Case Reports

Instituto de Anatomia Patológica, Universidad de Los Andes, Mérida, Venezuela; Department of Pathology, Faculty of Medicine, Hradec Králové, Czechoslovakia

Two Cases of Adiaspiromycosis

K. Salfelder, A. Fingerland, M. de Mendelovici and Z. Zambrano P.

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Summary

Pulmonary granulomas with adiaspores were found incidentally in two necropsies in which no gross pulmonary lesions existed. Adiaspiromycosis has not previously been described in Venezuela.

The literature on this newly identified rare deep mycosis in men is reviewed, and the importance of early diagnosis is emphasized, suggesting a lowered threshold of suspicion when granulomas are found.

This pulmonary mycosis, observed first in lower animals, has only recently been described in man.

Adiaspiromycosis forms part of a group of approximately twenty rare deep mycoses, most of which have been identified in the last decade (Emmons et al., 1970; Parker and Klintworth, 1971). As diagnosis is made mainly by pathologists, these new cases are published in this journal rather than in one specializing in mycology.

Case Reports

Case 1. An about 60 year old male of Russian origin who had lived in Mérida/Venezuela for about 30 years was brought to the Hospital in June, 1971. He was comatose and hypertensive and showed hemorrhagic cerebrospinal fluid. He died one day after admission. Autopsy revealed generalized arteriosclerosis, cardiac hypertrophy, and extensive red infarction of the brain. Grossly, no lesions were found in the lungs.

Histology did not reveal lesions in the main viscera. Sections from four blocks of lung tissue were reviewed. In one, a submiliary granuloma with a central cystlike structure measuring 150 μ in greatest diameter and with a capsular wall 17 μ in thickness was noted. The wall was strongly Grocott-
Fig. 1. Case 1. Granuloma with almost empty, thick-walled "cyst" in the center. H & E, x 100.

Fig. 2. Case 1. The cystlike formation shows a thick Grocott-positive wall. Grocott's method, x 260.
positive. In the interior of the cyst nonhomogeneous material was seen. The cyst was surrounded by epithelioid cells and a single multinucleated giant cell. Toward the periphery, the granuloma showed collagen fibers in laminated arrangement; lymphocytes were also present (figs. 1, 2). In serial sections of this block and others some partly scarred granulomas with deformed cystlike structures were observed.

Case 2. A 45-year-old Venezuelan woman from the neighborhood of Mérida was autopsied in September, 1971, to determine the cause of her sudden death. Grossly, a moderate cardiac hypertrophy was found with grayish nondescript foci in the myocardium. The lungs did not show any lesions. Marked chronic myocarditis without parasites was seen histologically in many sections. In Venezuela this type of myocarditis is considered by many people Chagas's myocarditis, despite the lack of leishmanias of Tr. cruzi in the myocardial tissue. Other viscera did not show any lesions.

In one of four pulmonary sections examined a granuloma was observed which showed a structure similar to that described in case 1. Acidophilic and homogeneous masses, possibly due to necrosis or hyalinosis, were observed in central parts around the fungus cell. In serial sections the granulomatous reaction was clearly seen, as were laminar structures of the wall and irregularities of the internal and external surface of the wall of
he fungus cell (figs. 3-7). No further granulomas were found in sections of the other lung tissue blocks.

Discussion

The large, spherical cystlike structures in the pulmonary granulomas of the two foregoing cases are thought to be adiaspores of Emmonsia cressens. Their size, the Grocott-positive thick wall with laminar membranes and irregular surfaces, the lack of endo- and exospores, and the tissue reaction point in this direction. The cystlike formations show certain similarities to spherules of Coccioides immitis and Rhinosporidium seeberi, but without endospores. The fungus cells and tissue reaction are identical with those in the case of KODOUSEK et al., 1971, which was studied for comparative purposes.

Fig. 4. Case 2. In serial sections, the cystlike formation of the granuloma shows a thicker wall in the central part and irregularities of the inner and outer surface. Grocott's method, X 275.

Fig. 5. Case 2. The cyst wall shows a wavy surface. Grocott's method, X 1,150.
In addition no internal structures were seen in the majority of adiaspores of our case.

The diagnosis in these two cases was made exclusively on morphologic grounds. Since there were no gross pulmonary lesions, lung tissue had not been cultured. In any case, positive cultural results have been obtained only in recent infections of lower animals. In humans and in chronic infections of lower animals fungus cells could not be cultured.

Regarding differential diagnosis, nonliving foreign bodies, parasites, or eggs of parasites must be taken into consideration. However, not all nonfungal structures show the strong Grocott-positivity of the wall. Probably, up to now, the reason that so few cases have been found in men is that lung tissue is histologically examined only when grossly visible lesions are detected.

The mycotic infection in our first case was more recent than in the second case; calcification had not yet occurred. In the first case, several granulomas, and in the second, only a single granuloma were found. The lack of multiplication of fungus cells in the tissue is probably the main obstacle for extrapulmonary dissemination. Multiple pulmonary granulomas apparently are due to inhalation of many adiaspores and not to secondary intrapulmonary spread.

Adiaspiromycosis has been described in the lungs of wild rodents, first in Transcaucasia and later in Arizona/USA (Kirschchenblat, 1939; Emmons, 1942; Emmons and Ashburn, 1942). The causative agent was called Rhino-
sporidium pulmonale in the USSR and Haplosporidium parvum in the USA. In 1959, the large cyst-like fungus cells in the lungs were named Emmonsia (Cifferri and Montemartini, 1959). In 1960, they were called adiaspores, and the mycosis adiaspiromycosis (Emmons and Jellison, 1960). Botanists call the fungus Chrysosporium (Carmichael, 1962). From 1942 on, these fungi have been found worldwide in numerous small mammals and in soil samples (Jellison, 1950; Jellison and Vinson, 1961; Parker and Klintworth, 1971). Four species were differentiated on the basis of size, thickness of wall, number of nuclei, and behavior of fungus cells in cultures: E. crescens, E. parvum, E. brasiiliensis and E. cifferina (Parker and Klintworth, 1971); however, only the first two mentioned are recognized by Emmons (Emmons et al., 1970).

Ten cases have been described since 1963 in men in five countries (Brazil, Czechoslovakia, France, Honduras, and India) (Batista et al., 1963; Batista et al., 1963; Chevrel et al., 1964; Doby-Dubois et al., 1964; Misra et al., 1966; Cueva and Little, 1971; Kodeusek et al., 1971; Fingerland and Vortel, in press; Slais et al., in press).

Adiaspiromycosis has not been the cause of death in any of the described cases, including the two presented in this paper. However, multiple granu-
lomas with adiaspores are assumed to lead to pulmonary insufficiency. In any event, this new lung mycosis should be better known. In cases of disseminated miliary granulomas, biopsy may lead to early diagnosis and hence to a generally favorable prognosis.

References


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Professor Dr. K. Salfelder, Departamento de Patologia, Universidad de Los Andes, Apartado 75, Mérida, Venezuela, S.A.