INTRA-MEDULLARY CYST OF THE SPINAL CORD DUE TO THE CESTODE MULTICEPS MULTICEPS IN THE COENURUS STAGE

REPORT OF A CASE

BY

J. W. LANDELLS

From the Bernhard Baron Institute of Pathology, London Hospital

(RECEIVED FOR PUBLICATION, OCTOBER 1, 1948)

The cestode Multiceps multiceps is well known as a cause of intracerebral cysts in sheep, but there are only three reports of similar human infestation certainly due to this particular parasite (Brumpt, 1936; Clapham, 1941; Cluver, quoted by Craig and Faust, 1943). The subcutaneous tissues in man have been more frequently involved; there are some eight reports from various continents. The case now to be detailed is the first in which the spinal cord has been affected. A parallel case of hydatid cyst in the spinal canal has been published by Rogers and Tudhope (1938); the measurements of the hooklets in their case and in the present case establish the different species involved.

Case Report

A girl aged 14 was admitted to the Brentwood Annexe of the London Hospital under Mr. Northfield on Feb. 15, 1947. She was suffering from spastic paraplegia of acute onset at the level of the sixth thoracic segment; her earliest symptom, “pins and needles in her legs,” appeared and increased in intensity only four weeks before motor and sensory paralysis became complete. Double incontinence followed three weeks later, but she never complained of any pain.

Laminectomy and exploration of the cord were undertaken on Feb. 19, 1947, on a clinical diagnosis of intra-medullary tumour, the spines and laminae of the fourth to the ninth thoracic vertebrae being removed. The dura was tightly stretched over a swelling of the cord, the surface of which was irregular and had a patchy bluish discoloration. A midline incision was made into the posterior surface of the cord at about the fifth and sixth thoracic segments, and a thin-walled cyst, which presented itself spontaneously through the opening, was easily and apparently completely delivered by gentle traction; it had no visible attachment to the cord. The paralysis did not improve, and a second operation a month later, exposing the whole length of the cord from the fourth to the ninth thoracic segments, showed no further cysts and no visible cause for the continuing paralysis. During ten months since this second operation there has been very slight sensory recovery but no motor recovery, and increasingly severe and painful flexor spasms have required relief by cordotomy.

Special investigations.—Four examinations of the blood showed no eosinophilia, 450 eosinophils or fewer being counted in 6,100 to 9,200 white cells. The Casoni test was negative, but was not tried until some weeks after the removal of the cyst.

In already reported cases there is no record of blood examinations. Casoni tests in two patients with subcutaneous coenuri—described as Multiceps serialis—were also negative.

Further inquiry about the patient revealed that she had never been abroad, but had been evacuated during the war to Glamorgan, where the worm is common enough, and here she had been in close contact with the sheepdogs at a farm. She returned to Kent a year before the onset of her illness; there had been possible contact with a young puppy—an unlikely carrier of the definitive stage—but no close or frequent contact with other animals.

Pathological examination.—The cyst (Fig. 1) was received already fixed in formol-saline, and consisted of a chain of vesicles attached to each other in a twisted and tangled group; individual vesicles were up to 0.6 cm. in diameter and had filmy transparent grey walls with white nodules (0.1 cm. in diameter), varying in number from two or three up to thirty, on the inner surface. These nodules were usually in small groups (up to eighteen) but also occurred singly.

When a nodule was compressed under a cover-slip and examined under low magnification it showed the four suckers and double row of hooklets characteristic of a Taenid cestode (Fig. 2): the multiple scolices in each vesicle identified the type of cyst as a coenurus, Professor Buckley, of the London School of Hygiene and Tropical Medicine, very kindly completed the identification by measurement of the hooklets as Multiceps multiceps, the sheep gid-worm. Unfortunately, as all the material was fixed in formalin, this could not be confirmed by identification of the
definitive stage by passage through a puppy. Further
helminthological details of this case have since been
published by Crusz (1948).

**Histology.**—A group of scolices and part of
the cyst wall were embedded in paraffin and sections were
stained with haematoxylin and eosin, iron haemato-
xylin and van Gieson, and Mallory's phospho-
tungstic acid haematoxylin.

The cyst separated cleanly from the tissues of the
spinal cord, no part of which was present in sections.
The outer wall consisted of homogeneous eosinophil
material 50 μ thick, thrown up into very numerous
rounded wrinkles 20 to 30 μ in diameter; the fibres
composing the wall formed a coarse reticulum. On
the inner surface was a spongy parenchyma with
numerous small rounded nuclei, often in groups; the
confluence of vacuoles of this parenchyma formed
the cyst cavity. The scolices were developed in
outgrowths of the parenchyma by invagination from
the cyst; this invagination appeared in section as
irregular cleft lined by the cuticle of the scolex
(Fig. 3). This was a smooth, uniform, fibreless mem-
brane (10 μ thick) much folded, which stained a deeper
and more purple tint in haematoxylin and eosin than
the outer cyst wall. The suckers and hooklets are
easily made out in the apex of the invaginations
(Fig. 4).

For further study of the reactions of the host
to the cyst I am indebted to Professor L. P. Garrod
for the loan of a slide, stained with haematoxylin
and eosin, of the cyst in Clapham's case, and to
Professor Buckley for a sheep brain containing a
coenurus.

The human material shows separation of the cyst
wall from the brain by a space of 250 to 500 μ.
This is clearly a natural line of cleavage of which
the surgeons in my case took advantage. From
the border of this gap, four zones can be made
out in the brain before normal nervous tissue is
reached, at the depth of some 3 mm. from the
cyst wall.

1. The first zone (100 μ deep) is extensively
necrotic and is composed of angular, sometimes
multinucleated cells, orientated at right angles to
the cyst and separated by numerous clefts and
spaces. Preservation is too poor to determine the
character of the cells but some undoubtedly are
macrophages.

2. The second, less well defined zone (100 to
300 μ) consists of delicate fibrous tissue, the fibres
lying parallel to the surface.

3. The main zone of cellular infiltration succeeds
this with an indefinite boundary and is up to
700 μ thick, though averaging only 200 μ. The
infiltration, denser round small blood vessels, is
mainly of small lymphocytes with a few plasma
cells. Hyaline, probably collagenous, areas with
spindle cells are also present, and a larger artery
is undergoing purulent inflammation and necrosis.

4. In the nervous tissue proper there is an
increase of large plump astrocytes which are
arranged in such a way as to suggest severe
compression; this zone is oedematous in places
and up to 400 μ wide. The adjacent cortex shows
chromatolysis and dropsical degeneration of a
large number of pyramidal cells.

Collectively these observations indicate a chronic
granulomatous inflammation forming a zone at
least 3 mm. deep around the parasite. In the
spinal cord a corresponding zone would be of
greater functional importance than in the brain,
and the clinical course of the case and the non-
recovery after the removal of the cyst are thus
easily explained.

In material from the sheep there is again a very
obvious line of cleavage between the cyst wall and
the reaction zone of giant cells, fibrin deposition,
and necrosis in the adjacent tissue. This zone is
a great deal wider than in the human, and the
giant cells are more numerous and much larger.
There is rather less small round-cell infiltration,
and no necrosis in the outer adjacent zone. Traces
of collagen are present in the edges of the more
intense reaction.

Although, therefore, there is a very intense
reaction to the presence of the cyst, this latter does
not become incorporated in the host tissues, and
can be expected to “shell” out on surgical manipu-
lation: and, unless compression has already resulted
in severe damage to important nervous tissues,
operative treatment should have a very good
chance of success.

**Literature**

Crusz (1948) has reviewed from the helmintho-
logical point of view the cases of coenurosis
reported in man, including the present case, and
both he and Clapham (1942) consider that the
three types given specific rank by previous authors
(for example Faust) are not true species but rather
physiological strains or immature stages of the
single species *M. multiiceps*. In Brumpt's report
a Parisian locksmith, aged 40, presented with fits
and aphasia and was found at autopsy eight
months later to have two cysts, one in the posterior
horn of the left lateral ventricle and one in the left
angular gyrus; the latter was degenerate and con-
tained only hooklets, but 75 scolices were found
in the ventricular cyst.

No details of the case reported by Cluver in the
lateral ventricle of an African are available: the
original reference (not given by Faust) cannot be
traced.
INTRA-MEDULLARY CYST DUE TO MULTICEPS MULTICEPS

For my knowledge of the clinical details of Clapham’s case I am indebted to Dr. D. H. Fulton, pathologist to Peterborough Hospital. A man of 40 was found in coma in his bedroom and died 20 hours later, his relatives apparently mistaking his illness for alcoholism. He had served in the Navy from the age of 18 to 32 and had been well apart from an attack of meningococcal meningitis at the age of 18. At the age of 36 he had the first of increasingly frequent and severe sudden headaches, which lasted up to 24 hours. He had no fits or paralyses. At autopsy the right cerebral hemisphere was larger than the left; on section a thin-walled cyst 5 cm. in diameter by 8 cm. from front to back was found above and lateral to the right lateral ventricle; its inner wall was studded with white, raised areas, and it contained clear fluid. The left hemisphere and the cerebellum were normal. A massive haemorrhage had occurred in the pons and medulla and was the cause of death. The thoracic and abdominal organs were normal. He had died with arms and legs extended and with hands tightly clenched. His headaches with their paroxysmal onset resemble those occurring with colloid cyst of the third ventricle or the headache of acute internal hydrocephalus, and a terminal pontine or mid-brain haemorrhage is a not uncommon sequel to increased supratentorial pressure. The infection was probably acquired in this country rather than during his Naval service. This is suggested both by the lapse of four years between leaving the service and the onset of the headaches, which may be taken as marking the onset of the cerebral lesion, and by the life history of the parasite. This develops to full maturity in the sheep in about eight months (Neveu-Lemaire, 1936); it is then about 3 to 5 cm. in diameter and causes softening of the overlying skull. It occurs all over the world where sheep-raising is carried on, the definitive stage being passed in the intestine of the dog or wolf and the intermediate cyst (Coenurus) stage in the central nervous system of herbivores. In this country it is fairly well known in the Welsh sheep farms; but the Chief Inspector of Abattoirs in Cardiff has only seen two cases in twenty years, since the sheep commonly die on the farms. In some cases the shepherds remove the cysts with their penknives, locating them by the softening of the skull. The name “gid-worm” is derived from the staggering of the sheep when affected by the parasite.

I am indebted to Mr. D. W. C. Northfield for the clinical notes, and to Professor J. J. C. Buckley, Professor L. P. Garrod, Dr. D. H. Fulton, and Dr. Phyllis Clapham for assistance with the parasitology and the records of previous cases. I also wish to thank Professor D. S. Russell for help in preparing the paper and Mr. A. J. King for the photographs.

REFERENCES

Intra-medullary Cyst of the Spinal Cord Due to the Cestode *Multiceps Multiceps* in the Coenurus Stage: Report of a Case

J. W. Landells

*J Clin Pathol* 1949 2: 61-63
doi: 10.1136/jcp.2.1.61

Updated information and services can be found at:
http://jcp.bmj.com/content/2/1/61.citation

These include:

**Email alerting service**

Receive free email alerts when new articles cite this article. Sign up in the box at the top right corner of the online article.

**Notes**

To request permissions go to:
http://group.bmj.com/group/rights-licensing/permissions

To order reprints go to:
http://journals.bmj.com/cgi/reprintform

To subscribe to BMJ go to:
http://group.bmj.com/subscribe/