

Ineffectiveness of Praziquantel Treatment for Human Sparganosis (A Case Report)

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Abstract—A male Korean farmer, who had received excisional operation on subcutaneous masses due to sparganosis, showed reappearance of subcutaneous masses at other sites. He was given praziquantel orally at the dose of 75 mg/kg/day for 5 days and again after one month. However, the masses did not regress and the patient complained of discomfort for the next 4 months. Finally the masses were excised and 3 actively moving spargana were recovered. In this case of human sparganosis with multiple lesions, praziquantel was not effective.

Key words: *Human sparganosis, Praziquantel, Subcutaneous mass*

INTRODUCTION

Praziquantel is a broad-spectrum anthelmintic against various trematode and cestode infections. It causes rapid contraction of worms and extensive vacuolization of their tegument and/or parenchymal layer which results in their death (Andrews *et al.* 1983). This drug is especially useful for its therapeutic effect on dermal and cerebral cysticercosis caused by the metacystode of *Taenia solium* (Rim *et al.* 1982).

In sparganosis, another larval cestode infection in human tissues (Chi *et al.* 1980), however, surgical removal is so far the only effective therapy. No chemotherapeutic trial has been reported successful (Moulinier *et al.* 1982). We also report here a clinical experience of unsuccessful treatment of human sparganosis with praziquantel.

CASE DESCRIPTION

The patient, a 55-year-old male farmer resid-

ing in Yongin-gun, Gyeonggi-do (Province), visited a local clinic in December 1984 because of several painful masses on his lower abdominal wall and right axillary area. Besides pain and itching at the lesions, the patient complained of severe fatigue. He said he had eaten raw snakes during the past 20 years. Sparganosis was suspected and excisional operation was done on the masses. From the excised masses 3 living spargana were recovered. Hence, this patient was thought to be completely cured.

Three to 4 months after surgery, however, the patient complained of reappearance of 3 thumb-sized masses on the left flank and right axilla. He visited Outpatient Department of Seoul National University Hospital (SNUH). He was seen in consultation by the Department of Parasitology, SNU for the feasibility of chemotherapy. Serological test by micro-ELISA (enzyme-linked immunosorbent assay) for sparganum-specific antibody (Kim *et al.* 1984) revealed a high serum antibody level of absorbance (abs.) 1.06 (positive criterion: abs. 0.22).

The patient was treated with praziquantel for his disease. He was dosed with 75 mg/kg (in 3 divided doses) for 5 days, and again after one



Fig. 1. Three spargana (arrow: scolex) recovered from the patient 4 months after the treatment with praziquantel. They were all alive when removed surgically.

month. During 4 months of follow-up observation, the masses did not regress significantly, although they became a little softer to palpation. He continuously complained of unrelenting pain and occasional itching sensation around the masses. At 4 months post treatment serum antibody titer in micro-ELISA for sparganosis was abs. 1.35, which was not a decreased level. Finally the masses were surgically removed. Each mass contained a live sparganum (Fig. 1) of which motility was significant.

DISCUSSION

The present trial of praziquantel treatment in a case of human sparganosis with multiple lesions resulted in failure. Most sparganosis patients are infected with a single worm and less than 30% of patients have multiple infections (Cho *et al.* 1975). Therefore, this rare case with multiple lesions was suitable for the drug evaluation.

A similar result was reported in a human case of proliferative sparganosis, a fulminating systemic disease caused by branching spargana (Moulinier *et al.* 1982). They used mebendazole and praziquantel alternately, but reported unsatisfactory results with both drugs. In experimental mice, administration of praziquantel was found ineffective for the treatment of sparganosis (Lee *et al.* 1986).

An interesting finding *in vitro* was that the spargana incubated in solutions containing

0.1-100 $\mu\text{g/ml}$ praziquantel were immobilized and severely destroyed especially at their neck portions, looking as if the whole worm was dead and disintegrating (Lee *et al.* 1986). However, they (Lee *et al.* 1988) successfully infected mice with scolices of the damaged spargana. The spargana were completely regenerated to reveal scolex, neck and body in experimental mice.

The failure of praziquantel treatment for sparganosis may have been due to inadequate dosage or bioavailability of the drug. It is known that the therapeutic dosage of praziquantel for the treatment of tissue parasites should be generally much higher than those for intestinal ones (Andrews *et al.* 1983; Lee and Chai 1985). For example, as much as 500-750 mg/kg in total dose is required for muscular or cerebral cysticercosis, while only 10-15 mg/kg single dose is highly effective for intestinal trematode or cestode infections (Andrews *et al.* 1983). In the present case, though two courses of treatment (at 1 month interval) each with 375 mg/kg dose were tried, the result was poor. It seems worthwhile to investigate whether even higher doses can bring about a successful result.

The resistance of sparganum to praziquantel is hard to explain. It may be of similar nature to those shown by other helminths, namely nematodes, trematodes such as *Fasciola hepatica*, and larval cestode such as hydatid cyst (Andrews *et al.* 1983). It would be interesting to investigate any similarity in the tegumental or muscular structures. Thick tegument may interfere with drug absorption, as in sparganum, hydatid and *Fasciola*, that are refractory to praziquantel.

Anyway, recent findings of relatively frequent cerebral involvement by sparganum (Chang *et al.* 1987) stress a necessity of searching for effective drugs for this disease.

REFERENCES

- Andrews P, Thomas H, Pohlke R, Seubert J. Praziquantel. *Med. Res. Rev.* 1983, 3: 147-199
- Chang KH, Cho SY, Chi JG, Kim WS, Han MC, Kim CW, Myung H, Choi KS. Cerebral sparganosis: CT characteristics. *Radiology* 1987, 165(2): 505-510
- Chi JG, Chi HS, Lee SH. Histopathologic study on human sparganosis. *Korean J. Parasit.* 1980, 18: 15-23
- Cho SY, Bae J, Seo BS, Lee SH. Some aspects of human sparganosis in Korea. *Korean J. Parasit.* 1975,

13: 60-70
 Lee SH, Chai JY. Praziquantel in the treatment of trematode and cestode infections. J. Korean Soc. Chemoth. 1985, 3: 95-118 (in Korean)
 Lee SH, Chai JY, Sohn WM, Hong SJ, Lee KC. *In vitro* and *in vivo* effects of praziquantel on sparganum. Seoul J. Med. 1986, 27(2): 135-142
 Lee SH, Chai JY, Hong ST, Sohn WM. Survival in mice of damaged spargana by praziquantel, 1988 (To be published).
 Kim H, Kim SI, Cho SY. Serological diagnosis of hu-

man sparganosis by means of micro-ELISA. Korean J. Parasit. 1984, 22(2): 222-228
 Moulinier R, Martinez E, Torres J, Noya O, De Noya BA, Reyes O. Human proliferative sparganosis in Venezuela: Report of a case. Am. J. Trop. Med. Hyg. 1982, 31(2): 358-363
 Rim HJ, Lee JS, Joo KH, Kim SJ, Won CY, Park CY. Therapeutic trial of praziquantel (Embay 8440; Biltricide) on the dermal and cerebral human cysticercosis. Korean J. Parasit. 1982, 20(2): 169-190

= 국문초록 =

인체 스파르가눔증에 대한 프라지칸텔 치료 실패(1 증례 보고)

서울대학교 의과대학 기생충학교실 및 중앙대학교 의과대학 기생충학교실*

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경기도 용인군에 거주하는 55세 남자 환자가 1984년 복부 및 액와부의 피하낭종 제거술을 받은 바 스파르가눔(sparganum) 총체가 적출되었음이 확인되었다. 그러나 수술후 3-4개월에 좌우 허리와 저드랑이 부근에서 엄지 손가락만한 종괴 3개를 다시 발견하였다. 이 때 스파르가눔 항원에 대한 항체가(ELISA흡광도)는 1.06으로 나타나 스파르가눔증의 재발로 생각되었다. 프라지칸텔에 의한 치료 가능성을 생각하고 하루 75 mg/kg 용량을 3분복하여 5일간 총 375 mg/kg을 경구투여하였다. 또 같은 용량으로 치료를 1개월후 한번 더 반복하였다.

치료후 4개월 동안, 종괴는 촉진상 다소 부드러워졌으나 환자는 계속 통증과 가려움증을 호소하였으며 항체값도 1.35로서 전혀 감소되지 않았다. 다시 수술을 시행한 바 종괴 3개에서 살아있는 스파르가눔 3마리가 회수되었다. 이상의 결과는 인체 스파르가눔증 치료에 있어서 프라지칸텔이 유효하지 못함을 나타내었다.