

DISSEMINATED MUSCULAR CYSTICERCOSIS WITH MYOSITIS INDUCED BY PRAZIQUANTEL THERAPY

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Abstract. Cysticercosis, the infestation with the encysted larval stage of *Taenia solium*, is increasingly recognized as a major problem in most countries in Latin America, Asia, and Africa, involving mainly the central nervous system. Muscle involvement is also common, although it usually remains asymptomatic. We describe a case of disseminated muscular cysticercosis followed by myositis (fever, diffuse myalgia, weakness of the lower limbs, and inflammatory reaction around dying cysticerci) induced by praziquantel therapy, an event not described previously.

Cysticercosis, the infestation with the encysted larval stage of *Taenia solium*, is increasingly recognized as a major problem in most countries in Latin America, Asia, and Africa. Within the past 20 years, the development of computerized neuroimaging studies and highly specific serologic tests have led to the recognition of neurocysticercosis as a common cause of neurologic disease worldwide.^{1,2} In Ribeirão Preto, Brazil, the coefficient of prevalence based on compulsory notification is 54 cases/100,000 inhabitants.³

Cysticercosis mainly involves the central nervous system although cysts commonly infect the muscle.¹ However, subcutaneous and muscle involvement in patients with neurocysticercosis is apparently less frequent in the New World.⁴ We describe a case of disseminated muscular cysticercosis followed by myositis induced by praziquantel therapy, an event not described previously.

CASE REPORT

A 26-year-old woman presented with complaints of partial complex seizures, headache, and lower limb weakness of three months duration. On examination, numerous relatively mobile subcutaneous and intramuscular nodules, measuring from 0.5 to 1 cm in diameter, were palpable on the face, trunk, and extremities. There was no enlargement of the limbs but there was paraparesis that prevented walking, with the patient being able to stand only with support. A muscle biopsy showed infestation with living *Cysticercus cellulosae*. On the second day of praziquantel therapy (50 mg/kg/day), the patient presented fever (39°C) and diffuse myalgia, mainly of the lower limbs. Laboratory data included a total white blood cell count of 18,600 (89% neutrophils, 10% lymphocytes, and 1% eosinophils), an erythrocyte sedimentation rate of 62 mm/hr, and a serum creatine kinase level of 117 units (normal = 50–250). Despite the administration of dexamethasone (6 mg/day) and the discontinuation of praziquantel on the seventh day, intermittent fever persisted for nine additional days and myalgia for 15 days. With clinical improvement, therapy with dexamethasone was gradually removed. One week later, a new muscle biopsy revealed cysticerci in the reabsorption stage and the adjacent muscle tissue showed occasional necrotic fibers with mild lymphohistiocytic infiltrates (Figure 1). During a six-year follow-up, the patient continued her anti-epileptic treatment, with no further muscle symptoms.

DISCUSSION

The scanty literature on muscle cysticercosis provides limited information.⁵ To our knowledge, this is the first description of disseminated muscular cysticercosis with myositis induced by praziquantel therapy.

There are reports of massive cysticercal infection of striated muscle with a clinical picture of pseudo-hypertrophic myopathy.^{5–8} The muscle biopsy is usually normal, with no pathologic evidence of denervation, dystrophic change, or hypertrophy,⁷ but Sawhney and others⁵ reported active myositis with changes in muscle fibers apart from an inflammatory reaction around cysts in a patient with pseudohypertrophic myopathy.

Despite the controversy over the clinical outcome,^{9,10} therapy for cysticercosis advanced with the advent of two drugs considered to be effective: praziquantel and albendazole.^{11,12} Most investigators have reported good tolerability of praziquantel.^{13,14} Some patients present some side effects including fever, headache, nausea, vomiting, dizziness, meningismus, increased intracranial pressure,^{11,12} and cerebral infarct. These adverse reactions are probably not due to a toxic effect of praziquantel but rather to an inflammatory reaction produced by the host in response to massive destruction of cysticerci in the central nervous system.¹⁵ It is not surprising that with treatment, degenerating cysts cause myositis; however, in most of the cases, this may be mild and may regress spontaneously. The possibility that some cases may be severe, such as ours, cannot be ruled out, and the fact that it has not been previously noted probably reflect under reporting. Although the muscle biopsy was normal, Wadia and others⁸ reported a painful enlargement of the muscles in patients with disseminated pseudo-hypertrophic cysticercotic myopathy. In our patient, the disintegration of large number of muscular cysticerci by praziquantel therapy may have caused the myositis. Therefore, in patients with massive and disseminated cysticercosis, both in the central nervous system and muscle, we recommend simultaneous administration of high doses of dexamethasone to prevent the deleterious host inflammatory response, even though dexamethasone reduces the plasma concentration of praziquantel.

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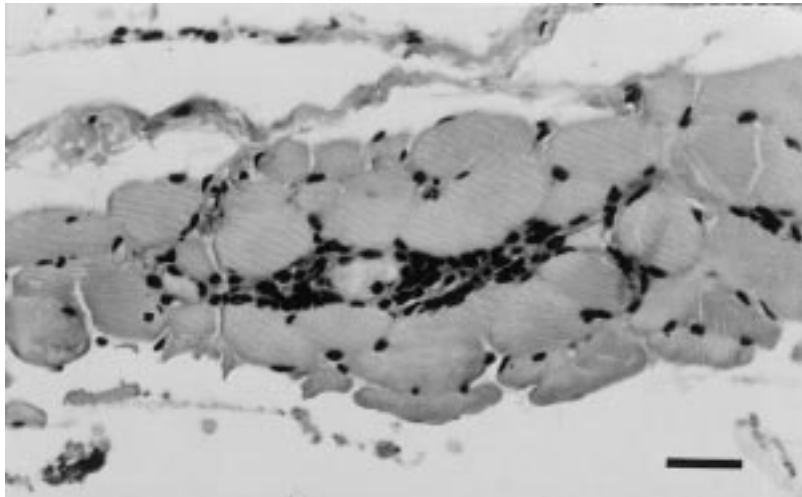


FIGURE 1. Muscle biopsy showing an occasional necrotic fiber with a mild inflammatory lympho-histiocytic infiltrate (hematoxylin and eosin stained, bar = 50 μ m).

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REFERENCES

- White Jr AC, 1997. Neurocysticercosis: a major cause of neurological disease worldwide. *Clin Infect Dis* 24: 101–115.
- Mitchell WG, 1997. Pediatric neurocysticercosis in North America. *Eur Neurol* 37: 124–131.
- Takayanagui OM, Castro e Silva AAMC, Santiago RC, Odashima NS, Terra VC, Takayanagui AMM, 1996. Notificação compulsória da cisticercose em Ribeirão Preto-SP. *Arq Neuropsiquiatr* 54: 557–564.
- Cruz I, Cruz ME, Teran W, Schantz PM, Tsang V, Barry M, 1994. Human subcutaneous *Taenia solium* cysticercosis in an Andean population with neurocysticercosis. *Am J Trop Med Hyg* 51: 405–407.
- Sawhney BB, Chopra JS, Banerji AK, Wahi PL, 1976. Pseudohypertrophic myopathy in cysticercosis. *Neurology* 26: 270–272.
- Armbrust-Figueiredo J, Speciali JG, Lison MP, 1970. Forma miopática da cisticercose. *Arq Neuropsiquiatr* 28: 385–390.
- Jacob JC, Mathew NT, 1968. Pseudohypertrophic myopathy in cysticercosis. *Neurology* 18: 767–771.
- Wadia N, Desai S, Bhatt M, 1988. Disseminated cysticercosis; new observations, including CT scan findings and experience with treatment by praziquantel. *Brain* 111: 597–614.
- Vazquez V, Sotelo J, 1992. The course of seizures after treatment for cerebral cysticercosis. *N Engl J Med* 327: 696–701.
- Carpio A, Santillán F, León P, Flores C, Hauser WA, 1995. Is the course of neurocysticercosis modified by treatment with antihelminthic agents? *Arch Intern Med* 155: 1982–1988.
- Takayanagui OM, 1995. Evaluation of the treatment of neurocysticercosis with praziquantel and albendazole. *Taeniasis/Cysticercosis Complex: Future Trends Toward Its Control*. Brasilia: PAHO/WHO.
- Takayanagui OM, Jardim E, 1992. Therapy for neurocysticercosis: comparison between albendazole and praziquantel. *Arch Neurol* 49: 290–294.
- Bittencourt PRM, Gracia CM, Gorz AM, Mazer S, Oliveira TV, 1990. High-dose praziquantel for neurocysticercosis: efficacy and tolerability. *Eur Neurol* 30: 229–234.
- Leopold G, Ungethüm W, Groll E, Diekmann HW, Nowak H, Wegner DHG, 1978. Clinical pharmacology in normal volunteers of praziquantel, a new drug against schistosomes and cestodes. *Eur J Clin Pharmacol* 14: 281–291.
- Livramento JA, Machado LR, Spina-França A, 1993. Immunobiology of neurocysticercosis. Fejerman N, Chamoles NA, eds. *New Trends in Pediatric Neurology*. Amsterdam: Elsevier, 307–312.