[Jap. J. Parasit., Vol. 32, No. 6, 501-508, December, 1983]

A Human Case of Eurytremiasis: Demonstration of Adult Pancreatic Fluke, *Eurytrema pancreaticum* (Janson, 1889) in Resected Pancreas

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(Received for publication; August 1, 1983)

Key words: human eurytremiasis, pancreatic fluke, *Eurytrema pancreaticum*, pancreatectomy, pancreatic carcinoma

Eurytrema pancreaticum (Janson, 1889) is a parasite commonly found in the pancreatic and biliary passages of cattle, hogs, sheep, goats, and waterbuffaloes mostly in Asian and South American countries. Several cases of human eurytremiasis have been also reported (Castellani and Chalers, cited by Faust *et al.*, 1970; Chang and Li, 1964; Asada *et al.*, 1966; Saito *et al.*, 1973; Ishii *et al.*, 1983), although diagnosis of these human cases was mostly made depending on the size and morphological features of the expelled eggs except for the two cases reported by Chang and Li (1964) and Ishii *et al.* (1983) who demonstrated adult specimens by a postmortem examination.

This paper describes a case of eurytremiasis in human pancreas which was resected as a diagnosis of pancreatic carcinoma.

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Case of human eurytremiasis

A 57-years-old Japanese female farmer living in Notsuharu, Oita, was admitted to the Hospital of Medical College of Oita, on 24 December, 1982. She complained hypochondralgia during the past three months. With a diagnosis of pancreatic carcinoma, she had a pancreatectomy on 14 January, 1983. Histopathological examination of the resected pancreas showed no evidence of malignant neoplasm, but discovered three flat fluke worms in the dilated pancreatic duct. After operation, the patient soon restored her health, and left the hospital on 10 February, 1983.

Morphology and identification of the parasites

The parasites submitted for identification were already fixed in 10% neutral formalin. The morphological features of these fixed specimens observed under a dissecting microscope are as follows: These three specimens (no. 1 and no. 2, partially injured near the posterior regions; and no. 3, cut off into two parts) resemble one another by the general appearance and creamy body color. They are all

Table 1 Measurements of the whole body, oral and ventral suckers, and eggs of three pancreatic fluke worms

Specimen No.	Whole body (mm)		Oral sucker (mm)		Ventral sucker (mm)		Eggs in uterus (μm)		
							No. examined	Length	Width
	Length	Width	Length	Width	Length	Width		Mean±SD*	Mean±SD*
1	15.0	5.5	2.5	2.3	1.7	1.8	21	54.47±2.49	34.12±1.85
2	15.8	5.2	2.5	2.2	1.7	1.7	22	54.13±2.19	34.47±1.81
3	15.6	6.4	2.4	2.4	1.8	1.6	14	51.81±2.25	32.47±1.30

* SD=Standard deviation.

characterized by the presence of the large oral and ventral suckers, the ventrally-everted projection between these two suckers (i.e. cirrus organ), and the conical caudal tip (Fig. 1).

All three worms were first immersed in distilled water, and then flattened ventro-dorsally between two slide glasses in 70% ethanol (Fig. 2). The size of the whole body and suckers were measured from these compressed specimens, and eggs taken from their uterus were also measured (Table 1).

One of these three specimens (no. 1) was further stained with borax carmine, dehydrated by the graded series of ethanol and then mounted with permount. The following morphological features were based on this stained specimen (Fig. 3): The whole body of the fluke measures 13.6 mm in length by 5.0 mm in breadth. It is flattened to a fusiform, with a conical caudal process and slightly ruffled margins. The oral sucker is large, measuring 2.2–2.4 mm in diameter, and is subterminally positioned. The esophagus is short, and two ceca terminate at some distance from the posterior end. The ventral sucker (1.6–1.7 mm in diameter) is smaller than oral sucker and is situated somewhat anterior to the middle of the body. The cirrus pouch is elongate, running obliquely towards the acetabulum. The genital pore is postbifurcal in position. The cirrus organ is everted far outside the genital pore. The ovary is a small, notched organ, lying submedially at three-fifths of the body distant from the anterior tip. The vitellaria are composed of small, finger-like follicles, occupying lateral fields along ceca in three-fourths of the body. The uterus is intricately coiled, occupying most posttesticular region medial to the ceca. It loosely ascends up to the genital pore. The eggs in the uterus are mostly ellipsoidal to ovoid in shape and are thick-shelled with a distinct operculum. They are deep golden brown in color and are generally embryonated (Figs. 4–6).

These characteristics mentioned above conformed to those of the genus *Eurytrema* Looss, 1906, as defined by Yamaguti (1958). And from the size of the worms as well as the proportional size of the oral sucker to the ventral one, these fluke parasites were identified as adults of *E. pancreaticum* (Janson, 1889), rather than *E. coelomaticum* (Glard and Billed, 1892), both of which are known to be distributed in Japan (Yamaguti, 1958). Furthermore, this identification was confirmed by comparing with specimens of the same fluke species from cattle slaughtered in Oita abattoir.

Histopathological examination of the resected pancreas

The resected pancreas is elastic soft in consistency and there are multiple foci of small fatty necrosis in the peripancreatic adipose tissue. The pancreatic duct was opened before fixation. It shows moderate degree of stenosis at the portion of 5 cm

from the pancreatic tail and dilatation of the peripheral duct. Three parasites were observed in the dilated pancreatic duct. The mucosa of the pancreatic duct is pale in color.

On multiple cut surfaces across the pancreatic duct, the pancreatic parenchyma is light yellow in color and appeared to be atrophic. Five specimens were taken from the resected pancreas for histological examination.

Histologically, the pancreatic parenchyma, both pancreatic acini and islets of Langerhans, shows no remarkable changes. There are many foci of fatty necrosis here and there (Fig. 8). The mucosa of the pancreatic duct is unremarkable except for the portion of stenosis in which the epithelium shows squamous metaplasia (Fig. 9). Focal infiltration of inflammatory cells consisting of lymphocytes, plasma cells, monocytes and eosinophilic leukocytes, and increased collagen fibers are observed in the wall of the stenotic duct.

Laboratory examination of the blood, urine and fecal samples

In seven preoperative examinations, the patient's blood was shown to be normal. For example, eosinophils averaged 2 or 3% with a single exception of 7% on 27 December, 1982. The value of Ig G which was measured once only (on 27 December, 1982) was within normal range (i.e. 1300 mg/100 ml). On the other hand, serum amylase was increased, ranging 171 U/l to 198 U/l (average, 189 U/l). However, it decreased rapidly to 151 U/l three days after operation, and continued to be almost the same level (148–158 U/l) until the day when she was allowed to leave the hospital, although it rose up to 180 U/l on 24 January, 1983.

The patient's urine and fecal samples examined twice before operation were found normal. Unfortunately, parasite's eggs in the feces were not searched before operation.

Follow-up stool examinations of the patient and her family (husband and son) were negative, but stool sample of the cow raised by this family was found to have eggs of *E. pancreaticum* (average, 51.8 μm long $\times$ 30.0 μm wide, see Fig. 7).

Discussion

A majority of *E. pancreaticum* infections in man have been diagnosed based on the size and shape of the eggs expelled in the feces, although Chang and Li (1964) and Ishii *et al.* (1983) found adult worms of this parasite at autopsy. In our case three worms were accidentally recovered in the resected pancreas from the patient with a preoperative diagnosis of pancreatic carcinoma. All these worms were found to be matured, and were identified as *E. pancreaticum* by their morphological characters.

The morphology and maturity of these three specimens (Fig. 3) were essentially similar to those of specimens recovered from cattle. However, these worms seem to be somewhat longer than the adult specimens detected by a postmortem examination (12–13 mm by Chang and Li, 1964; 10–11 mm by Ishii *et al.*, 1983). In both previous cases large numbers of adult worms were found in the pancreatic duct, e.g. 43 specimens by Chang and Li (1964) and "many specimens" by Ishii *et al.* (1983) (although the accurate number of parasites were not mentioned by the latter authors). Therefore, the difference in the body size may be attributed to the number of the worms parasitizing the pancreatic duct, although it is also considered that the body size of the parasite varies depending on the duration elapsing after ingestion of metacercariae. Similarly, uterine eggs of these three specimens seemed to be slightly longer than those (average, 47.06 μm) reported by Ishii *et al.* (1983) and the eggs

expelled in human faces (47–49 μm by Asada *et al.,* 1966, and an average of 48.3 μm by Saito *et al.,* 1973).

With regard to the histopathological examination of the pancreas infected by the parasites, Chang and Li (1964) reported dilatation of the pancreatic duct and chronic proliferous inflammation in the wall of the duct. Ishii *et al.* (1983) observed fatty infiltration into pancreatic parenchyma as well as dilatation of the pancreatic duct. In the present case, pathological findings were essentially similar to those of these reports, and the most conspicuous change was observed in the pancreatic duct which showed stenosis caused by squamous metaplasia of overlying epithelium and infiltration of inflammatory cells associated with mild fibrosis in the submucosa. These changes were considered to be resulted from chronic irritation by the parasites. There were many foci of fatty necrosis in the pancreas and peripancreatic fatty tissue. However, granulomatous lesions which were often seen in the usual host (Basch, 1966) were not observed.

According to the previous reports (Asada *et al.,* 1966; Saito *et al.,* 1973), no clinical symptoms have been exhibited in the patients infected by *E. pancreaticum* at all. In this connection, it is noteworthy that the patient reported herein complained the abdominal pain probably caused by stenosis of the pancreatic duct.

Laboratory findings showed that serum amylase was high (171–198 U/l) before operation but decreased to the normal range thereafter. This may indicate that the increase of serum amylase was associated with a stenosis of the parasitized pancreatic duct. Thus, measurement of this enzyme will be of some diagnostic value for the infection of *E. pancreaticum* if followed by a careful fecal examination, especially in areas endemic for bovine eurytremiasis, although increased amylase is known to be generally caused by pancreatic lesions.

The infection of cattle with *E. pancreaticum* is common in Kyushu (Itagaki, 1979). Our examination showed that the cow raised by the patient's family was infected with the same fluke species. Moreover, orthopterous insects belonging to *Conocephalus* and *Oecanthus,* some of which were reported to act as a second intermediate host of *E. pancreaticum* elsewhere (Basch, 1965; Nadikto and Romanenko, 1969), are known to occur in southern Japan (Itagaki, 1979). Under these circumstances, there will be a possibility that metacercariae of the pancreatic fluke are ingested by men, especially by those who are engaged in pasturage. However, no insectivorous habits are seen among the inhabitants in Oita, including the patient in the present case. It may be, thus, probable that the infection of men with *E. pancreaticum* is usually accidental, although the actual mode of infection remains to be studied yet.

Summary

A human case of eurytremiasis was reported. Three worms were recovered in the pancreatic duct resected from a 57-years-old Japanese female farmer from Notsuharu, Oita. All three parasites were identified as adults of *Eurytrema pancreaticum* (Janson, 1889) and their morphology and size including uterine eggs were found normal.

Histopathological examination of the resected pancreas revealed that the pancreatic duct was somewhat distended in the peripheral portion where parasites were present, and there was a stenosis at 5 cm from the pancreatic tail so that the parasites were confined behind this constricted site.

Laboratory findings showed that serum amylase of the patient was high (171–198 U/l) before pancreatectomy.

Acknowledgements

The authors wish to express their gratitude to Prof. I. Nakayama, Department of Pathology, Medical College of Oita, for his valuable comments on the manuscript. We are also grateful to Prof. P. Luh, Institute of Microbiology and Epidemiology, Beijing, China, for his kindness in sending us a copy of the relevant paper on a human eurytremiasis case in China.

Thanks are due to Drs. K. Nakahata and T. Sasaki, Meat inspection office of Oita prefecture, who kindly gave us specimens of pancreatic flukes from cattle for comparison.

Finally, we wish to thank Mr. S. Yano, Department of Pathology, Dr. M. Baba, Miss. C. Aoki and Miss. K. Ogata, Division of Medical Zoology, Medical College of Oita, for their assistance in this work.

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膵蛭成虫の人体寄生例

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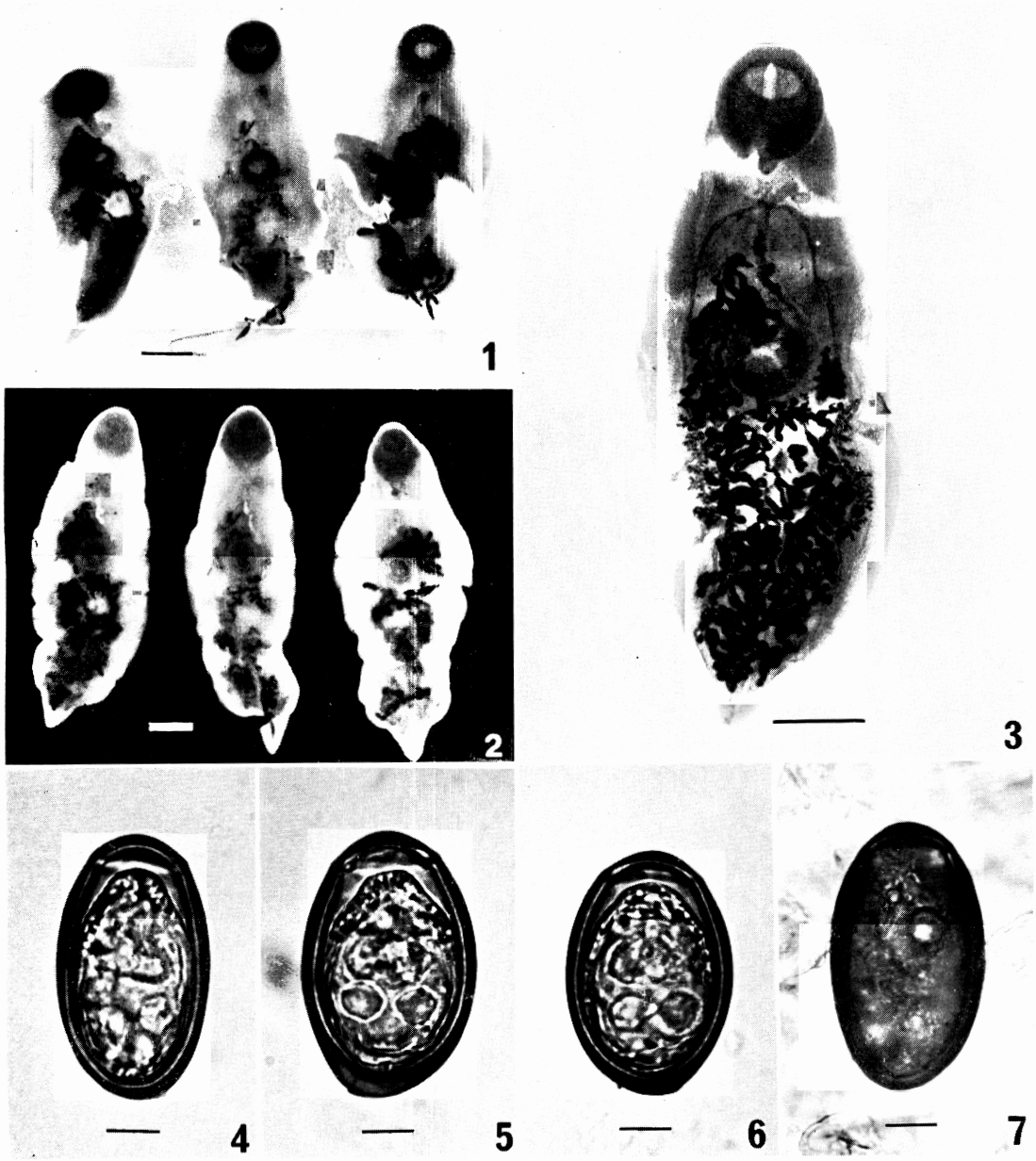
膵臓癌の疑いで膵臓摘出手術を受けた患者の切除部の膵管より膵蛭の成虫体が見いだされたので，本吸虫の人体寄生例として報告した．患者は大分県大分郡野津原在住の57歳の女性で，術前に左側季肋部腹痛を訴え，膵臓癌の疑いで手術を受けたが，悪性腫瘍の所見は認められず，術後27日目に退院した．虫体は膵管末尾近くに3個体発見された．虫体および子宮内の卵の形態的特徴と大きさなどから，膵蛭 *Eurytrema pancreaticum* (Janson, 1889) と同定された．摘出膵臓の組織病理学的所見では，実質部に著変はなく，寄生膵管の拡張と一部膵管上皮細胞の扁平上皮化生による膵管の狭窄が観察された．また，この狭窄部の膵管壁には炎症性細胞の局所的浸潤が認められた．血液の臨床検査では，術前に血清アミラーゼの上昇がみられた以外は正常であった．



Explanation of Figures

Fig. 1 Adults of *E. pancreaticum* preserved in 10% neutral formalin (specimen no. 1, 2 and 3 from left to right). Ventral view. Scale, 2 mm.

Fig. 2 Adults of *E. pancreaticum* compressed and fixed in 70% ethanol (specimen no. 1, 2 and 3 from left to right). Ventral view. Scale, 2 mm.

Fig. 3 Adult of *E. pancreaticum* stained with borax carmine (specimen no. 1). Ventral view. Scale, 2 mm.

Figs. 4–6 Eggs in the uterus of *E. pancreaticum* recovered from the patient. Scale, 10 μm.

Fig. 7 Egg of *E. pancreaticum* found in the fecal sample of the cow raised by the patient's family. Scale, 10 μm.

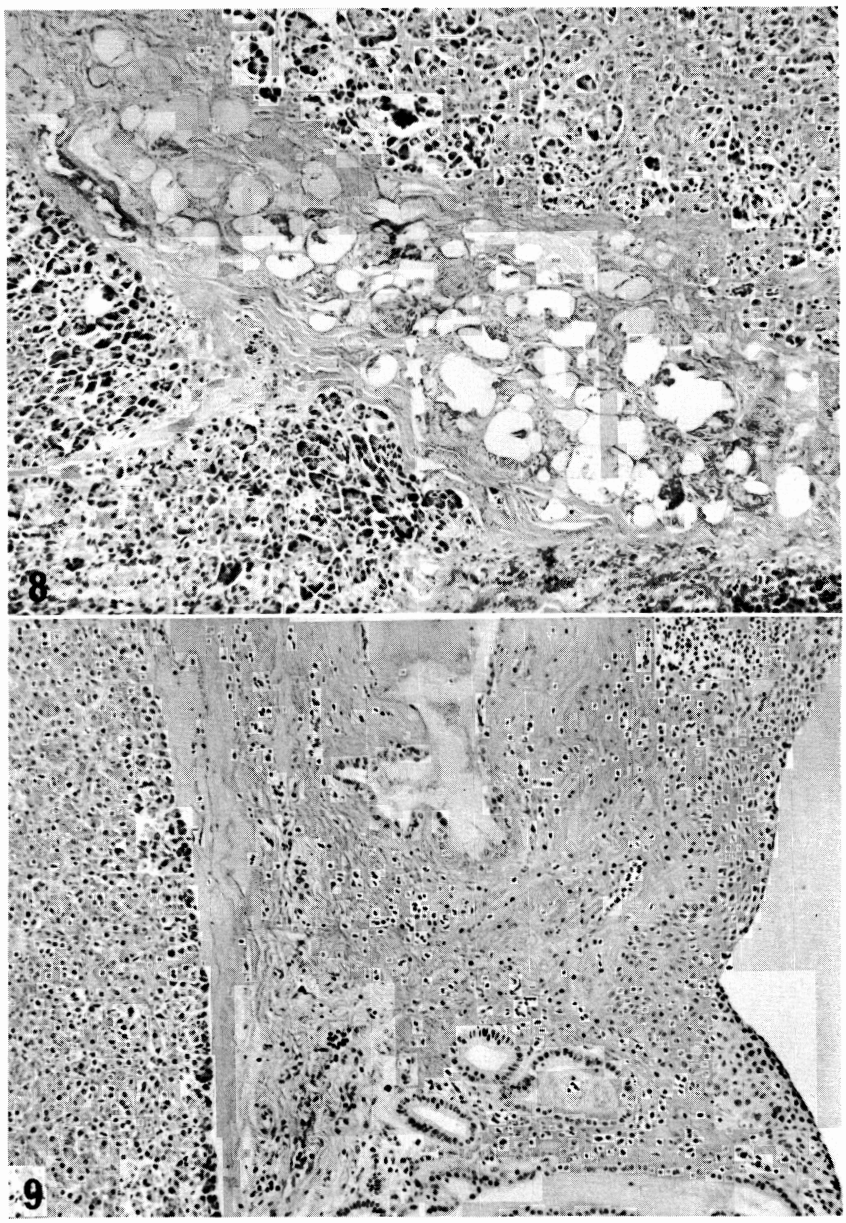


Fig. 8 Fatty necrosis in the interstitial tissue. H. E.-stain. ×700.
Fig. 9 Squamous metaplasia and inflammatory cell infiltration in the stenotic duct. H. E.-stain. ×700.