries.[9,10] It has been reported that the highest prevalence and intensities of human schistosomiasis occur in school-aged children, adolescents, and young adults who also suffer from the highest morbidity and mortality.[4]

While all previous reported cases from our environment and adjoining regions were diagnosed from orchidectomy samples,[2,3,5,7,8] one of our present cases had an incisional biopsy with histopathologic diagnosis of TS, praziquentel therapy and thus testicular preservation. This is in contrast with most cases in literature that had diagnosis after excisional biopsy (orchidectomy). This index case highlights the importance of high index of suspicion, biopsy diagnosis and treatment in preventing unwarranted orchiectomies and thus preservation of testicular function in TS.

Though noninvasive techniques such as Ultrasonography (USS) can detect schistosomiasis as hypoechoic lesions and raise the suspicion for granulomatous lesions, Magnetic Resonance Imaging (MRI) is more sensitive as it shows irregular tunica.[11] However, neither ultrasonography nor MRI is confirmatory as only biopsy and histopathologic analysis could confirm TS. Also, neither USS nor MRI was done in all three of our cases because the patients had problems of poor finance, and biopsy

and histopathologic analysis were selectively done for confirmation of diagnosis and conservation of financial resources.

There are reports of use of Frozen Section Analysis (FSA) for intra-operative analysis of suspicious testicular lesions aimed at avoidance of unnecessary orchidectomy.[12,13-15] FSA is a valuable tool assisting testicular preservation but when the lesion size is correlated with incidence of malignancy, FSA may best be used for small testicular lesions suitable for excision biopsy.[16]

In addition to the presentation of painless testicular swelling, all three of our cases lived in the endemic areas with stream as their source of water. The finding of living near rivers, lakes, and other water bodies such as dams and reservoirs, usually contaminated with snail intermediate hosts constitute high risk for schistosomiasis,[4,17] and it is in agreement with that of previous reports from our environment,[2] as well as those from other regions.[10,18] This also emphasizes the importance of considering TS as a differential diagnosis of testicular tumors, especially in patients who reside in endemic areas, and underscores the need for clean sources of water as a vital control and prevention strategy. It is also important to obtain a thorough history, to elucidate exposure to endemic areas and inform whether biopsy, and subsequent testicular preservation, may be appropriate.[19] In one of our cases, a high index of suspicion in addition to a history of significant period spent in endemic areas, influenced the decision to do an incisional biopsy rather than an orchidectomy. Histopathologic confirmation of schistosomiasis and subsequent treatment with praziquantel led to avoidance of orchidectomy and conservation of testicular function.

Our cases also had a varied duration of illness before presentation ranging from 1 to 3 years. These findings are also in keeping with the reported varied presentations of TS.[8,10,18] This could be due to poverty and inability of these patients to afford standard health care.

Although two of our patients were mildly anemic at presentation and there were no other remarkable hematological or biochemical alterations observed in all three patients, they all had normal hemoglobin levels and conservation of the remaining testicular tissue after treatment.

Schistosomiasis is a medically treatable disease and a stat dose of praziquantel of 400 mg is usually enough for treatment. All three of our cases also had praziquantel therapy, which is the mainstay for the treatment of schistosomiasis worldwide.[8]

Conclusions

TS remains a rare but important cause of testicular swelling within the pediatrics age group, presenting with unique clinicopathological features. TS should always be considered an important differential in all male children and adults presenting with testicular masses especially when they live in endemic regions. A high index of suspicion, incisional biopsy, proper histopathologic evaluation and treatment with praziquantel will prevent unwarranted orchiectomy and thus preserve testicular function.

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Figure 1. Photomicrograph showing granulomas with intense surrounding eosinophilic infiltration and fibrosis surrounding ova of Schistosoma worms (short arrows) [H & E x 100].

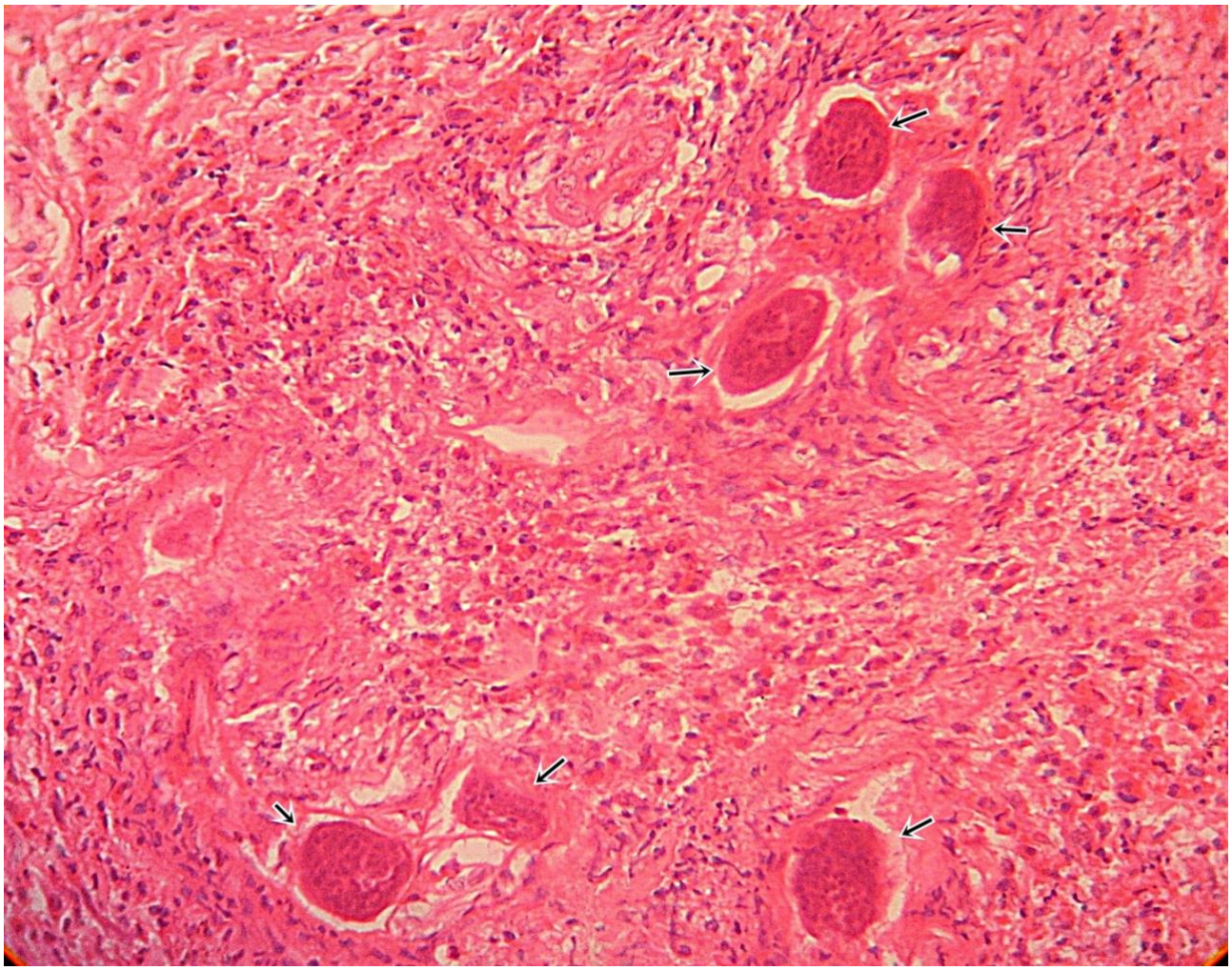

Figure 2. Photomicrograph showing granulomas containing Langhans Type Multinucleate Giant Cells (LMGC) surrounding ova of Schistosoma worms (short arrows) [H & E x 200].

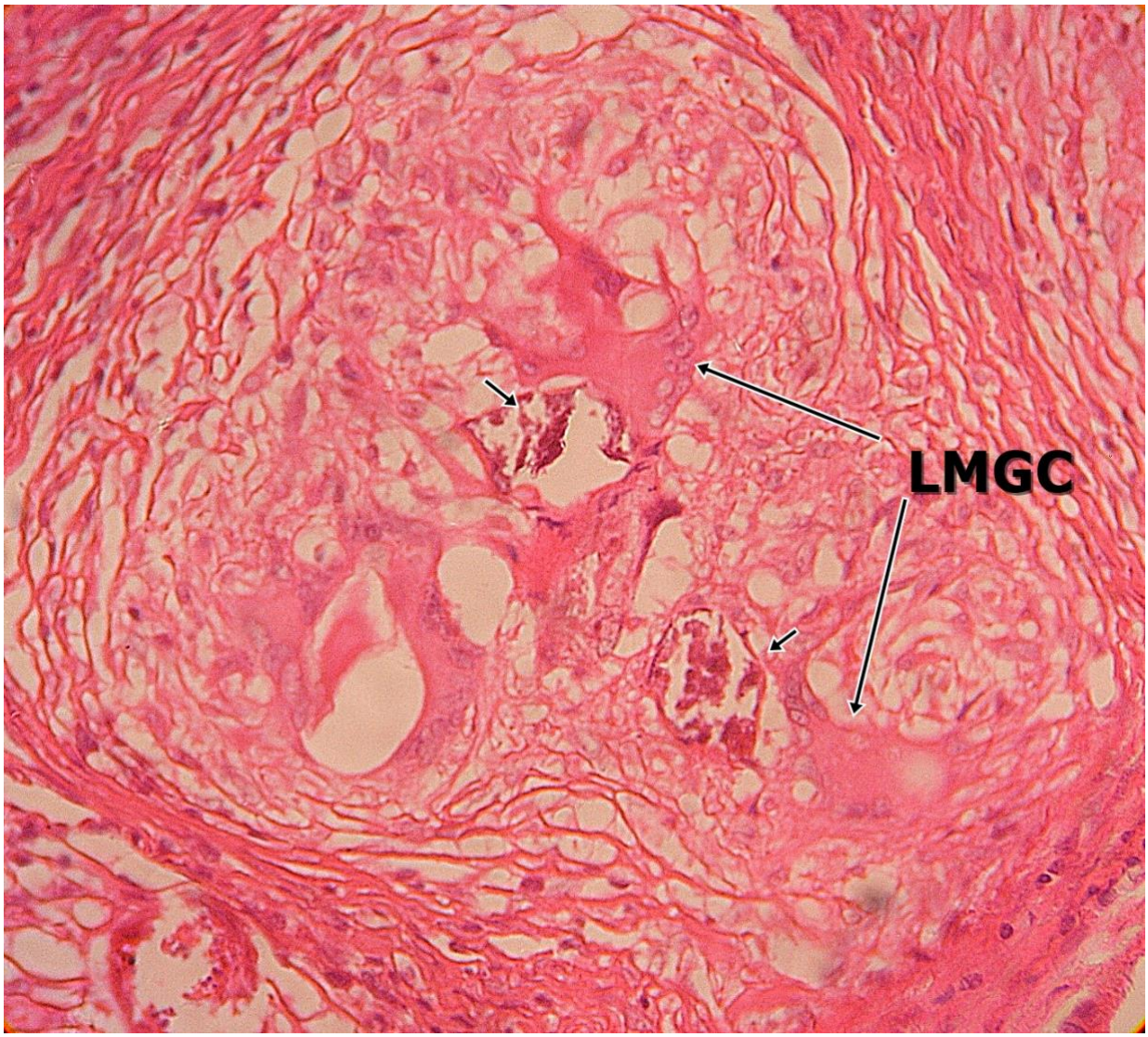

Figure 3. Photomicrograph showing granulomas containing Langhans Type Multinucleate Giant Cells (LMGC) surrounding ova of Schistosoma worms (short arrows) [H & E x 100]. [H & E x 200].

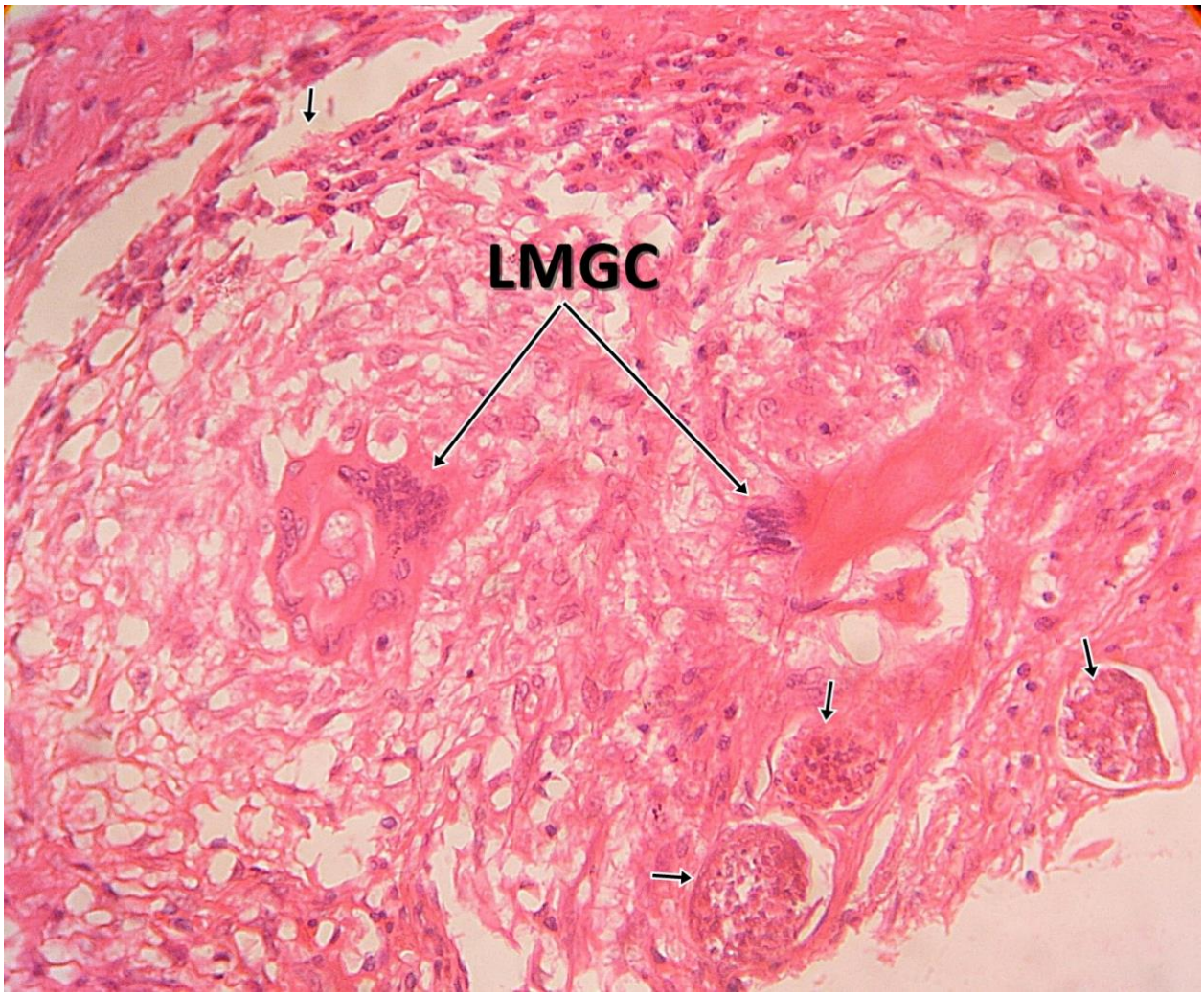

Figure 4. Photomicrograph showing numerous granulomas containing Langhans Type Multinucleate Giant Cells (LMGC) surrounding ova of Schistosoma worms (short arrows) [H & E x 100].

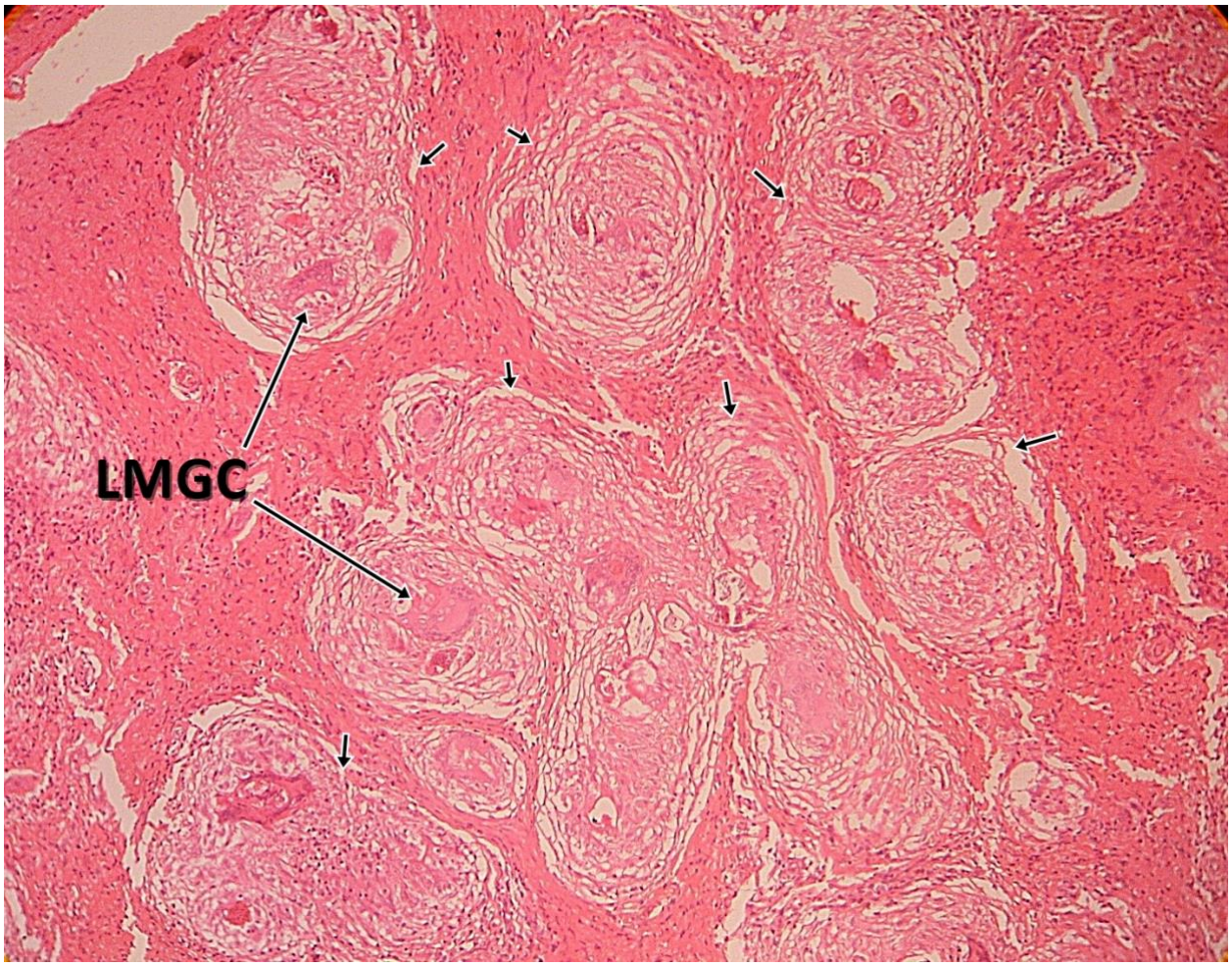

Figure 5. Photomicrograph showing numerous granulomas (G) containing Langhans Type Multinucleate Giant Cells surrounding ova of Schistosoma worms (short arrows) [H & E x 200].

Figure 6. Photomicrograph showing granulomas containing Langhans Type Multinucleate Giant Cells (LMGC) surrounding ova of Schistosoma worms (short arrows) [H & E x 200].

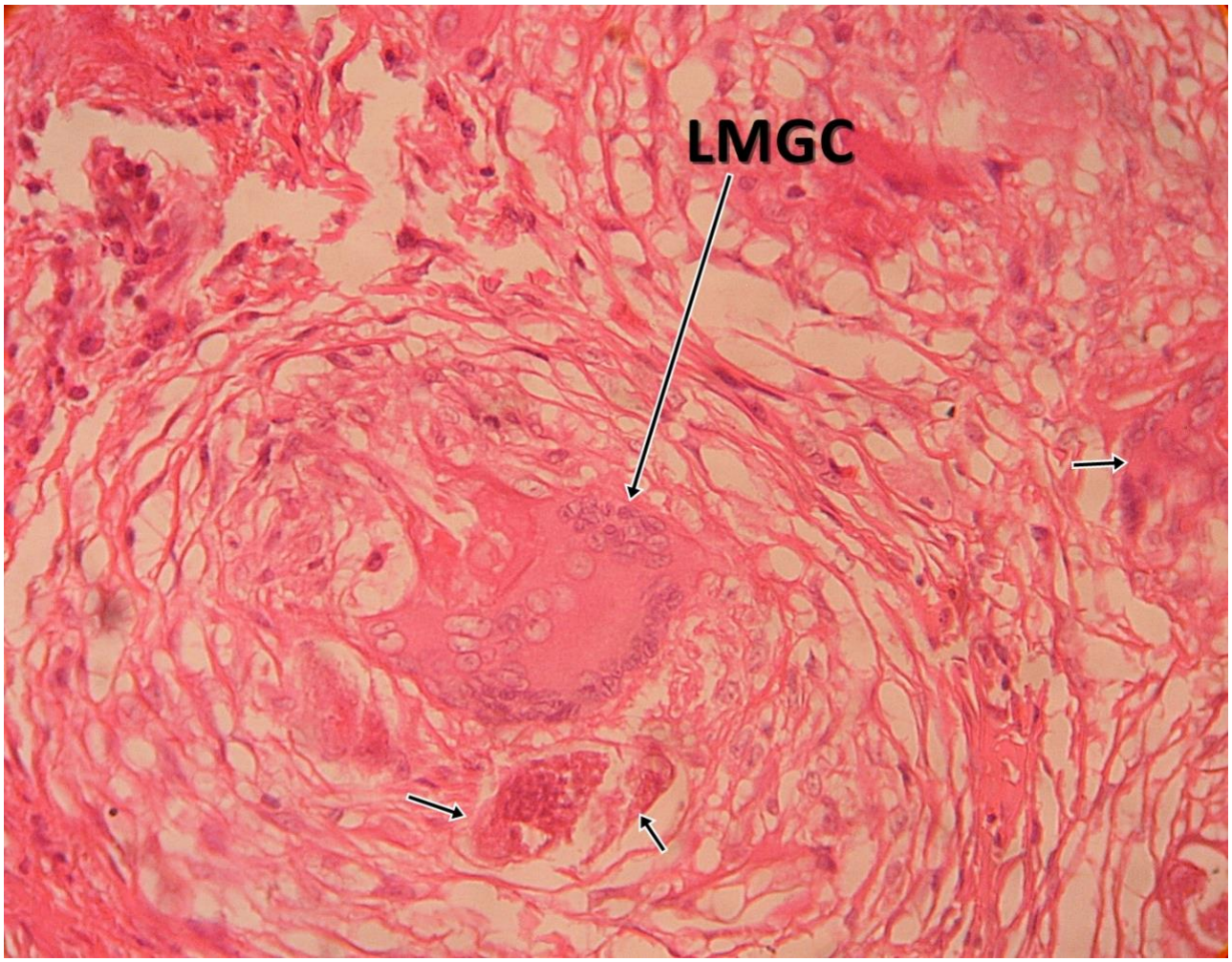

Figure 7. Photomicrograph showing an enlarged vein within which there is an adult pair of worms (SW) seen wrapped in the copulatory embrace with the slender female adult lying within the gynaecophoric groove of the male with surrounding ova of Schistosoma worms (short arrows) [H & E x 100].

Figure 8. Photomicrograph showing an enlarged vein within which there is an adult pair of worms (SW) seen wrapped in the copulatory embrace with the slender female adult lying within the gynaecophoric groove of the male with surrounding ova of Schistosoma worms (short arrows) [H & E x 200].

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S/N		Case 1	Case 2	Case 3
1	Age	7 yrs	12 yrs	14 yrs
2	Place of presentation	CHOP	PSC	GOPD
3	Symptoms	Left testicular swelling	Left testicular swelling	Left testicular swelling
4	Signs	Mild tenderness	Not tender	Not tender
5	Duration of symptoms before presentation	1 year 6 months	3 yrs	1 year
6	History of hematuria	Terminal and intermittent hematuria of 1 yr	Intermittent hematuria of 2 yrs	Intermittent hematuria of 6 months
7	Pallor	Mild pallor	Mild pallor	Not pale
8	Weight loss	No weight loss	Mild weight loss	No weight loss
9	History of endemic region	Positive	Positive	Positive
10	Spleen, liver and kidney involvement	Mild splenomegaly	Mild splenomegaly	Nil
11	Provisional diagnosis	?Testicular tumor	?Testicular tumor	?Testicular tumor R/o orchitis
12	Type of biopsy	Excisional biopsy (orchidectomy)	Incisional biopsy	Excisional biopsy (orchidectomy)
13	Therapy	Praziquantel, 400 mg stat	Praziquantel, 400 mg stat	Praziquantel, 400 mg stat
14	Duration of follow–up	The patient was lost to follow-up 3 months after	The patient was lost to follow-up 1 year after.	The patient was lost to follow-up 6 months after
15	Outcome	Right testis preserved	Significant reduction in size of masses, both testes preserved and testicular function fully preserved.	Right testis preserved

Table 1. Summary of clinical presentations.
CHOP, Children Out Patient; PSC, Pediatric Surgical Clinic; GOPD, General Out Patient Department

S/N	Test	Case 1	Case 2	Case 3
1	Hb	8.5g/dL	9.6 g/dL	10.3 g/dL
2	PCV	26%	29%	31%
3	ESR	90 mm 1st hour	80 mm 1st hour	100 mm 1st hour
4	Platelet count	108000/mm³	Not done	Not done
5	Total WBC	4.5x109/L (N-60%, L-38%, E-2%)	$6.8x10^9$/L (N-59%, L-38%, E-2%, M-1%)	$8.3x10^9$/L (N-62%, L-36%, E-02%)
6	SEUCr	Within normal limits	Within normal limits	Within normal limits
7	Urine and Stool M/C/S examinations	No ova seen	No ova seen	No ova seen
8	Mantoux test	Negative	Negative	Negative
9	HIV I & 2 antibodies	Negative	Negative	Negative
10	Chest X-ray	Within normal limits	Within normal limits	Within normal limits
11	Scrotal ultrasonography	Not done	Not done	Not done
12	Biopsy	Excisional biopsy (orchidectomy)	Incisional biopsy	Excisional biopsy (orchidectomy)
13	Gross features	An orchidectomy specimen measuring 4.0x2.5x1.5 cm was received	Three pieces of greyish white tissues aggregating to 1.2 cm were received	A piece of greyish white tissue measuring 6.0x 5.0x 2.0 cm was received
14	Microscopic features	Non-caseating granulomas around viable and degenerating Schistosoma eggs with calcification	Non-caseating granulomas around viable and degenerating Schistosoma eggs with calcification; adult worms within vessels	Granulomas around Schistosoma eggs with adult worm pairs wrapped in the copulatory embrace in scrotal and testicular
15	Histopathological diagnosis	Testicular Schistosomiasis	Testicular Schistosomiasis	Testicular Schistosomiasis

Table 2. Summary of laboratory findings.

Hb, Hemoglobin concentration; PCV, Packed Cell Volume; ESR, Erythrocyte Sedimentation Rate; Total WBC, Total White Blood Cell Count; N, Neutrophils; L, lymphocytes; E, eosinophils; M, monocytes; SEUCr, Serum Electrolyte Urea, Creatinine Concentration