SUBCUTANEOUS TAENIA CRASSICEPS INFECTION IN A PATIENT WITH NON-HODGKIN’S LYMPHOMA

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Abstract. Infections with larvae of Taenia crassiceps are rare in humans and have mostly affected patients with acquired immunodeficiency syndrome. We report the first case of a patient with malignancy (non-Hodgkin’s lymphoma) and infection of the subcutis and muscles of the hand and forearm. Surgery and antiparasitic chemotherapy led to a complete cure.

INTRODUCTION

Taenia crassiceps is a tapeworm whose adult form lives in the intestine of carnivores, mostly foxes, in North America, Europe, and Russia. Rodents are natural intermediate hosts, and they harbor the cyst-like larvae (metacestodes, cysticerci) in the peritoneal cavity, where they multiply by asexual budding. Humans serve as intermediate hosts when food or water contaminated with feces from infected canids or felids is consumed. In humans, metacestodes develop in the subcutis and among muscular tissue. Most infections occur in patients with acquired immunodeficiency syndrome.1–3 Thus, immunosuppression seems to be a prerequisite for infection. In addition, immunocompetent patients have been reported in whom larvae settled in the anterior chamber of the eye or grew subretinally (Table 1). In this report, we describe the first case of an infection with the larval form of T. crassiceps in a patient with underlying malignancy. The patient was treated for a non-Hodgkin’s lymphoma and was thereby immunosuppressed. Combined surgical removal of the cystic larvae from a subcutaneous site and treatment with albendazole and praziquantel led to a complete cure.

CASE REPORT

An 82-year-old German woman was hospitalized because of progressive pain and swelling in the left forearm and back of the left hand. The symptoms had started six weeks earlier after a fall on the hand. Soft tissue injury with lymphedema but without fracture was diagnosed by her family physician and massage therapy was initiated. Despite this conservative treatment, the symptoms worsened. On examination at the General Hospital Trostberg (Trostberg, Germany), the patient was febrile (38°C). The left forearm and back of the hand were swollen. The skin was pale brown with an irregular bosselated surface, tense, and tender to palpation. Perfusion and sensation were normal and there was no lymphadenopathy. The patient had a history of a centroblastic-centrocytic B cell non-Hodgkin’s lymphoma (stage III, grade III a) that was diagnosed in 1998. She had received the last of six cycles of chemotherapy with fludarabine phosphate and cyclophosphamide two months before admission. She had undergone radiotherapy of retrocardiac and iliac lymph nodes. The patient also had colon cancer for which a curative hemicolectomy had been conducted one year earlier.

At the time of admission, the patient was anemic (hemoglobin = 7.6 g/dL) and leukopenic (2,700 cells/µL) with an eosinophilia of 10%. Platelets were in the lower normal range (142,000 cells/µL). The C-reactive protein level was elevated (82 mg/L) and the erythrocyte sedimentation rate was 80 mm/hour. Serumelectrophoresis showed low albumin (50.1%) and immunoglobulin (10.3%) fractions. α1 (11.6%) and α2 (17.6%) fractions were elevated. The IgE level was normal. There were no clinical, radiologic, or laboratory signs of recurrence of hematologic or intestinal malignancy. Ultrasoundography of the limb showed massive edema of subcutaneous tissue and in between muscles and tendons. Multiple, well-defined, cystic lesions were demonstrated intermuscularly. The findings were confirmed by magnetic resonance imaging tomography (Figure 1B).

On the fifth day after admission, the patient developed a massive swelling of the forearm and a lump (diameter = approximately 8 cm) on the back of the hand. Initial radiographs of the forearm were reviewed, and a fracture of the distal radius was diagnosed retrospectively. The patient underwent emergency fasciotomy because of compartment syndrome. Jelly-like tissue containing multiple spherical masses with diameters of up to 2 mm, similar to fish spawns, was discovered and removed from the subcutis, muscles, and tendons by multiple excisions (Figure 1A). Whole-specimen morphologic and histopathologic examination of the tissue showed the larval stage of T. crassiceps. The larvae had a typical appearance with an ellipsoid cystic body, a long and retractable neck, and a single protoscolex. The protoscolex was armed with four suckers and two rows of hooklets. The number and size of the hooklets varied. Additionally, asexual buds at the posterior end of the larvae were observed (Figure 2). Serologic findings for Echinococcus multilocularis, E. granulosus, and Taenia solium were negative. All wounds were left open to heal by secondary intention and flushed twice a day with saline. The patient was given praziquantel (1,000 mg twice a day) and albendazole (400 mg twice a day) as anthelminthic chemotherapy. Praziquantel was administered for 28 days, but albendazole had to be discontinued after 18 days because of leukopenia (600 cells/µL). The wounds healed quickly and the forearm and hand regained their original size and shape. There were no signs of recurrence after a follow-up period of nine months.
When we used a co-culture system originally developed for the metacestodes of *E. multilocularis*[^4^], *T. crassiceps* larvae could be kept in vitro. During a four-month observation period, the markedly vivid and motile larvae grew to a size of approximately 1–1.5 cm, including posterior buds. Moreover, the larvae could be propagated intraperitoneally in Mongolian gerbils (*Meriones unguiculatus*) as previously described[^5^].

DISCUSSION

We describe the first case of a *T. crassiceps* infection in a patient with underlying malignancy and iatrogenic immunosuppression. It is one of the rare reports of a human infection with this parasite (Table 1) and the second case described in Germany[^3^]. In three previous reports[^1^]–[^3^], patients were immunosuppressed because of advanced infection with human immunodeficiency virus (HIV), and the cestode larvae developed subcutaneously and intermuscularly. In the case in this report, the patient’s immune function was impaired because of non-Hodgkin’s lymphoma and antineoplastic chemotherapy. Suppression of the cellular immune response, as in HIV infection, non-Hodgkin’s lymphoma, and antineoplastic chemotherapy, seems to predispose for infection with this parasite. With respect to cestode infections, such an association has only been established for *Hymenolepis nana*[^6^] and possibly *Skrjabinoporus merops*[^6^].

A useful model for studying the immune mechanisms involved in determining the disease outcome has been established in experimental murine cysticercosis with *T. crassiceps* larvae. A Th1-type response is essential for the development of immunity against an experimental infection[^7^]. In humans, *T. crassiceps* has a tropism for subcutaneous and muscular tissues, similar to cysticercosis caused by *T. solium* and coenurosis caused by *T. serialis*. Except for the eye[^8^]–[^10^], no other organ systems were involved in human cases of infection with *T. crassiceps*. In contrast to cysticercosis caused by *T. solium*, there are no reports on infection of human neural tissues with *T. crassiceps*. In the natural rodent intermediate host, *T. crassiceps* metacestodes develop intraperitoneally in the pleural cavity or subcutaneously, where they can multiply by asexual budding. Canids and occasionally felids who serve as definitive hosts accommodate the adult hermaphroditic 7–14-cm long worms in their digestive tract, where they attach to the intestinal wall. Similar to humans, canids can become intermediate hosts and harbor larvae in their visceral or subcutaneous tissues[^11^]–[^12^]. Interestingly, one report describes a visceral infection of a presumably immunocompromised dog[^12^].

Transmission to intermediate hosts is believed to occur via the oral route when fecally contaminated food or water is consumed. In one case, human transmission could be linked to a pet dog[^9^]. In the case in this report, the patient owned a

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**TABLE 1**

Case reports on human *Taenia crassiceps* infections[^1^]

<table>
<thead>
<tr>
<th>Year of publication</th>
<th>Author, (reference)</th>
<th>Context location, (contact)</th>
</tr>
</thead>
<tbody>
<tr>
<td>1998</td>
<td>Maillard (1)</td>
<td>Subcutis and muscle, arm (AIDS patient), France</td>
</tr>
<tr>
<td>1998</td>
<td>Francois (2)</td>
<td>Subcutis and muscle, forearm (AIDS patient), France</td>
</tr>
<tr>
<td>1997</td>
<td>Chuck (8)</td>
<td>Intraocular (subretinal), USA</td>
</tr>
<tr>
<td>1992</td>
<td>Arocker-Mettinger (9)</td>
<td>Intraocular (anterior chamber), Austria</td>
</tr>
<tr>
<td>1992</td>
<td>Klinker (3)</td>
<td>Subcutis and muscle, back (AIDS patient), Germany</td>
</tr>
<tr>
<td>1973</td>
<td>Shea (10)</td>
<td>Intraocular (anterior chamber), USA</td>
</tr>
</tbody>
</table>

[^1^]: AIDS = acquired immunodeficiency syndrome.
stray dog that she had imported from Crete 18 months earlier. Results of examination of the dog’s feces were negative. However, stool examination was conducted only once and the dog was dewormed thereafter as a precaution. Therefore, the source of infection in this case remains uncertain, but the patient had no other known contact with canids or felids. In North America, *T. crassiceps* is one of the most common adult taeniids in foxes and poses a zoonotic risk for the increasing number of immunocompromised humans. Studies in southwest Germany in 1982 demonstrated an infection rate of 24% in foxes and 1% in cats. Hygiene measures and anthelmintic treatment of pet dogs and cats may be useful in controlling the distribution of this parasite.

Interestingly, our patient developed the parasitic lesions after trauma and on the bruised limb, similar to two cases with injured immunosuppressed patients. The reason for this remains unclear. In these two cases, anemia and fever attacks were reported. Our patient received two blood transfusions because of a low hemoglobin concentration. The anemia could be a side effect of the antineoplastic chemotherapy. One report describes coagulation problems leading to suffusion, but neither were detected in our case. The combined antiparasitic chemotherapy with albendazole and praziquantel was empirical. Both drugs were effective *in vitro* and in mice. However, the effectiveness of conservative medical treatment is uncertain. In our case, thoracic surgical excision seemed to be the most appropriate therapeutic approach. Relapses have been reported in two HIV-infected patients in whom a combined antiparasitic chemotherapy was initiated after surgery. A prolonged treatment course (> 6 months) was therefore suggested by Maillard and others. In addition, patients should be followed-up closely. In the case described here, the patient was asymptomatic during a follow-up period of nine months.
REFERENCES


