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Zoonotic Onchocerciasis of the Shoulder

A Case Report

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Investigation performed at the Minneapolis Sports Medicine Center, Minneapolis, Minnesota

Infections caused by *Onchocerca* species, a filarial parasite, other than those caused by *Onchocerca volvulus,* are rare in humans. Six cases of *Onchocerca gutturosa* or *Onchocerca cervicalis* infection in humans have been reported previously[1-5]. These cases occurred in Switzerland, Crimea, Canada, Illinois, and Japan (two cases). We describe the case of a Minnesota resident who had a subdeltoid mass that was caused by *Onchocerca gutturosa.*

Case Report

A fifty-year-old, left-hand-dominant woman presented with a one-year history of a gradually enlarging mass in the anterior part of the left shoulder. She reported a recent increase of pain in the involved shoulder. The patient had no history of injury or unusual medical conditions, and she had not traveled outside of the United States. Physical examination demonstrated a generalized enlargement of the anterior part of the left shoulder. A fluid-filled, subdeltoid mass measuring 4 cm × 5 cm was noted. Both neurovascular function and rotator cuff function were intact. The patient reported pain with range of motion. The Hawkins and Neer impingement tests, consisting of forced forward flexion of the shoulder to 90° and 180° in internal rotation, were both positive. The anterior region of the shoulder was tender to palpation in the area of the mass. Anteroposterior, supraspinatus outlet, and axillary radiographs revealed a mass with a soft-tissue density in the anterior part of the shoulder. No acute or chronic osseous changes were noted.

Coronal, sagittal, and axial proton-density-weighted and T2-weighted magnetic resonance images and coronal fast STIR (short tau inversion recovery) magnetic resonance images revealed a subacromial and subdeltoid soft-tissue mass (Fig. 1). The mass demonstrated increased signal intensity, compared with that of muscle, on the proton-density-weighted images as well as the T2-weighted images. Areas of lower signal intensity were evenly dispersed throughout the mass and were believed to represent fluid with debris.

A definitive diagnosis could not be reached on the basis of the history, physical examination, and imaging studies, and an attempt to obtain a tissue specimen by means of a needle biopsy was unsuccessful. After informed consent was obtained, the patient was taken to the operating room for an open biopsy. An anterolateral incision was made in line with the deltoid fibers. Splitting of the deltoid fibers revealed a fibrous capsular rind surrounding the mass. The fibrous capsule was incised, and a thick, gelatinous fluid with yellow granular debris was noted. An excisional biopsy was performed, and the mass was submitted for histological evaluation. The mass contained a worm, which on initial histopathologic examination was thought to be of the *Dirofilaria* genus. Further review of the slides was performed by members of the Armed Forces Institute of Pathology, who concluded that the morphologic features of the worm were most consistent with those of *Onchocerca gutturosa*.

Histologic examination revealed connective tissue with a caseating granuloma (Fig. 2). The area of necrosis was surrounded by palisading epithelioid cells. Chronic inflammatory cells composed primarily of lymphocytes, plasma cells, and histiocytes were seen, but eosinophils were rarely seen. The adjacent tissue showed perivascular cuffing by lymphocytes and plasma cells. A coiled, degenerated, filarial nematode lay within the area of caseating necrosis.

Fig. 1
T1-weighted axial magnetic resonance image demonstrating the subdeltoid mass.

Fig. 2
Photomicrograph demonstrating a coiled, degenerated worm in the area of caseating necrosis (hematoxylin and eosin, ×30).

The patient recovered uneventfully from the excisional biopsy and, following rehabilitation of the shoulder, resumed her usual activities. Medication for a presumed *Dirofilaria* infection was considered until further review of the slides indicated that no additional treatment was necessary after excision of the mass. The patient had had no further symptoms referable to the *Onchocerca gutturosa* infection by the time of the six-year follow-up examination.

Discussion

Zoonotic *Onchocerca* infection has been rarely observed in humans. *Onchocerca gutturosa* has been found in the ligamentum nuchae of cattle, and *Onchocerca cervicalis* has been identified in the cervical ligaments of horses[2]. The six previously reported cases of *Onchocerca* infection in humans were all believed to have been caused by *Onchocerca gutturosa,* as was the infection in our patient. *Onchocerca gutturosa* infection in cattle has been reported to be prevalent in Louisiana, Georgia, and Minnesota[2]. Our patient reported that she had not traveled outside the Midwest in the few years prior to the onset of symptoms.

The transmission of *Onchocerca gutturosa* involves ingestion of microfilariae by a black fly when it feeds. The microfilariae mature into infective juveniles, which are injected into a human when the fly feeds again. These juveniles mature into adults. The female adults produce microfilariae, which lie in the subcutaneous tissues. These microfilariae are then ingested by another black fly when it feeds, thereby renewing the cycle. No other mode of transmitting *Onchocerca gutterosa* is known to exist.

The six previously reported infections in humans involved the eye-muscle tendons of a fifteen-year-old girl, the knee of an adult male patient, the wrists of three adult female patients, and the foot of a two-year-old girl[1-5]. The infection in our patient involved the shoulder. These anatomic sites are appropriate locations for *Onchocerca gutturosa* infection; whereas *Onchocerca cervicalis* infection usually involves dense, regular connective tissue, *Onchocerca gutturosa* is typically found in the loose connective tissues that surround the tendons[1-5].

The case of our patient demonstrates that unusual diagnoses must be considered in the presence of unusual findings and symptoms. In retrospect, no other tests would have helped us to reach a diagnosis prior to surgery. Thorough pathological evaluation can help the physician to reach a definitive diagnosis when a rare illness is suspected. ■

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