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Case Report

Human subcutaneous dirofilariasis infection in Grodno Region, Belarus

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ABSTRACT

A major worldwide concern, dirofilariasis is a zoonotic helminth disease that is transmitted by vectors. The disease is caused by nematodes of the species *Dirofilaria* of the family Filariidae. Mosquitoes are intermediate hosts, along with dogs, cats, and wild carnivores. The larva that penetrates human tissues after a mosquito bite begins to grow, develops and spreads the disease. A multidisciplinary approach is required because the clinical symptom is dependent on the location of the helminth. A 60-year-old woman presented with a tumor-like formation in the right temporal region of the scalp. Preliminary diagnosis was made as “epidermal cyst of the right temporal region” upon the examination. An ultrasound of the soft tissues of the right temporal region of scalp was performed and it demonstrated a hypoechoic formation of spherical shape. A thread-like formation which was curled into a ball was found following the surgical excision of the tumor-like formation. Diagnosis was confirmed following the pathohistological examination. The result of the postoperative pathohistological examination was; fibrous tissue with focal chronic inflammation with the presence of a helminth. The patient was diagnosed with “dirofilariasis of the subcutaneous fat of the right temporal region” (ICD-10, B74.8). Patients with subcutaneous Dirofilariasis should be treated with surgical excision following a thorough ultrasound examination. Early and efficient diagnosis of this disease can prevent the long suffering of the patient while misdiagnosis can lead to unnecessary investigations.

Keywords: Subcutaneous dirofilariasis, Surgical treatment, Ultrasound

INTRODUCTION

Dirofilariasis is a vector borne zoonotic helminth disease which is a major consideration in the European Union specially in countries like Belarus, Russia and Ukraine.[1] It is caused by *Dirofilaria immitis* and *Dirofilaria* repens. The disease is caused by nematodes of the Filariidae family of the genus *Dirofilaria.*

The species listed below are members of the genus Dirofilaria: *Dirofilaria immitis* - a cardiac helminth of dogs, parasitize the heart's right ventricle, pulmonary artery, and hollow veins *Dirofilaria repens*- in dogs, *Dirofilaria tenuis*- in raccoons and *Dirofilaria ursi*- in bears.[2] Heartworm disease, also known as canine cardiopulmonary dirofilariasis, is caused by *Dirofilaria immitis*, which is present in the pulmonary artery and the right chambers of the heart. *Dirofilaria repens* causes subcutaneous dirofilariasis because it is found in subcutaneous tissues.[3]

The clinical signs caused by the migration of immature helminths in human tissues or internal organs have a long course and gradual development.[4]

Dirofilariasis is characterized by the feeling of a live "worm" moving and crawling inside a seal, tumor, or subcutaneous node. Headache, nausea, weakness, and discomfort where the helminth is located, coupled with radiation along the nerve trunks, are all possible symptoms of dirofilariasis. The location of the helminth determines the clinical symptoms. Ophthalmologists, dermatologists,

dentists, urologists, otolaryngologists, therapists, oncologists, surgeons, infectious disease experts, and others can be contacted by patients with dirofilaria because the clinic varies, and the lesion's location varies.[1]

The parasite has a filamentous body covered with a thin outlined cuticle of milky or greyish yellow color. Sexually mature females *D. repens* 130-150 mm long, up to 0.5 mm in diameter, *D. immitis* 180-300 mm long, up to 0.5 mm in diameter, males 100-110 mm long, up to 0.4 mm in diameter. The body consists of a skin-muscular sac represented by a cuticle, hypodermis and muscle elements. In the body of animals, the parasites themselves live from 4-5 months to 2 years. Dirofilaria are viviparous, their larvae are called microfilariae, have a length of 0.22-0.30 mm. Females give birth directly into the blood of a parasitized animal. Microfilaria circulates for 2.5 years, without changing morphologically, or until they get into the intestine of the mosquito during bloodsucking.

Carnivorous animals from the dog, cat and viverra families are the definitive owners of dirofilaria. When a mosquito (*Culex*, *Aedes*, or *Anopheles*) sucks blood, blood-containing microfilaria enters its intestines, progresses to the invasive stage, and can then spread to the next host. When mosquito saliva enters the human skin, microfilaria actively travel through the subcutaneous tissues before developing into an adult dirofilarial.[2]

Dirofilaria develops with a double change of hosts. Sexually mature dirofilaria live in the subcutaneous tissue (*D. repens*) and the blood system *(D. immitis*) of the definite hosts - dogs, cats, wild carnivores.[4] Females *D. repens* and *D. immitis* are viviparous, after fertilization, the development of microfilaria occurs directly in the uterus, the larvae of the first stage (L1) of microfilaria are reborn. Up to 30,000 microfilariae are reborn per day, which circulate in the blood of carnivores (the final host) for up to 2.5 years. Microfilaria doesn't have cuticles, their length is up to 0.3-0.4 mm, width is up to 0.006-0.008 mm, which is comparable to the diameter of an erythrocyte.

The causative agent of dirofilariasis penetrates the human body through the transmissive mechanism of infection. Blood microfilaria from infected animals is non-invasive and does not directly endanger people or other animals. Being intermediary hosts and carriers of these parasites, mosquitoes contract the disease when they feed on infected animals (L1 stage). In intermediate hosts, such as mosquitoes, microfilaria larvae settle within their bodies. The parasite larvae appear in the insect's intestine 24 hours after blood sucking. On the second day, they move to the Malpighian vessels, where they change and grow over the next 16 days until they reach the invasive stage L3, at which point they enter the insect's body cavity and penetrate the lower lip of the mosquito, reaching a length of 0.8-0.9 mm. and live there until the bite.[2]

Invasive larvae enter the bloodstream from the insect's proboscis, penetrate the skin, travel through the bloodstream to different tissues, and reach the L4 stage of the microfilaria in 9–15 days, which is how humans (or animals) become infected. They change from young, immature beings after two months to adults by actively migrating through subcutaneous tissue. Human tissues for dirofilarial are an unfamiliar environment in which they do not develop to a sexually mature state. Thus, a person is a dead-end host: in most cases, microfilariae that have fallen into human blood die; also, and this is usually an unfertilized developing female, due to the absence of a male, fertilization does not occur, microfilaria does not revive. The larva that gets into the human tissues at the site of a mosquito bite begins to grow and increase in size.[2]

CASE REPORT

We are presenting a 60-year-old woman, who presented to surgical clinic complaining of a tumor-like formation in the right temporal region of the scalp. The anamnesis revealed that the formation had appeared suddenly about 2 months ago. Upon the examination it was found that the tumor-like formation is 1 cm in diameter, spherical in shape, elastic upon palpation, smooth, movable, painless, and the skin above it is unchanged is visualized in the right temporal region of the scalp.

Preliminary diagnosis was made as "epidermal cyst of the right temporal region" upon the examination. Then a preoperative examination was performed using ultrasound of the soft tissues of the right temporal region of the scalp and a complete blood count.

The ultrasound of the soft tissues of the right temporal region of scalp demonstrated a hypoechoic formation of a spherical shape with a clear, even contour, measuring 7×7 mm, is localized subcutaneously in the right temporal region. The results of the complete blood count were; erythrocytes 4.1×10^{12}/l, hemoglobin 126 g/l, leukocytes 4.3×10^{9}/l, ESR 13 mm/hour, color index 0.92, platelets 187×10^{9}/l, formula: eosinophils 5%, segmented neutrophils 54%, lymphocytes 38%, monocytes 3%, and total cells: 100.

Table 1: Results of the complete blood count.

Complete blood count	Values
Erythrocytes	4.1×10^{12}/l
Hemoglobin	126 g/l
Leukocytes	4.3×10^{9} /l
Platelets	187×10^{9}/l
Eosinophils	5%
Segmented neutrophils	54%
Lymphocytes	38%
Monocytes	3%
Total	100%
ESR	13 mm/hour
Color index	0.92

After the pre-operative examination, the patient was offered surgical intervention. After obtaining the patient's consent for the operation, excision of the tumor-like formation was carried out. The surgical excision was carried out under local anesthesia; sol. lidocaine 2%-2 ml. Surgical field was cleaned with 70% ethanol solution prior to the incision. A 2 cm long incision was made over the tumor-like formation, an insignificant amount of fluid (1 ml) of gray color was released. A thread-like formation about 7 cm long, 0.5 mm thick, transparent-white, elastic consistency, which was curled into a ball was found inside the tumor-like formation (Figure 1). During the excision, an area of slightly altered subcutaneous fat tissue possibly with remnants of the capsule of the thread-like formation was also removed. At the end of the surgical intervention, hemostasis was performed and sutures were put on the wound.

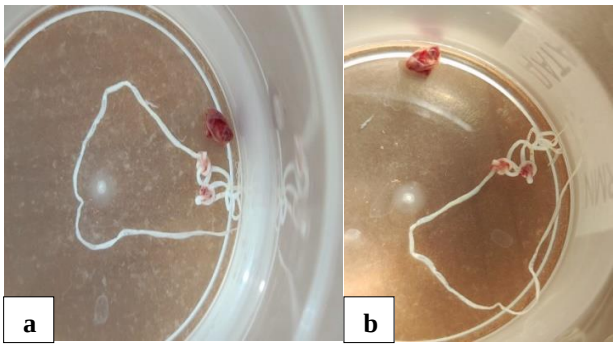

Figure 1 (a and b): Thread-like formation (helminth) removed during the operation (7 cm long, 0.5 mm thick, transparent-white, elastic consistency, which was curled into a ball) The excisional biopsy specimen contains a subcutaneous mass on the left side and the helminth on the right side.

According to the case discussed above, the removed formation was sent for pathohistological examination after the surgical intervention. The result of the histological examination was; fibrous tissue with focal chronic inflammation with the presence of a helminth. The patient was diagnosed with "dirofilariasis of the subcutaneous fat of the right temporal region" (ICD-10, B74.8). As there were no postoperative complications, the stitches were removed on the 7th day after the operation.

DISCUSSION

Dirofilariasis is a rare condition in Belarus, but currently there is a tendency for the growth and wider spread of this disease. The recent discovery of an adult heartworm in a wild wolf and increasing human cases of *Dirofilaria* in mosquitoes suggest a high likelihood of autochthonous transmission in Belarus. Two *Dirofilaria*-positive pools were detected in Belarus, indicating a high prevalence of the disease for both mosquitoes and vertebrate hosts despite a low number of mosquitoes analyzed.[5] In the Old World, there are more cases of subcutaneous/ocular dirofilariasis caused by D. repens than pulmonary dirofilariasis produced by *D. immitis*.[6] Isolated cases of Belarus were described until the middle of the last century, which were deemed to be imported. Then recently the number of observations has increased. Human infections with *Dirofilaria* have been identified on a regular basis since 1997; some of these infections are regarded as autochthonous.[5]

Experts associate the expansion of the area of this invasion with global warming. Temperature increases can decrease the life cycle and generation period of many vector species, making them more appropriate for the possible spread of infections that cause disease. [7] For mosquitoes to thrive and evolve into the infectious third stage, temperature and environmental factors are crucial. Bandar Abbas's climate is conducive to the development and reproduction of various mosquito species as vectors.[8]

As well as with the movement of citizens and animals, which contributes to the "introduction of diseases" from other countries. Also, the expansion of the distribution area is associated with a number of epidemiological and environmental factors: Environmental factors, patient habits, and sanitation have a direct impact on the incidence of parasite eye illnesses.[6] An increase in the number of stray dogs (the final hosts of the parasite) and *Dirofilaria* species *D. immitis*, *D. repens*, and *D. tenuis* are the most common *Dirofilaria* species that infect humans, as they naturally inhabit dogs and other wild canids.[7] A high level of their invasiveness, a large number of vectors (mosquitoes), because of human disease in the amphibian population (frogs) contract the disease by mosquito bites from species such as *Aedes*, *Anopheles*, *Culex*, and *Mansonia*.[7] Since there are no targeted anti-epidemic measures to limit the spread of dirofilariasis in the Republic of Belarus, there will be an even greater increase in the future. Hence, it should be expected in the near future.

The clinical manifestations are caused by the migration of immature helminths in human tissues or internal organs, characterized by slow development and a long course.

The clinical finding of our case study was only presented with a smooth, movable, painless tumor-like formation in the right temporal region. However, unusual clinical symptoms in another case study about human dirofilariasis in the forehead included headache, nausea, facial and forehead puffiness, as well as localized itching, irritation, and mild discomfort.[8]

The disease can be diagnosed in humans using the clinical signs and parasitological, serological, histopathological, ultrasound (sonography), and molecular methods.[6]

In our case study, the preliminary diagnosis was done upon the examination and found an epidermal cyst on the right temporal region. The ultrasound showed a hypoechoic formation of a spherical shape with a clear, even contour, measuring 7×7 cm, is localized subcutaneously in the soft

tissues of the right temporal region. The first-line imaging method with the highest specificity for detecting a cystic nodule with an interior serpiginous structure made up of parallel echogenic lines is ultrasound. In lymphatic filariasis, the serpiginous structure exhibits an active spinning movement known as the "filarial dance sign".[9] Within a cyst, ultrasonography reveals tubular, actively moving entities with parallel echogenic stripes.[7] Complete blood count was done in laboratory tests. Despite the parasitic infection, IgE levels showed no elevation. In laboratory studies, elevated IgE levels and peripheral blood eosinophilia are uncommon.[7] The definite diagnosis was done by pathohistological examination, and it was confirmed to be Dirofilariasis of the subcutaneous fat of the right temporal region in our case study. The conclusive diagnosis of Dirofilaria is obtained through histopathologic demonstration. A multilayered, thick cuticle with longitudinal ridges and a well-developed circumferential musculature broken up by lateral chords are characteristics of Dirofilaria.[7] Transverse striation and external longitudinal cuticular ridges are characteristics that distinguish *D. repens* from other Dirofilaria. Only after examining the fully developed worm may it be feasible to identify the species precisely. However, the lack of longitudinal ridges and transverse striation distinguishes *D. immitis* from *D. repens.*[10] PCR reaction can also be used. Identification of species using molecular sequences can bolster the findings of this investigation.[8] One disadvantage of this study is that the test has not been performed on patient tissue.

Limitations

The study did not include follow-up assessments after the surgical excision, limiting our ability to evaluate long-term outcomes.

CONCLUSION

Dirofilariasis of the subcutaneous fat is a rare zoonotic helminth disease in Belarus. The preliminary diagnosis of our patient was made as “epidermal cyst of the right temporal region” upon the examination. Then a preoperative examination was done using Ultrasound of the soft tissues of the right temporal region of the scalp and a complete blood count. Characteristic features in ultrasound examination of the tumor-like formation can play a major role in suspecting the presence of a helminth prior to the surgical intervention. The diagnosis was made as ‘Dirofilariasis of the subcutaneous fat of the right temporal region’ using a pathohistological examination following the surgical excision. Swift and early diagnosis of Dirofilariasis is important in order to reduce the suffering of the patient.

This case study hopes to bring more awareness and understanding about human subcutaneous Dirofilariasis. Also, it is important to suspect the presence of Dirofilariasis even in non-endemic areas when the patient comes with a similar clinical presentation as we have discussed above.

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