

An acephalic budding *Cysticercus* (= *Racemose cysticercus*) found at the abdominal wall of a man

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INTRODUCTION

Racemose cysticercus is a rare, large, cystic, lobated or grape-like metacestode which has been predominantly found in the intracranial cavity of man, especially at the base of brain (Slais, 1970). Its origin has been thought to be *Cysticercus cellulosae* because of the almost identical features of the bladder wall (Slais, 1970) and because some cases of racemose cysticercus infection were found to be simultaneously infected with *C. cellulosae* (Haskovec, 1929; Henner *et al.*, 1946). Racemose cysticercus, however, is devoid of typical scolex and its bladder wall has abnormal growth.

Since the metacestode of *Taenia multiceps* also has large, cystic bladder of similar histologic features, the racemose cysticercus may be a sterile coenurus (Slais, 1970; Jung *et al.*, 1981). Although Slais (1970) described the histologic differences of bladder wall of *C. cellulosae* from those of *C. bovis* and coenurus, their differentiation was not always easy in every clinical materials (Voge, 1967); therefore some uncertainties are still present on the origin and other aspects of racemose cysticercus.

In this context, further informations concerning the racemose cysticercus are necessary to understand this entity more correctly. Recently we had a patient from whom a *Cysticercus* was removed surgically at the abdominal wall. The metacestode showed many interesting aspects in

its morphology as well as its infection site. We describe the worm in order to supplement the knowledge on the racemose cysticercus.

CASE RECORD

A Korean man (YCK) living in Seoul, a factory worker aged 26 years was admitted to Department of Dermatology, St. Paul's Hospital, Catholic Medical College on March 10, 1983 because of a solitary mass palpated at the left lower quadrant of abdominal wall. The mass was firstly recognized about 4 months before the admission. It was localized, firm, thumb-tip sized, indolent mass without spreading tendency to adjacent areas.

Laboratory tests undertaken before surgery revealed: Hb. 16.7g/dl, WBC 9,300/ μ l with differential count of 45% neutrophils, 54% lymphos and 1% eosinos; bleeding and coagulation times were normal; findings of urinalysis, chest X-ray film, blood chemistry and electrocardiogram were all in normal range. Stool examination was not done.

Excisional biopsy of the mass was undertaken. At surgery, an irregular pale purple to brown coloured rubbery mass of 2.0 \times 1.2 \times 1.8cm was removed between subcutaneous fat and fascia of external oblique muscle of abdomen. The cut section of the mass, fixed in formalin, revealed slightly wrinkled generally smooth cavity from which a white round worm of 1.2 \times 0.8 \times 0.4cm was found. Histologic examination showed

hyalinized, thick fibrous wall infiltrated heavily with eosinophils and lymphocytes.

PARASITOLOGICAL FINDINGS

The fixed worm from the cavity looked like a normal *C. cellulosa* at first glance, which had a cystic bladder and a parenchymatous scolex body. To confirm the diagnosis, a part of bladder was cut to process histologically. The remaining worm was stained by Semichon's acetocarmine.

The histology of the normal bladder was compatible with that of *C. cellulosa* (Fig. 5). Tegumentum (mean thickness $2.5 \mu\text{m}$) had fine microtriches on outer surface. Wart-like protuberances, made by contracted subtegumental muscles were $17\sim 32$ (mean 22) μm in width at their base, and $10\sim 23$ (mean 17) μm in height. These superficial protuberances may be absent while many were formed in certain areas. Subtegumental cells aligned under tegumentum. The average thickness of total bladder wall was $86\pm 12 \mu\text{m}$. Sections of excretory canals were found in the fibrous connective tissue of bladder wall together with scattered cells.

Acetocarmine stained, total mounted specimen, however, revealed differences from *C. cellulosa* in two aspects (Fig. 1). First, the *scolex body* (in this paper this term referred the round parenchymatous structure accomodating the suckers, rostellum, hooklets, invaginated neck, spiral canal and adjacent connective tissue), which was placed to outside of teased bladder and was made flat under a slide glass pressure, revealed some abnormal findings. The scolex of *sensu stricto* (composed of suckers, rostellum and hooklets) was absent and had no vestiges of their ghost, suggesting agenesis rather than degeneration. The invaginated neck was conical shape occupying only the central portion of the scolex body (Fig. 2). Spiral canal did not encircle the entire inner tissue of scolex body. The vestibule around the margin of scolex body was not evident (Figs. 1A & 2)

Another abnormal finding was that there were

buds on the bladder wall around the opposite of scolex body. The exact number of the buds was hardly countable, but at least 8 were recognized (Fig. 1). Some buds were definitely cystic (Fig. 3) while some were stained darkly by acetocarmine suggesting cellular nature (Fig. 4).

To observe the nature of different buds and to prove the vestibule histologically the acetocarmine stained specimen was rehydrated and the portion of bladder wall with buds and the lower half of scolex body were removed, sectioned serially and stained by haematoxylin and eosin.

The sections of the lower scolex body (Fig. 6) showed that the neck portion occupied central part of so-called spiral canal. Spiral canal was lined by a simple layer, which was composed of tegumentum, subtegumental cells and thin connective tissue, without forming complicated infoldings into the canal. The surrounding tissue of the spiral canal was spongy, whirl-like connective tissue. Sections of the vestibule were not recognized at the outer margin of the connective tissue of scolex body.

The buds were of two different types again. The first type of bud was covered with tegumentum of normal thickness and had subtegumental cells of normal density. The tegumentum had microtriches. Its inner cavity was filled with loose connective tissue with scarce cellular elements and excretory canals. The neck of bud was short and narrow. The bud may have a daughter bud (Fig. 7).

The second type of bud was characterized by the local proliferation of subtegumental cells, as shown by the increased number in a locus (Fig. 8). It accompanied the significant thickening of the covering tegumentum as well as the thickening of inner fibrous tissue of bladder wall. The tegumentum of the buds had microtriches too. In addition, round or irregular shaped lacunae were present in tegumentum (Fig. 10), under tegumentum (Fig. 11) or under a layer of subtegumental cells (Fig. 10). The lacunae varied in their size. Some sections revealed filamentous or various shaped projection to outside of the bladder wall, which were composed of tegumen-

tum with fibrous tissue (Fig. 12). Subtegumental cells may be found at the thickened tegumentum or up to the outgrown projections.

DISCUSSION

Tangled cystic branchings covered with normal tegumentum was one of features of racemose cysticerci. As shown in Fig. 29A in his review of Slais (1973), in Fig. 2 of Jung *et al.* (1981) and in Fig. 31-14A of Beaver *et al.* (1984), sections of cystic branchings may give an impression of forming separated daughter cyst. However the racemose cysticercus of this type was grossly one cyst, which had complicated branchings (Zenker, 1882). Histologically the first type of the abnormal buds of the present worm corresponded to those branchings.

Talice and Gurri (1950) described the histologic findings of a racemose cysticercus surgically removed from an Uruguayan man as follows: The irregularities in thickness of bladder wall with numerous infoldings and structural irregularities such as variability of thickness of subtegumental fibrous layer and the presence of multiple lacunae of variable size. These findings could be explained as results of multifocal and temporary proliferation of subtegumental cells as observed in the second type of bud in the present worm. Evidently the proliferation of subtegumental cells and their disorderly alignment were other features of the abnormal buds as seen in microphotographs of the racemose cysticercus presented by them. However, the second type of bud of present metacestode was not exactly same histologically with that of Talice and Gurri (1950). The more marked proliferation of subtegumental cells and presence of outgrown projection on the bud characterized the second type bud of present metacestode.

Voge and Berntzen (1963) described tapeworm cysts collected from a fatal dog of ascites. The cysts showed disorderly alignment and proliferation of subtegumental cells. Their findings together with the present experience suggested that the proliferation of subtegumental cells may be

a common histologic manifestation of abnormal growth or asexual reproduction of larval tapeworms.

The present metacestode was not that of *Taenia crassiceps* (Freeman, 1962) which multiplied asexually by forming daughter metacestodes on the bladder wall. Our worm did not make such cystic buds of *T. crassiceps*. It was not a coenurus either, because only one scolex body was present. We could not diagnose the present worm as a metacestode of any *Taenia* spp. of wild animals because of agenesis of scolex and because of paucity of information on taeniid of wild animals in Korea. Only one report of adult recovery is available from a weasel (Kim and Song, 1972). No *C. pisiformis* were reported to infect man.

Though Voge (1963) stressed the importance of scolex morphology in differentiation of *C. cellulosae* from *C. bovis*, the presence of vestibule around the scolex body was indicated as a differential key between two species by Slais (1966) together with the different morphology of tegumental protuberances. Voge (1967) agreed the vestibule as different, but she declined the tegumental protuberance as another key. This was the reason why we tried to demonstrate the vestibule at scolex body of the present worm, in which we could not find it. Because the development of scolex body was evidently anomalous in many aspects, the last process of forming vestibule may also be deleted in the present worm. Although we could not find any evidence of *C. cellulosae* in scolex body, the tegumentum of the bladder wall was compatible with that of *C. cellulosae* by Slais' criteria.

The metacestode described in this paper, therefore, had two types of abnormal buds simultaneously on the bladder as well as normal bladder. The worm also showed agenesis of scolex. Based on these findings we identified the present worm as a racemose cysticercus by the criteria of Beaver *et al.* (1984). We suspected *C. cellulosae* as a species of origin but evidences were few.

The present worm was different from conventional racemose cysticerci in some respects: (1)

the location of infection was abdominal wall rather than intracranial cavity, (2) size of the bladder was that of usual *C. cellulosae* rather than huge cyst, (3) two types of abnormal buds were simultaneously found in a worm (usually one histologic type constituted a racemose cysticercus), and (4) the scolex body was relatively well preserved.

As far as we can trace, the racemose cysticercus infection outside the intracranial cavity of man was once reported by Firket (1895) at the heart wall. So far, the extracranial location has been extremely rare among the rare occurrence of racemose cysticercus. It is evident that extracranial infection is possible.

The present worm raised a question what is the structural definition of "acephalic" (either agenetic or degenerated) in racemose cysticercus. In conventional racemose form, no *scolex body* was recognized. Unlike conventional, the scolex body was formed anyway, though with multiple anomalies. In the present paper, therefore, we interpreted "acephalic" as the absence of the scolex of *sensu stricto*.

The present racemose form suggested that the process of abnormal growth was a making of subtegumental cells as speculated by Voge and Berntzen (1963) and Voge (1967). The present worm also suggested that the transformation into a racemose form, whatever the species of origin were, occurred through genetic mechanism because multiple anomalies were associated with budding.

SUMMARY

An acephalic budding *Cysticercus* of 1.2 cm long was removed surgically at the abdominal wall of a Korean man. The worm revealed abnormal buds on the bladder wall and absence of suckers and hooklets in the scolex body. The buds were of two histologic types; branching bud covered with normal tegumentum and with subtegumental cells of normal density, and buds of proliferated subtegumental cells with lacunae formation. On the bases of the morphologic fea-

tures, it was identified as a racemose cysticercus. This case confirms that its extracranial location is possible.

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＝국문초록＝

두절기형과 분아형성이 있는 낭미충(=포도낭미충)에 의한 복벽 감염

중앙대학교 의과대학 기생충학교실*, 가톨릭의대 피부과학교실** 및 기생충학교실***

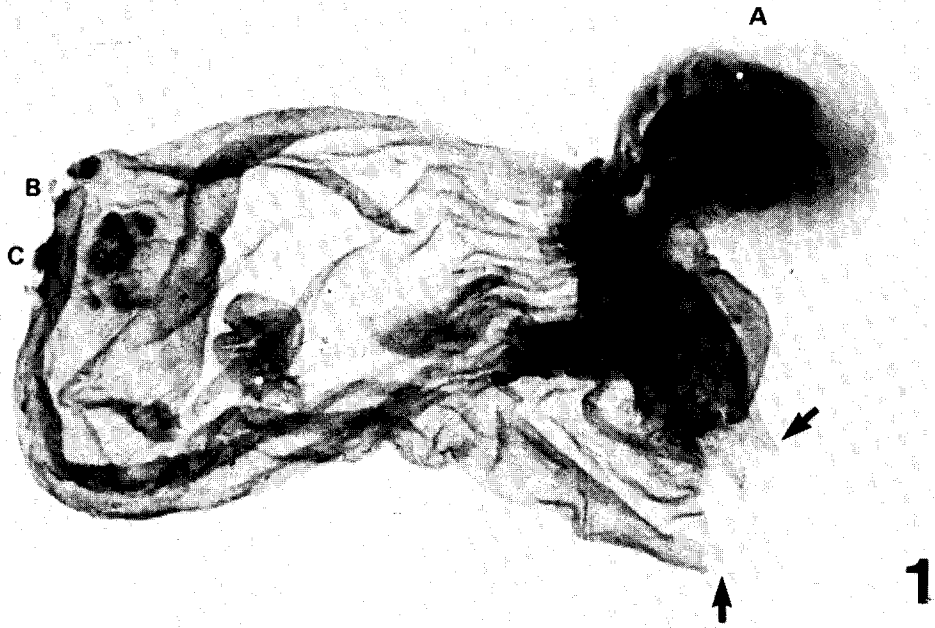
조 승 열* · 조 백 기** · 최 원 영***

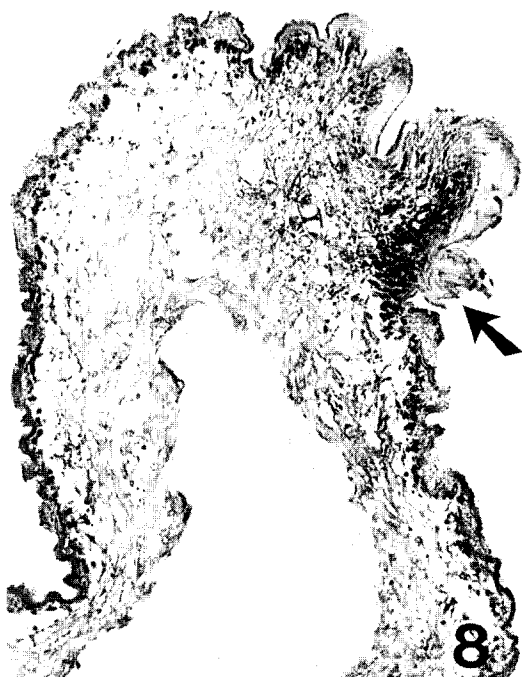
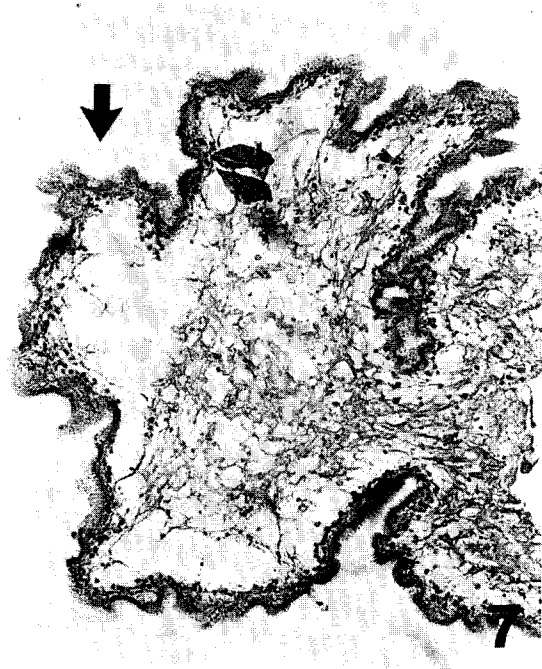
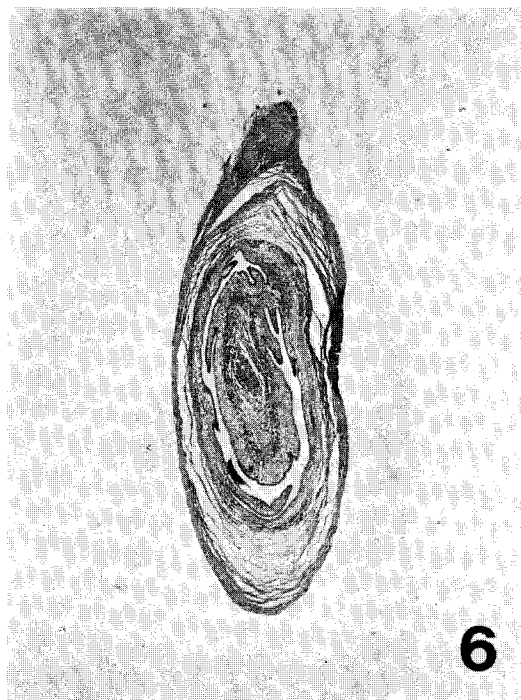
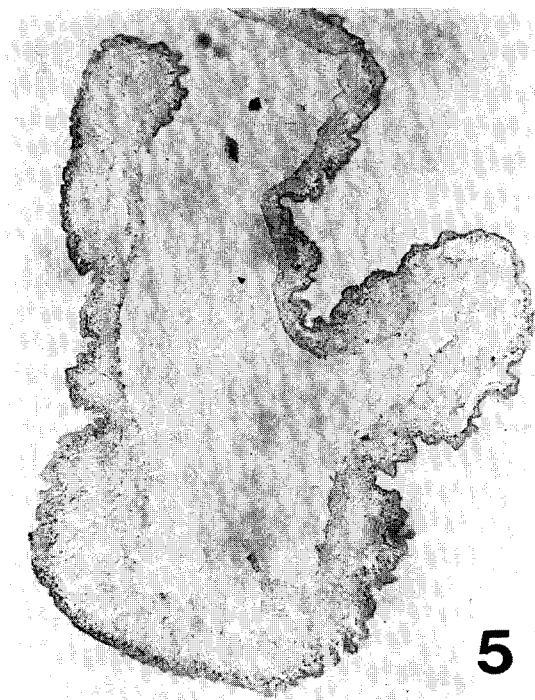
1983년 3월 좌하부 복벽(左下部 腹壁)의 종괴(腫塊)를 주소(主訴)로 한 26세 남자 환자를 수술하고 병변부위의 육아종(肉芽腫)에서 길이 1.2cm인 낭미충 한마리를 얻었다.

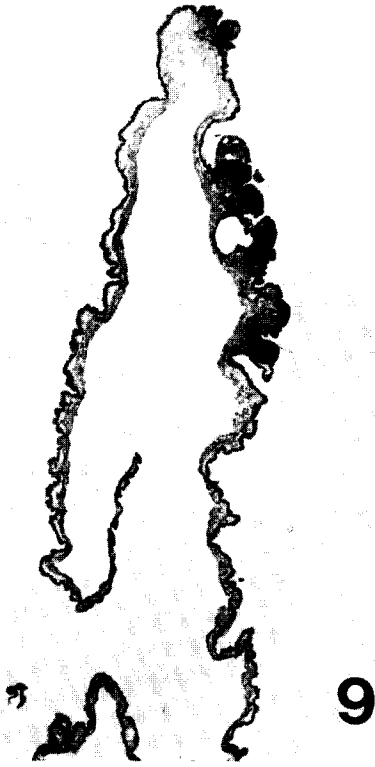
이 낭미충은 낭벽(囊壁)에 비정상적인 분아(分芽)를 형성하고 있었고 내번(內臟)되어 있는 두절부위(頭節部位)에는 빨판과 갈고리가 없는 이상소견을 보이고 있었다. 낭벽의 분아는 조직학적으로 두가지 형태를 하고 있었다. 제 1형은 정상두개인 표피(表皮)로 덮여 있고 표피하세포(表皮下細胞)의 밀도가 정상이면서 분지(分枝)하는 분아였고, 제 2형은 표피하세포가 증식하면서 표피 및 표피하조직이 두꺼워지고 표피 및 표피하조직에 크기가 다른 공동(空洞; lacunae)을 여러개 만드는 것이었다.

분아의 조직학적 소견과 두절기형을 기초로 이 총체를 포도낭미충(葡萄囊尾蟲, racemose cysticercus)으로 동정하였다.

이번 증례는 포도낭미충 감염이 두개강(頭蓋腔) 이외의 장소에서도 일어남을 확인하게 한 흥미있는 증례이다.







EXPLANATION OF FIGURES

- Fig. 1.** Acetocarmine stained total mount of the metacestode. "A" indicated the parenchymatous scolex body, which was placed at outside of bladder (see Fig. 2). "B" and "C" were abnormal buds which were enlarged in Figs. 3 and 4 respectively. Two arrows at lower right indicated the removed bladder wall for examination of normal histology (Fig. 5). $\times 9$.
- Fig. 2.** Magnification of Fig. 1A. Suckers and hooklets were absent. Neck and spiral canal were simple. Vestibule was not evident. Acetocarmine stained, $\times 40$.
- Fig. 3.** A cyst-like bud on the bladder wall (magnification of "B" in Fig. 1). Acetocarmine stained, $\times 40$.
- Fig. 4.** A parenchymatous buds on the bladder wall ("C" in Fig. 1). The mid-portion of the bud was darkly stained by acetocarmine. Random projections were present at outer surface. A cystic structure was seen within the bud. Acetocarmine stained, $\times 40$.
- Fig. 5.** A section of bladder wall where abnormal buds were not present (indicated by two arrows in Fig. 1), H&E, $\times 100$.
- Fig. 6.** A section of mid-portion of the scolex body. Neck situated at the centre; spiral canal was poorly developed, simply encircled the neck; the linings of the spiral canal had few infoldings projected to the lumen of the canal. Vestibule was absent. H&E, $\times 25$.
- Fig. 7.** A section of a branching bud, covered with tegumentum of normal thickness and had subtegumental cells of normal density. The neck of bud was short and narrow. A daughter bud (indicated by an arrow) arose the bud. H&E, $\times 250$.
- Fig. 8.** A section of the second type bud. The arrow indicated the locus where subtegumental cells proliferated; covering tegumentum and inner connective tissue thickened. H&E, $\times 250$.
- Fig. 9.** A section of abnormal bud of the second type, showing the multifocal occurrence. H&E, $\times 250$.
- Fig. 10.** Magnification of a part of Fig. 9. Increased number of subtegumental cells was recognized. Numerous cystic lacunae were present in the tegumentum or under a layer of subtegumental cells. H&E, $\times 250$.
- Fig. 11.** A section of the second type bud. A large lacuna was formed between tegumentum and subtegumental tissue. Subtegumental cells proliferated and were intermixed with fibrous elements. H&E, $\times 250$.
- Fig. 12.** Another section of bud of the second type. A lacuna was formed under tegumentum. Thickened tegumentum sloughed partly, and projections of tegumentum with fibrous tissue were seen. H&E, $\times 250$.