Human Dirofilariasis Presenting as Scrotal Mass

Flavia Tripi, Francesco Scarlata, Vincenzo Verde, Giuseppe Li Voti, Cinzia Chiaramonte*  

1 Department of Mother and Child Care, Pediatric Surgery, University of Palermo, AOU Policlinico Paolo Giaccone  
2 Department of Mother and Child Care, Division of Infectious Diseases, University of Palermo, AOU Policlinico Paolo Giaccone  
3 Department of Radiology, Section of Radiological Sciences, University of Palermo, AOU Policlinico Paolo Giaccone

Abstract

A large spectrum of pathology may present in children as an asymptomatic scrotal mass (ASM) ranging from congenital to neoplastic lesions.

We observed a 11 months old child referred to us for suspicion of neoplasm presenting with an asymptomatic scrotal mass. High frequency scrotal sonography enabled diagnosis of nematode infection (Dirofilaria repens): viability of the adult worms was assessed by looking for the animated documentation of filarial dance sign (FDS). Differentiation from malignancy allowed an appropriate management consisting in minor surgery.

Extra-testicular pathologies are encountered relatively infrequently but awareness of different possible etiology avoids unnecessary extensive surgery.

Keywords: Asymptomatic Scrotal Mass (ASM); Human Subcutaneous Dirofilariasis (HSD); Children; Ultrasound;

Introduction

Extra-testicular cystic and solid scrotal masses are frequently observed in pediatrics. The most common pathologies are benign (i.e.: appendage torsion, epididymitis and varicoceles); however para-testicular rhabdomyosarcoma represents 50% of solid extra-testicular masses [1]. It could be difficult to differentiate from malignancy because sonographic findings are patognomonic only in 37% [2]. Human dirofilariasis is a rare event as genital location but is frequently indistinguishable from a tumor. Human subcutaneous dirofilariasis (HSD) should be kept in mind in differential diagnosis of scrotal masses as it is considered an emergent zoonosis.

Patient presentation

A child aging eleven months presented to our Pediatric Surgery Unit complaining a painless nodule on his left hemiscrotum (ASM). He had neither pathological history nor was in contact with dogs or other animals and never visited other countries.

The physical examination revealed no pathological findings except for a single painless hard nodule of 1 cm at the apex of the scrotum with any connection to the testis. The skin over the nodule was normal without any signs of inflammation and the inguinal lymph nodes were not palpable.

Routine laboratory tests of the blood showed no abnormal values for the age. Tumors markers as βHCG and α-fetoprotein were within normal range.

Suspecting an extra-testicular neoplasm an ultrasound scan with high-resolution linear probe 10 – 12 MHz of the nodule was performed. It showed the presence of an anechoic cystic formation without any vascularization in its contest or vascular pole at the color-doppler integration. Within a cyst mass a tubular coiled structure with parallel echogenic wall in continuous movements was found: to identify the species of worm specialist opinion was requested.

The characteristic pattern movements of the nematode (Filaria dance sign-FDS) led to diagnosis of human subcutaneous dirofilariasis (HSD). Testicular tissue was normal (FIGURE 1).

The surgery was performed under general anesthesia with minor surgery approach by a transversal scrotal incision of less than 1 cm over the nodule. The mass wasn't in connection with testicular or funicular structures. It was isolated from the dartos without any damage to the capsule and was enucleated intact (FIGURE 2).

The histopathological examination showed a simil-cystic structure with granulomatous inflammatory wall with a high number of eosinophils and a tubular structure of 10 cm of length inside moving when placed in physiological solution (FIGURE 3).

The diagnosis of infection from Dirofilaria repens was based on the morphological characteristic of the worm that displayed the typical features of a female.

Abstract

A large spectrum of pathology may present in children as an asymptomatic scrotal mass (ASM) ranging from congenital to neoplastic lesions.

We observed a 11 months old child referred to us for suspicion of neoplasm presenting with an asymptomatic scrotal mass. High frequency scrotal sonography enabled diagnosis of nematode infection (Dirofilaria repens): viability of the adult worms was assessed by looking for the animated documentation of filarial dance sign (FDS). Differentiation from malignancy allowed an appropriate management consisting in minor surgery.

Extra-testicular pathologies are encountered relatively infrequently but awareness of different possible etiology avoids unnecessary extensive surgery.

Keywords: Asymptomatic Scrotal Mass (ASM); Human Subcutaneous Dirofilariasis (HSD); Children; Ultrasound;

Introduction

Extra-testicular cystic and solid scrotal masses are frequently observed in pediatrics. The most common pathologies are benign (i.e.: appendage torsion, epididymitis and varicoceles); however para-testicular rhabdomyosarcoma represents 50% of solid extra-testicular masses [1]. It could be difficult to differentiate from malignancy because sonographic findings are patognomonic only in 37% [2]. Human dirofilariasis is a rare event as genital location but is frequently indistinguishable from a tumor. Human subcutaneous dirofilariasis (HSD) should be kept in mind in differential diagnosis of scrotal masses as it is considered an emergent zoonosis.

Patient presentation

A child aging eleven months presented to our Pediatric Surgery Unit complaining a painless nodule on his left hemiscrotum (ASM). He had neither pathological history nor was in contact with dogs or other animals and never visited other countries.

The physical examination revealed no pathological findings except for a single painless hard nodule of 1 cm at the apex of the scrotum with any connection to the testis. The skin over the nodule was normal without any signs of inflammation and the inguinal lymph nodes were not palpable.

Routine laboratory tests of the blood showed no abnormal values for the age. Tumors markers as βHCG and α-fetoprotein were within normal range.

Suspecting an extra-testicular neoplasm an ultrasound scan with high-resolution linear probe 10 – 12 MHz of the nodule was performed. It showed the presence of an anechoic cystic formation without any vascularization in its contest or vascular pole at the color-doppler integration. Within a cyst mass a tubular coiled structure with parallel echogenic wall in continuous movements was found: to identify the species of worm specialist opinion was requested.

The characteristic pattern movements of the nematode (filaria dance sign-FDS) led to diagnosis of human subcutaneous dirofilariasis (HSD). Testicular tissue was normal (FIGURE 1).

The surgery was performed under general anesthesia with minor surgery approach by a transversal scrotal incision of less than 1 cm over the nodule. The mass wasn't in connection with testicular or funicular structures. It was isolated from the dartos without any damage to the capsule and was enucleated intact (FIGURE 2).

The histopathological examination showed a simil-cystic structure with granulomatous inflammatory wall with a high number of eosinophils and a tubular structure of 10 cm of length inside moving when placed in physiological solution (FIGURE 3).

The diagnosis of infection from Dirofilaria repens was based on the morphological characteristic of the worm that displayed the typical features of a female.
The patient has a normal post-operative recovery and has not been treated with any antielminthic medication.

**Discussion**

The painless scrotal masses are extra testicular in 28% of cases and 50% of solid extra-testicular masses in children are malignant (rhabdo myosarcoma) [1]. Patient age distribution shows 2 peaks at 0 to 1-year and 13 to 14-year interval, more than one third of the painless scrotal masses were found during the first year of life, predominantly congenital anomalies (in utero torsion) and neoplasia [2]. Benign lesion is distinguishable by their clinical presentation and ultrasound appearance. Less commonly may not be easily classified and the patients undergo explorative inguinotomy in order to rule out any malignancy. In some region of the world Nematodes infection as Dirofilariasis should be considered in the differential diagnosis of the ASM.

Human dirofilariosis is an emerging zoonotic infection caused by Dirofilaria repens or, less frequently, Dirofilaria immitis.

Many species are known to parasite wild and domestic animals. D. repens and D. tenuis occurring in Mediterranean countries a, middle east, Africa and southeast asia. D. tenuis (parasite of raccoon) is cause of most human cases in USA. D. ursi (bear) e subdermata (porcupine D) in North USA and Canada. These worms cause dirofilariasis in dogs, cats and wild carnivores transmitted by Aedes, Culex and Anopheles mosquitos. The adult worm live in in subcutaneous tissue of its natural host producing microfilariae that circulate in the blood. Mosquitos take up the first stage larva (L1 - microfilaria) while feeding infected animal. Within the vector, L1 develops in 7 days in L2 stage and after other 6-7 days the larvae became infective (L3) and migrate to the proboscis of the insect. The parasite penetrates a new host during subsequent meal. The larva develops into adult form in both humans and animals in subcutaneous tissues (D. Tenuis, D. repens) [3].

Adult D. repens worm reaches a diameter of 0,5 mm and a length 5 to 15 cm. Usually female worm measures 220-660 microns across; the central intestine and the genital organs (two uteri) are located in the pseudo-coelom; the thick, multilayered cuticle is provided with 95-105 sharp, longitudinal ridges, the latter being separated from each other by a distance of 12 microns; the circumferential muscle cell layer, covering the inner side of the cuticle, is bilaterally interrupted by the large chord cells, two to five nuclei of which are discernible in each cross section. [4]

In our case the worm was a female of 10 cm of length.

The adult worms in infested definitive hosts of D. immitis are located in the pulmonary arteries and the right-hand heart chambers and cause a life-threatening condition known as canine and feline heart-worm disease [1]. Adult D. repens worms are found in subcutaneous tissues of animals, and it is the cause of asymptomatic infections, subcutaneous nodules or allergic dermatitis. Humans are not fully suitable hosts and so that larvae L3 transmitted by mosquitos bite generally dies before reaching sexual maturity and does not release viable microfilaria.

In a large series studied in Ukraine (1465 cases) 3 patients had 2 lesions. Each lesion contain1 parasite (majority females) [5].

In spite of its reduced pathogenicity for dogs, D. repens has been recently considered as an emerging zoonosis in Europe. In fact the effects of climatic changes on the density of the mosquitos and the spreading of a new competent vector as. Albipictus so as the increased translocation of dogs has favored the recent increase of infection rates in regions where the parasite is endemic and also its spreading to areas (e.g. northern and eastern Europe) previously free from this infection. In these countries the cases of autochthonous human dirofilariasis were usually preceded by imported cases as well as canine dirofilariasis or dirofilariasis of...
wild carnivores. Italy is the western European country with the highest infection rate in dogs. In a large survey carried out on 2,512 dogs in the nine provinces of Sicily, a very high prevalence of D. repens microfilariae was found in Trapani Province while in other provinces the infection rate was regarded from 0.4 to 4.7%. The most important vectors in Italy and in many European countries are the opportunistic feeders Aedes albopictus and Culex pipiens [6].

In Humans the worms are usually single and immature, and so the microfilariae are not present in peripheral blood. However females carrying microfilariae have been described so full development and fertilization of D. repens in humans is possible.

Only one report of circulating microfilaria in a human exists in medical literature [7] and a case report showing the present of microfilaria in a nodule examined by fine needle aspiration [8].

The incubation period in the vertebrate host is 6-8 months [9]. Migration of the worm from the bite side may result in local swellings with changing localization (cutaneous larva migrans). Immune-mediated formation of subcutaneous nodule surrounding the larvae is the most frequent pathologic finding associated with the infection.

The common sites of the lesions are face and eyelids, chest wall and upper arms. The nodule may be painful and erythematous. The ocular infection includes nodule formation in the orbital zone and eyelids, as well as the presence of intact migrating worm in sub conjunctival and intra vitreous tissues [10] as well as in intra orbital musculature [11].

Intra Dural dirofilariasis was reported in a 6-years-old girl presented as a extra-spinal tumor. Other localizations of Dirofilaria were peri-tendon of the hand, breast infection, retroperitoneal, pulmonary [12, 13, 14, 15, 16].

Dirofilariosis of the male genitalia remains a rare event. (Only 21 cases reported) [17, 18] and can be mistaken for scrotal malignancy, resulting in unnecessary extensive operation [19].

A3-years-old child underwent orchectomy as infestation by Dirofilaria repens was mimicking an acute scrotum [18].

Ultrasound examination performed in our child by a high-resolution probe showed a tubular coiled structure with parallel echogenic wall in continuous movement (FDS) that has already observed only in Bancroft an filariasis [20] however the host inflammatory response can destroy worm’s morphology making identification of the parasite difficult.

Absent vascularization inside the mass and the absent of a vascular pole allowed differential diagnosis with a neoplastic mass as para-testicular rhabdomyosarcoma, in which is mandatory a more aggressive treatment [1].

In Dirofilariasis, no reliable signs, symptoms or laboratory parameters are available. Eosinophilia and elevated IgE levels are almost always absent although elevated IgE level could confirm the diagnostic suspicion. We found only eosinophilic infiltration in the capsule of the cyst at the histopathological examination of the cyst.

Treatment of nodule consists of complete excision. Studies carried out on the effects of some drugs on anthropodonic filariae obtained discordant results. Ivermectine or Diethylcarbamazine may be advised for deeper multiple lesions. However, it seems that only doxycycline may have a possible adulticidal effect due to its action on the bacterial endosymbiont Wolbachia, present in most filarial species (D.repens included).

**Conclusion**

Extra-testicular pathologies in children are relatively rare: awareness of their clinical and ultrasound characteristics may allow to appropriate management. Testicular ultrasound proved to be highly reliable in differentiating intra testicular from extra testicular lesions but it demonstrated poor specificity because of extensive overlap between benign and malignant pathologies. HSD of the male genitalia remains a rare event: 21 cases reported in literature involving scrotum, epididymis spermatic cord. Considering the climate changes and the increasing of migration and international travels phenomena, the presence of an unclear nature subcutaneous nodule should prompt suspicion of infestation by nematodes and in particular of D. repens.

To our knowledge this is the first genital case of HSD reported in an infant aging less than 1 year of age a live D. repens mimicking an extra testicular neoplasm.

**References**


9. Klinteberg K, Petersen E, Pehuschinayna N.Y., Ermakova LA, NagornySA, Larsen CS - Peribortal Dirofilaria repens imported...


