CASE REPORT

# Temporary cardiac ischemia in the young adult. A complication of cardiac echinococcosis. A case report

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**Cardiac echinococcosis is an unusual isolated manifestation of the hydatid disease. There are no official reports in Mexico about this incidence. We described the case of a male 19 years old with dyspnea and typical angina. The electrocardiogram showed subepicardial inferior ischemia. The images studies displayed a cardiac cyst in the right ventricle. He was operated on cardiopulmonary bypass and all manifestations of ischemia disappeared immediately after surgery, with a complete success at a two years follow-up.**

***Key words*: Cardiac echinococcosis; Cardiac ischemia, Hydatid cyst.**

**La equinococcosis cardíaca es una manifestación aislada inusual de la enfermedad hidatídica. No hay registros oficiales en México sobre su incidencia. Describimos el caso de un varón de 19 años con disnea, y angina típicA. El electrocardiograma mostró una isquemia inferior subepicárdica. Los estudios de imágenes mostraban un quiste cardíaco en el ventrículo derecho. Fue operado en circulación extracorporea y todas estas manifestaciones de isquemia desaparecieron inmediatamente después de la cirugía, con buena evolución a dos años de seguimiento.**

***Palabras clave:* Equinococosis cardiaca; Isquemia cardiaca; Quiste hidatídico.**

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Hipocrates first described the echinococcosis on the IV century BC. Its worldwide distribution is mainly on rural areas [1]. It is a zoonotic and accidental infection acquired by the human due to the metacestode form of one of the four species of the tapeworm Echinococcus. The Echinococcus granulosus causes cystic echinococcosis, which is characterized by the growth of cysts on the target organ. The cyst is formed by larva that develops on its interior and the larva and germinal epitelium is called hydatid. The cystic or hydatid equinococosis can appear on any organ, mainly on the liver or lungs. Only 10% of cases develop on the rest of the body. Isolated form of the hydatid cyst on the heart is very rare, from 0.5% to 3% of the series [2].

Since the hydatid cysts develop slowly during months or years, most of the patients are asymptomatic. The rupture of the cyst into the heart causes dyspnea, ischemia, arrhythmias, myocardial infarct, tamponade, pericarditis, or sudden death.

## CASE REPORT

A 19-year old male, with no contact to animals nor cardiovascular precedents, he started one month before admission, with dyspnea, initially treated as a pulmonary disease. Afterwards, he presented progressive-effort dyspnea and typical angina-like chest pain. Physical exploration showed no relevant data. The electrocardiogram showed negative symmetric suggestive T waves of subepicardial inferior ischemia (**Fig. 1A**). Transthoracic echocardiogram (TTE) showed a 53x50mm round multiseptated cyst located on the right ventricle (RV) (**Fig. 1B**). The axial computated tomography (ACT) showed a cystic tumoration on the right ventricle (**Fig. 1C**).

The patient underwent surgical procedure, and a small pericardial effusion and multiple pericardial-epicardic adhesions were found. In the cardiopulmonary bypass, with an aortic and pulmonary clamp, a nodular lesion was found in the outer surface of the heart. This lesion had a 70 mm highest diameter in the base of the RV that rests on the diaphragm. In asystole and through the right auricle a nodule protruding 4cm towards the cavity of the RV near the tricuspid annulus was found. The ventricular muscle was intact, suggesting an extra cardiac approach. In this approach, a 3cm tumor bordering the right atrioventricular union in the inferior wall was observed. Subsequently, 17% sodium hypertonic solution was injected into the cystic cavity, and the cyst was opened and drained a pink, thick liquid. A 0.2 mm thick thin wall was

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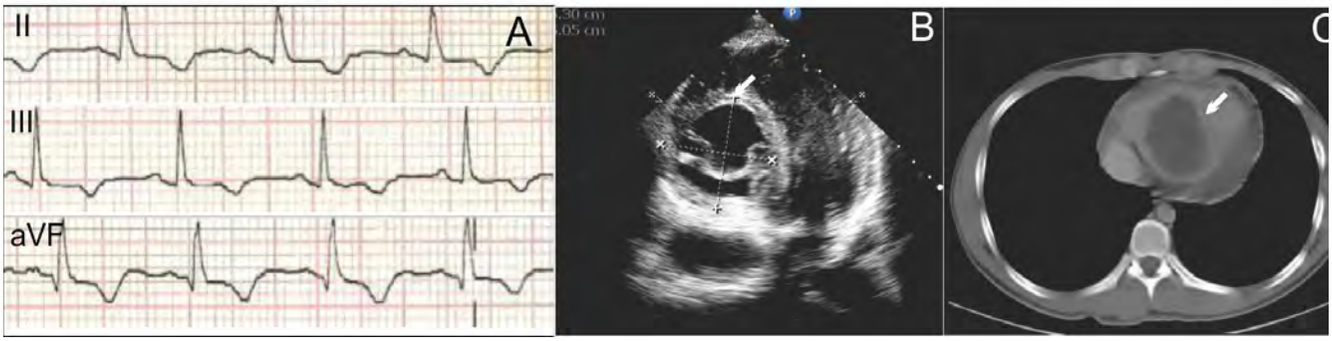


*Figure 1. A= The electrocardiogram showed negative symmetric suggestive T waves of subepicardial inferior ischemia. B= The transthoracic echocardiogram showed a 53x50mm round multiseptated cyst located on the right ventricle. C= The axial computated tomography showed a cystic tumor on the right ventricle and pericardial effusion.*

observed and extracted. The wall was found bordered by a fibrous tissue capsule, which was surrounded by muscle and epicardium and closed by monofilament, teflon felt separate sutures. The right auricle was closed, and the surgical procedure continued in a habitual manner. (**Fig. 2 A**) (**Fig. 2 B**) (**Fig. 3C**). In the immediate postoperative period, Albendazol 400 mg each 12 hours was given to the patient. Hospital discharge was at sixth day after operation. Macroscopic characteristics of the dry specimen showed a thin, white, nacreous, translucent membranous-like, with small nodular nacreous, bulky prominences of fragile consistency (**Fig. 3A**). Histologically a lax fibroconnective tissue laminated wall can be found. Into it, scolex of Ecchinococus granulosus was found (**Fig. 3B**). In the cytological preparation of the cystic cavity liquid, scolex fragments irregularly invaginated were found (**Fig. 3C**). His clinical evolution was good after 4 weeks, and the ECG does not show T inverted waves on the inferior wall (**Fig. 4A**), the TTE (**Fig. 4B**) and the CAT showed no signs of lesions on the RV 2 years after the surgery (**Fig. 4C**).

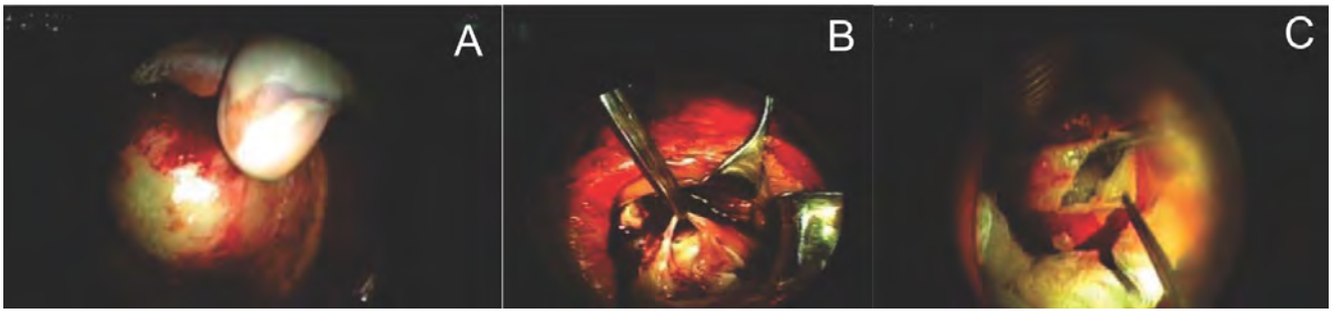


*Figure 2. A = Hydatid cyst in the right ventricle. B= Intracardiac view through the right auricle. The cyst protruding in the base of the rigth ventricule behind the tricuspid valve. C= Extracardiac view of the right ventricle. The hydatid cyst wall opened.*

## DISCUSSION

In this case the course of the disease was insidious, especially in a region where it is not endemic. The WHO shows that the Echinococcus granulosus is present worldwide [3]. However, there are no official records in Mexico about its incidence, and the WHO reports only a sporadic incidence in the northern of the country [4-6]. Cardiac Echinococcus (CE) is present in patients of any age or gender, mainly between 20-40 years-old group, with a 1.7-1 ratio between women and men. 61% of the CE cases are isolated, and the predominant location of the cyst is in the left ventricle (LV) (60%), followed by the interventricular septum (9-20%) and the RV (4-17%). In 75% of all cases, the hydatid cardiac cyst grows subendocardically in the right part of the heart, and subepicardically on the left part [7]. The most dangerous complication of the CE is perforation, which occurs more commonly on LV cysts towards the pericardial cavity. The RV gets pierced less often, but when it does, its perforations occur more commonly towards the RV cavity and are accompanied by embolic, pulmonary complications and sepsis. 75% of the patients with perforations die [8]. The implantation of the CE on the RV has some characteristics of its own: 1) its less frequent implantation is due to the anatomy of the anterior right coronary, which does not easily allow the entrance of the scolexes; 2) in the RV, it is easier for the CE to have an intracavity and subendocardically growth, perhaps because the RV has thinner wall and myocardium, and 3) the RV has less irrigation than the LV. In the heart, clinical manifestations depend of the size and cyst localization. The symptoms are effort dyspnea, non-specific thoracic pain and arrhythmias. In most of the cases, non specific changes in the ST/T segment appear in the ECG. [9].

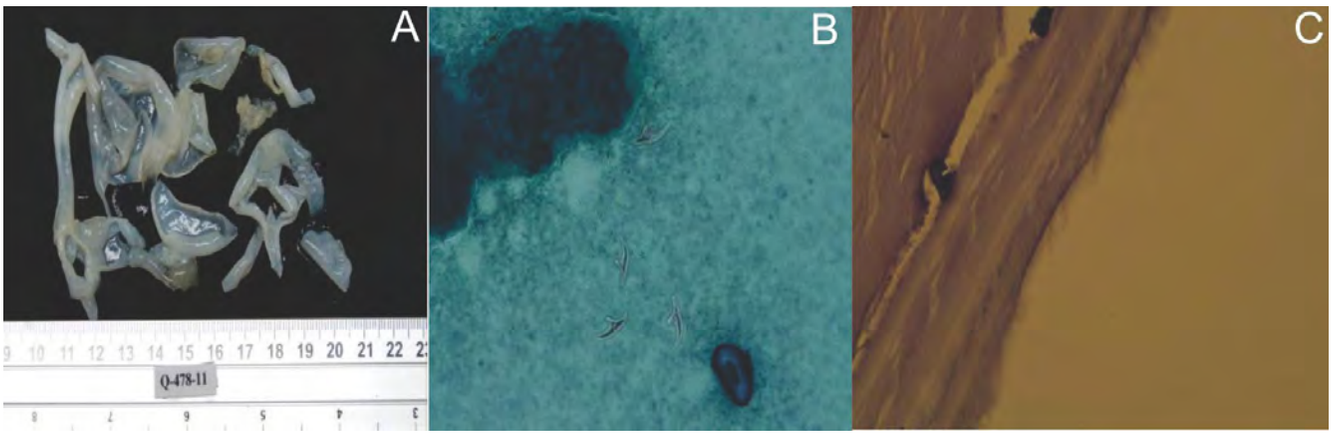


*Figure 3. A= The macroscopic characteristics of the dry specimen show a thin, white, nacreous, tanslucent wall with a membranous aspect where small nodular nacreous, bulky prominences of fragile consistency. B= Material with amorphous clear material with fragmented scolex. C= Laminated wall of fibroconnective tissue with scolex in thickness.*

Other less frequent manifestations are those of pain by ischemia, which are manifested by untreatable angina or acute myocardial infarction without specific electrocardiographic data. Pericardial multiple cysts have been found, causing multiple compressions on the ventricular and auricular cavities, on the coronary arteries and on the venae cava; showing vena cava syndrome or coronary ischemia data [10-12] .

The electrocardiographic data of ischemic cardiopathy are not specific; they may or may not be present. Some reports indicate that they can be appear even when the cyst is the pericardial cavity. To differentiate atherosclerotic coronary disease from CE, an imaging technique of the cyst in the heart is needed; the echocardiography is the procedure of choice. The CE must be included in the differential diagnosis of patients with heart tumors and patients with normal coronary angiography but with angina-like symptoms. Patients with CE are asymptomatic during many years or they can present minor non-specific symptoms that may lead to lethal complications if they remain undiagnosed or untreated [13-15].

Most of the surgical series belong to the endemic areas reported by the WHO, such as Turkey, Syria, Italy and Russia. Due to their load experience, in most of the cases coronary revascularization procedure wasn't necessary because the drainage and decompression of the cyst reduced ischemia. Similarly, other procedures were conducted in special cases like mitral repairs when the cyst was located on the mitral papillary muscle, or other complementary resection of lung cysts [16-17]. The cysts were removed by median sternotomy on extracorporeal circulation by atrial and biatrial approaches, and by left or right ventriculostomy, and in some other cases via trans aortic. Some cysts were operated on without extracorporeal circulation. In this case, once the cyst was disconnected from the RV through extracorporeal circulation, the compression on the right coronary artery disappeared, and the clinical and electrocardiographic data suggestive of inferior ischemia disappeared too. It was showed clearly on the postoperative follow up with ETT and CT without relapse for two years. Therefore, we can say that not all suspected ischemic cardiopathy is atherosclerotic.

In this case, hypertonic sodium solution (17%) was applied into the cyst before we touch it. We clamped the pulmonary

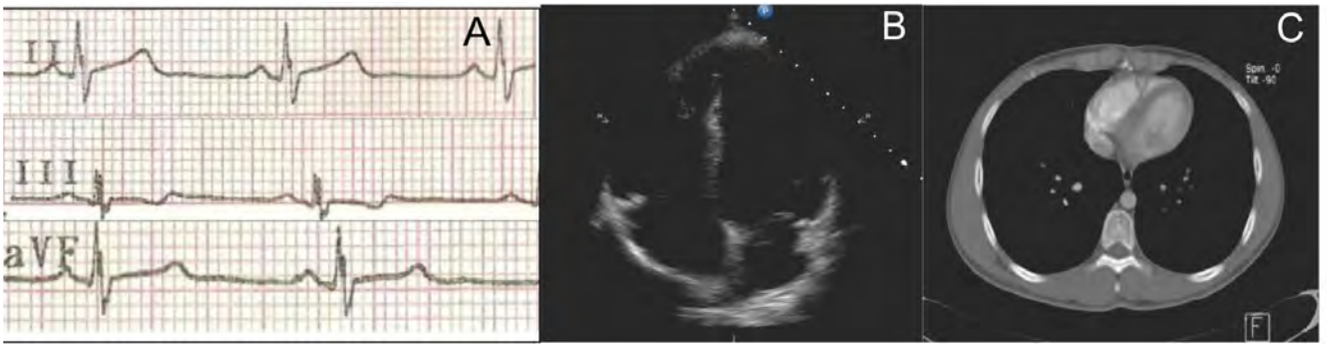


*Figure 4. A= ECG does not show T inverted waves on the inferior wall after surgery. B and C= No lesions in the right ventricle in the postoperative transthoracic echocardiogram (B) and computed axial tomography (C) at 2 years follow-up.*

artery to minimize embolism risk. Other useful solutions that contain alcohol, iodine, and silver have been proposed. Macroscopic and cytological findings of this case confirm the presence of a white, membranous wall of cystic cavity with small, white, bulked prominences that indicate forming cysts (**Fig. 3A**) and material with amorphous clear material with fragmented scolex (**Fig. 3B**) (**Fig. 3C**). It is recommended to begin medical treatment once the cyst has been removed, because to do it before the removal would implicate a higher risk of rupture and embolization. The suggested treatment is 400 mg of anthelmintic albendazol twice a day for six months, beginning the day of the surgical procedure.

As a conclusion, cardiac hydatidosis is a condition that must be kept in mind when making a differential diagnosis of ischemic cardiopathy, especially in young patients. The surgical treatment is the best choice once the diagnosis has been made. This case presented here had an excellent two-year follow up and is one of the few documented cases of CE in Mexico. Due to its behavior and especially the evolution, it is possible to state out that not all suspicion of ischemic cardiopathy means atherosclerosis.

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