

Multiple cysticercus nodules in skin and brain in a Balinese woman: A case report

Sri S. Margono,* Sutisna Himawan,** Teddy A. Purnama,[∞] Rizal Subahar,* Abdulbar Hamid,^f Akira Ito,^ξ

Abstrak

Dilaporkan kasus seorang perempuan Bali, berumur 33 tahun, agama Hindu dengan nodul multipel sistiserkosis di bawah kulit dan otak. Ditemukan gejala kejang sejak remaja yang kemudian tidak diobati. Sejak tiga tahun sebelum dirawat di rumah sakit, penderita menemukan nodul multipel di dalam kulit, mulai pada dahi dan sejak setahun yang lalu juga di bagian lain dari kepala dan tubuh, seperti pada bahu, dada dan punggung. Ternyata sampel serum yang diperiksa dengan cara immunoblot positif terhadap antigen *Taenia solium*. Hasil tes kopro-antigen yang juga positif memberi petunjuk adanya cacing dewasa *T. solium* di usus. Penderita diterapi dengan praziquantel terhadap infeksi dengan cacing dewasa dan kemudian dengan albendazol terhadap stadium larva, yang berupa kista. Tidak berhasil ditemukan cacing dewasa di dalam tinja 24 jam. Setelah tiga minggu jumlah kista yang teraba di dalam kulit sangat berkurang, demikian juga di dalam otak. Setelah setahun tes immunoblot masih positif. (*Med J Indones 2002; 11: 169-73*)

Abstract

A case of multiple subcutaneous and cerebral cysticercosis in a 33-year-old Balinese female, is reported. The patient suffered from seizures since adolescence, which was not treated. Since three years before admission she started developing multiple nodules in the skin, starting from her forehead and since a year ago also in other parts of the head and body such as shoulders, chest and back. Serum sample tested against cysticercus antigen by immunoblot assay against antigen of *Taenia solium* was positive. The copro-antigen test was also positive, indicating the presence of the adult worm in the intestines. The patient was treated with praziquantel for the adult *T. solium* infection and thereafter with albendazole for the larval stages, which resulted in obvious reduction of the cerebral cysts and most of the subcutaneous nodules disappeared. However the adult worm was not recovered in the 24 hours stool specimen and after one year the immunoblot test was still positive. (*Med J Indones 2002; 11: 169-73*)

Keywords: cysticercosis, *Taenia solium*, praziquantel, albendazole

Cysticercosis, especially neurocysticercosis, is now considered as a common infection, not only in developing countries, but also in countries such as the United States due to a large number of immigrants from Latin America.¹ It is a disease spread all over the world, although the disease is mostly acquired in developing countries. The improvement of diagnostic techniques

and the increase of international travel are contributing to the increase of cases in several areas of the world.

Cases of cysticercosis/neurocysticercosis were found in Bali since years ago.^{2,3,4,5} Ngoerah² reported on four cases of suspected neurocysticercosis. Besides the involvement of the central nervous system, multiple cysts were found in the subcutaneous tissue and muscles. After the imaging technique, i.e. the CT-Scan, was introduced in the General Hospital in Bali, 25 patients suffering from neurocysticercosis could be detected during the years 1995-1997.^{3,4}

Cases of cysticercosis were also reported from Jakarta from time to time.⁶ In this report a case of cysticercosis/neurocysticercosis found in Jakarta is described.

*Department of Parasitology, **Department of Anatomic Pathology and ^fDepartment of Neurology, Faculty of Medicine, University of Indonesia, Jakarta, Indonesia

[∞]Department of Surgery, Mitra Kemayoran Hospital, Jakarta, Indonesia,

^ξDepartment of Parasitology, Asahikawa Medical College, Asahikawa, Japan

CASE REPORT

A 33-year-old Balinese female, religion Hindu, was referred to the Surgical Department of the Mitra Kemayoran Hospital, with complaints of multiple nodules in the skin. Nodules were found all over the body, especially in the skin of the upper part of the body. The nodule on the forehead was known to exist for three years, whereas the other nodules on the head, shoulders, chest and back were detected since one year. She was a teen-ager when she started to suffer from seizures, which was treated. The last attack of seizures was in November 1999. Before the seizures started, she complained of hemianopsia, emesis and a feeling of impending fainting. The duration of the seizures was about half an hour. In March and June 2000 she again had attacks of hemianopsia and was going to faint, however seizures could be avoided. As a toddler the patient sometimes visited her grandmother, who had pigs in husbandry. She had the habit of consuming various dishes of pork.

On May 16th, 2000 several nodules were excised from the forehead, near the right side of the lips, temporal side of the face, neck and shoulder, measuring 1.5 to 2 cm. The pieces of tissue were fixed in 10% formalin solution. The pathologist received seven pieces of tissue, 3 with an irregular texture, each about the size of a corn seed, 2 round pieces of tissue, each with a diameter of 3 mm and 2 pieces of thin walled cysts, which were already opened. The pieces were embedded in paraffin and routine haematoxylin and eosin stained slides were prepared for histopathological examination. The specimen consisted of skin tissue, striated muscle, fatty tissue and connective tissue, infiltrated by chronic inflammatory cells and a few multinucleated giant cells. Cystic structures were observed with thick collagenous walls. One among the three cysts showed an invaginated scolex with several hooklets. Suckers and spiral canals were also observed. Based on these findings the diagnosis of subcutaneous cysticercosis *T. solium* was made.

Plain and contrast computed tomography revealed multiple round hypodense lesions in both hemisphere and right cerebellum, less than 1 cm in size. In the right hemisphere 13 cystic lesions, in the left hemisphere 15 lesions and in the cerebellum 2 lesions were found respectively. In the soft tissue of the right temporal cerebrum 2 lesions were discovered. With contrast imaging no definite enhancement was found. Three and a half months after treatment the CT-Scan showed improvement; only four small cystic lesions,

not more than 0.5 mm in size, were found in the left hemisphere. The extra-cranial lesions at the right frontal side decreased in size. Pathological intra-cranial enhancement was not detected.

The electroencephalogram, before and three months after treatment, showed foci of irritation in frontal, temporal and parietal region of both hemispheres.

A serum sample was tested before treatment by immunoblotting assay with antigen of *T. solium* cysticerci fractioned by single-step isoelectric focusing. Glycoprotein components of the antigen were highly specific and sensitive.⁷ Strong response was found against the antigen, which was a typical result for cysticercosis. One year after treatment a serum sample was examined again and was still positive against cysticercus antigen.

A fecal sample was examined using a commercially available G enzyme Virotech GmbH-approved ELISA-coproantigen kit, for the detection of *Taenia* specific antigen in feces. The result of the coproantigen test was positive, which indicated the presence of the adult worm in the intestines, however segments of the worm could not be found in the feces collected during 24 hours for three consecutive days.

During hospitalization symptomatic treatment for the headache and seizures was given since June 15th with methampyrone, 3 times a day and phenytoin, 3 times a day. At the same time dexamethason, 3 times 1 ampulla was injected on June 15th, tapering to one ampulla on June 17th. Hereafter a tapering dose of oral dexamethasone was given during 30 days. Also on June 17th a single dose of 1050 mg praziquantel, followed by purging with 30 gram magnesium sulphate solution was administered for the intestinal taeniasis.

On June 19th the patient was treated for the multiple cysts with albendazole, 800 mg as a single dose for 10 days. At the same time a single dose of methyl prednisolone 750 mg was injected intravenously. Starting from September 27th carbamazepine one tablet b.i.d. was given daily.

DISCUSSION

The diagnosis of the case was based on neurological signs and symptoms, neuroimaging criteria, laboratory tests, epidemiological history and histo-pathological

confirmation. The patient suffered from cystic neurocysticercosis, with cysts located in the parenchyma of the brain in addition with multiple cysts found in the subcutaneous tissue. The main symptom was general epileptic seizure, starting during adolescence. This symptom is one of the most prominent symptoms and most frequently encountered in neurocysticercosis. In his study Cruz et al.¹⁰ found seizure disorder as the most frequent symptom among 100 patients. Sudewi et al.^{3,4} found epileptic seizures in 68% of 25 neurocysticercosis patients, 24% had cephalgia and 8% had other neurological symptoms.

The patient was initially treated with a single dose of praziquantel, 20 mg per kg bodyweight due to the positive copro-antigen result, which is diagnostic for intestinal taeniasis. Misra et al.¹⁰ treated taeniasis with a single dose of 400 mg albendazole during one or three consecutive days. A cure rate of 57.1% was obtained with a single 400 mg dose as compared to 100% when the same daily dose was given for three consecutive days. We suggest that if a patient suffers from both intestinal taeniasis as well as cysticercosis, the patient should be given praziquantel first for the elimination of the adult worm, before trying to cure the cysticercosis. In the case of intestinal taeniasis praziquantel, which is a fast acting drug, should be used to prevent the possibility of eggs, which partly were not destroyed, to regurgitate to the stomach by retroperistalsis and initiating a cysticercosis infection. In cases of taeniasis without cysticercosis we prefer to use also praziquantel. In the present case albendazole was used for the cysticercus infection. No side effects were found. This drug seemed to be somewhat more effective than praziquantel for cysticercosis.⁷ In general chemotherapy using praziquantel or albendazole in neurocysticercosis was effective in approximately 80% elimination or reducing the cyst numbers and in more than 90% reduction in number of seizures.^{11,12} Different dose and schedules of albendazole and praziquantel have been used with good results. Sotelo et al. administered albendazole at a daily dose of 15 mg/kg of body weight for one month and praziquantel at a daily dose of 50 mg/kg of body weight for two weeks. Steroids were given if adverse reactions to treatment were intense considering recommendations not to use steroids as a preventive measure of adverse reactions.¹³ Simultaneously administration of dexamethasone and praziquantel caused a decrease to 50% plasma levels of praziquantel.^{7,13} In our case dexamethasone was given together with praziquantel and albendazole to avoid side effects.

A twenty-four hours stool specimen was collected from this patient. However, not a single segment of the adult worm was recovered, probably due to several constraints. Praziquantel changed the morphology and color (from whitish to black) of the worm segments because of destruction so that segments were not recognized. Also possibly part of the stool material was thrown away as a result of misunderstanding between the physicians and nurses.

The round hypodense lesions without enhancement on CT-Scan indicated the presence of viable cysts in the brain. Therefore a single dose of albendazole, 15 mg per kg body weight during 10 consecutive days was administered to the patient, which resulted in much improvement, shown on CT-Scan three and a half months after treatment. Four small cystic lesions were still found in the left hemisphere. A decrease of size was noticed of the extracranial lesions at the right frontal side. During the three and a half months the patient had no seizures, headache or other complaints. Most of the subcutaneous nodules on the body and extremities were not detected anymore by palpation. Although much improvement was found after treatment a second course of chemotherapy should be considered.

The serum sample was still positive after one year, which was in accordance with results of other authors. The copro-antigen test was not repeated.

Histopathologically viable cysticerci cause virtually no inflammatory response or only insignificant inflammatory reaction consisting of lymphocytes and plasma cells. However, when cysticerci degenerate, there is an infiltration of various inflammatory cells. In specimens with recently dying cysts, polymorphonuclear leucocytes predominate, while in those specimens with non-viable cysts since a longer period, polymorphonuclear leucocytes as well as lymphocytes and macrophages are found. Eventually, in specimens with cysts undergoing further degeneration, a granulomatous reaction consisting of histiocytes, epithelioid cells and foreign body giant cells develops, leading to fibrosis and calcification.^{14,15} In our case the cysts caused a chronic inflammatory cell infiltration with a few foreign body type multinucleated giant cells.

CONCLUSIONS

A patient with cysticercosis and intestinal taeniasis was first treated with praziquantel for the adult *T.*

solium infection and thereafter with albendazole for the infection of larval stages. The patient suffered from seizures since adolescence age; other symptoms were denied. After three months cysts were much reduced in the brain and subcutaneous nodules were not detected anymore. No side effects were found. The immunoblot was still positive after one year.

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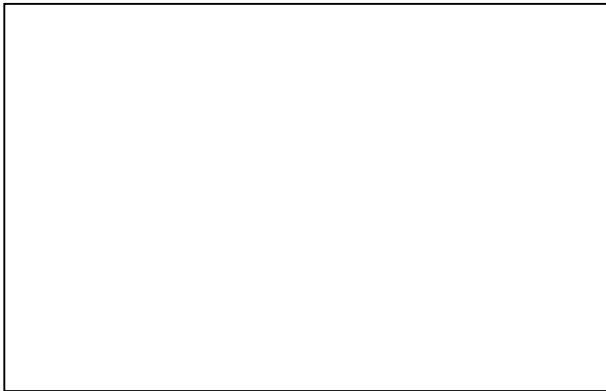


Figure 1. Section of scolex cysticercus *T. solium* showing sucker (Hematoxylin and Eosin staining)

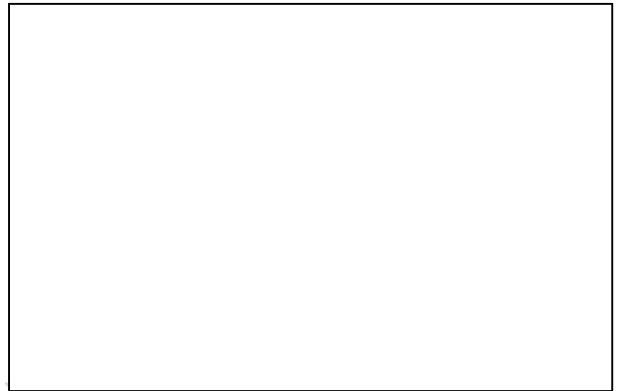


Figure 2. Section of *T. solium* cysticercus skin nodule showing spiral canal (H & E staining)

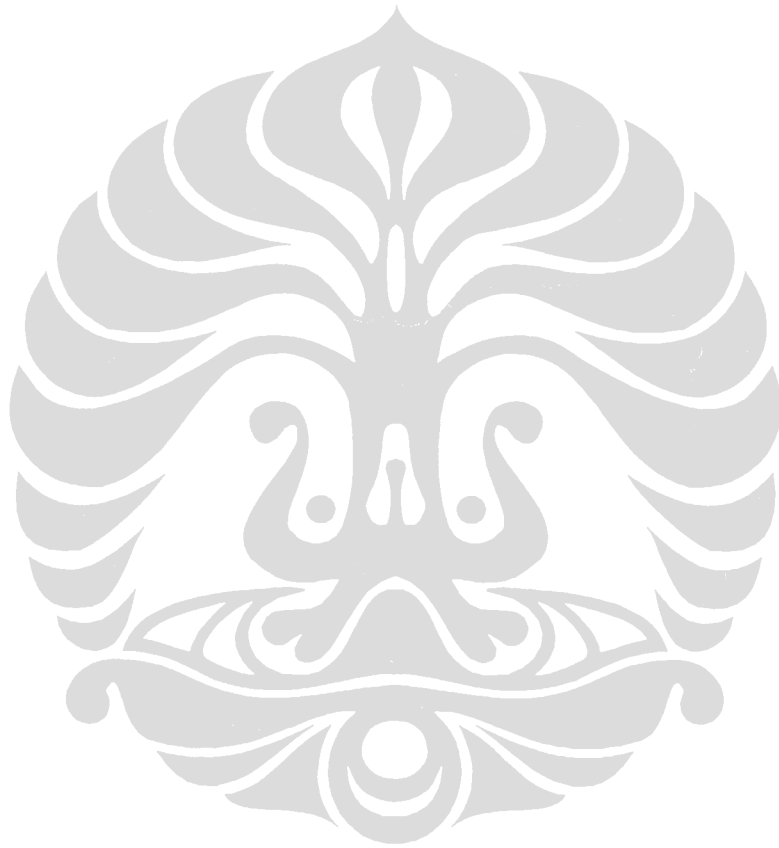


Figure 3. Axial CECT scan shows multiple small cystic lesions in the corticomedullary junction of both upper parietal lobes. No edema is present and no cyst wall enhancement is seen. The picture represents the vesicular stage of infection.



Figure 4. More caudal CECT scan shows non-enhanced small cystic lesions in both occipital lobes; some are located in the left parieto-occipital cortex.





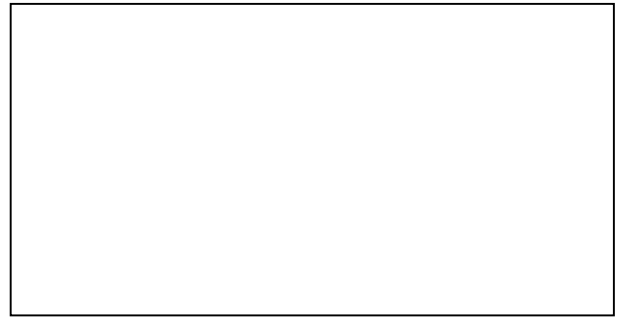
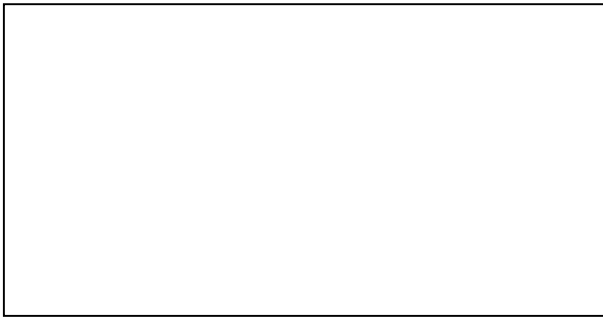


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