# Some New Cases of the Occurrence of Bothriocephalus liguloides Lt.

By

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With Plate V *bis.*

As far as we are aware, that larval Cestode parasitic in man, first described by Cobbold as *Ligula Mansoni* and renamed by Leuckart as *Bothriocephalus liguloides*, has hitherto been found in only two cases (Leuckart: die menschlichen Parasiten des Menschen. II Aufl. p. 941–951). The one case was that of a Chinaman, in whose corpse Dr. Manson of Amoy found no less than 12 pieces, one free in the pleural cavity and all the rest in the sub-peritoneal connective-tissue in the region of Fossa iliaca behind the kidneys. The other case was that of a Japanese and was observed by Dr. Scheube, then of Kyōto Hospital and was communicated by him to Prof. Leuckart. In this case the worm was discharged from the urethra.

Of late we have come to know of at least six new cases of the occurrence of Bothr. liguloides. Notes on them have already appeared in some medical journals written in Japanese, but we believe, no apology is needed for the reproduction here of the accounts of these cases with such additional remarks as suggest themselves to us. The six cases are as follows:

**Case I.**—*Bothr. liguloides discharged from the urethra.**—We owe the knowledge of this case to the kind communication of Mr. K. Namba, a physician in the province of Echigo, who also sent us the worm for examination. He writes to the following effect: the patient was a boy, scrofulous and of weak bodily constitution. When three years old he suffered from frequent swelling of the scrotum on the right side, consequent on inguinal hernia. This complaint ceased, but after the lapse of several years, when he was nine years of age, he began one day (July 1886) to experience difficulty in urination, which had to be done often but only drop by drop. Two days passed in this way, when, while making efforts for the passage of urine, a tapeworm-like body came out of the urethra to the length of about 10 cm. On being drawn it contracted and tore off (to what length is not stated). On the following day, the patient came to Mr. Namba, who put him in warm-bath and carefully wound out the worm, that still hung out of the urethral opening and showed signs of movement. The piece thus obtained measured over 20 cm. After this, the urine passed unobstructedly and an inquiry made many days afterwards showed that the boy had since felt in his usual health.

The measurement above given must be considered as giving only a fairly approximate length of the piece extracted. The piece of the worm sent to us in spirit was only 8 cm. long (*Fig. 1*). We do not know whether this piece is the whole that was pulled out by Mr. Namba. At all events, numerous wrinkles and folds observable on the surface, show that it has greatly contracted. Both ends are not natural, so that it was not possible to determine which is the anterior and which is the posterior end. One end was greatly disfigured while the other was deeply notched in the middle-line and plainly indicated that the cut at this place must have been made when fresh. The

* This case was independently published by Murata in *Chūgai-Iji-Shimbō*. No. 181. 1887.

breadth of the piece was fairly uniform, at one place measuring 10 mm. The body was fleshy, reaching about 1.75 mm. at the thickest part; its lateral edges were rounded. The color was whitish, slightly translucent.

On the one surface there was a median depression running through the whole length. It was by no means sharply defined. The lateral halves of the body were, for the most part, more or less reflected toward the side on which this depression lay, so that at some places the cross-section would present a V-shape. Numerous wrinkles, mostly transverse, gave the lateral margin an uneven outline. The specimen appeared as if it were swollen, but sections showed that such was not the case.

**Case II.**—*Bothr. liguloides from the urethra.*—*Igakushi* S. Saitō of Kyōto reported, at one of the meetings of Kyōto Medical Society, of two cases of tapeworm-like parasite, which we have recognized as Bothr. ligulodes. This report was published by one of us in Nr. 185 of the *Chiugai-Iji-Shimpō*. One of the cases will be described here and the other afterwards as Case V.

According to Saito's report, the patient was a man (son of a farmer in Sayama Village, near Kyōto), 25 years of age and strong in body. Five years previously (1882) he is said to have suffered from violent gonorrhoea, at one period passing blood with urine. In half a year he recovered, but sometime afterwards, the desire for passing urine began to be frequent, sometimes as much as 15 or 16 times in a day. However it was only by great efforts that he could discharge urine. Besides, he felt now and then itching or pressing sensations at the perinæum. This state continued until Oct. 14th 1887, when, while endeavoring for the passage of urine, a moving worm protruded itself from the urethra. Mr. Ogino, a physician of the village carefully

pulled it out until it tore off leaving a part of the body behind. The piece obtained measured then 2 feet in length, about 6 mm. at the broadest and about 1.5 mm. at the narrowest part. For two days afterwards the patient felt pain in passing urine, which moreover contained blood. The frequent but scanty discharge of urine continued longer. When Saito examined him some time after, the urine was transparent and amber-colored, without precipitate or other abnormality.

We do not know what had since become of the piece of the worm that was left in the urethra nor of the complaint in urination.

Through the kindness of *Igakushi* Saitō we were enabled to examine the piece (*fig.* 2) of the worm extracted by Ogino. It was preserved in spirit, very much twisted, rather tough and greatly shrunken, probably the effect of having been thrown into strong alcohol. It was 245 mm. long. The one end was torn and the other, with which the worm undoubtedly first protruded itself from the urethra, was natural. The latter was no doubt the head-end. At this end the body had very much contracted, forming in contrast to the long-drawn part that followed, a rounded disc about 3 mm. broad and about one-third as thick. On its surfaces fine transverse wrinkles were discernible and at the middle of the anterior margin, there was a small but distinct indentation, showing the position of withdrawn rosterum. Behind this disc-like portion, the body was very thin, almost thread-like (1 mm. in breadth) for some distance and then gradually broadened toward the hind end, where it was thin and measured about 3 mm. in breadth. Transverse wrinkles were especially abundant near the margin and numerous longitudinal ones in the median portion. At some places, three longitudinal grooves (one median and two lateral on the one surface and two (lateral) on the other, were more prominent than others. In view of the shrunken state of the specimen, no importance can be attached to these grooves.

**Case III.**—*Bothr. liguloides from the urethra.*—This case was observed by Mr. Toyoda, a specialist in helminthiasis in Kyōto. He has published a note of the case in *Iji-Hyōron*, Nr. 2, 1888.

The patient was a citizen of Osaka, 42 years old. On the morning of May 8th 1884, he began to discharge blood with urine, and in the afternoon a white worm appeared from the urethra while urinating. Toyoda was immediately called for. He succeeded in pulling out the worm entire. This measured about 364 mm. in length and about 12 mm. in breadth. Put in a vessel (with water?) it continued to contract and stretch and move about for nearly two hours. It was then put into glycerine for preservation. As the worm was new to Toyoda, he tried various means to identify it but in vain.

As we read his note, there was scarcely any doubt as to the identity of the worm with Bothr. liguloides. In compliance with our request, Toyoda has kindly sent us the specimen for inspection. Examination of it showed the correctness of our assumption.

The specimen (in glycerin) is fleshy and well preserved but had been colored by carmine and cut open at some places, probably in attempt at dissection. It is figured in *Fig. 3*. The length was about 105 mm. and the greatest breadth, 6.5 mm. Both ends are natural so that the diminution in size, as compared with Toyoda's measurement, is entirely due to contraction. The body was narrower near one end than the other. The narrower end was no doubt the head, the configuration of which could not however be definitely ascertained on account of an unfortunate cut at this place. The broader hind-end showed a shallow indentation at the middle-line.

The two surfaces presented the following difference, distinctly for the most part. The one surface had a distinct depression, running longitudinally at the median line. Most of the irregular transverse depressions proceeded from this median line (see the upper part of

*Fig. 3*). The other surface (the lower part of the same figure) was divided into three longitudinal areas (one median and two lateral) of about equal breadth, by two lines of shallow depression. The distinctness of these areas was brought forth more by the fact that the lateral areas greatly bulged out in comparison with the median, than by the presence of those grooves. Moreover transverse constrictions were to be found mostly on lateral areas.

**Case IV.**—*Bothr. liguloides from the eye.*—This case has been kindly communicated to us by *Igakushi* R. Satō of Utsunomiya. He also placed the worm at our disposal. It was preserved in spirit, 25 mm. long, narrow (1.5–4 mm.) and flat. It was very much twisted and shrunken up, but microscopical investigation left no doubt of its being Bothr. liguloides. The one end was cut and the other broken into shreds.

As to its origin Sato informs us that it was at the end of 1883 when he came across a patient with blepharitic symptom. He was a young man, 17 years of age, living at Kanazawa in Province Kaga, where Satō then resided. (In *Chiugai-Iji-Shimpō*, No. 181, in which one of us published a note of this case, it was stated by mistake that the locality was Utsunomiya). The affected place was the region of the inner angle of the left eye. At this place, not only the eye-lids but also a part of the conjunctiva around the Plica semilunaris was in a state of severe inflammation. At a spot just over the Caruncula lachrymalis, Satō observed a whitish spot which seemed to protrude itself. This was taken hold of by a pincette and pulled out, when it proved to be the worm in question.

**Case V.**—*Bothr. liguloides from the eye.*—We know one more case in which the parasite was located in the orbit. For information and

the specimen, we are indebted to *Igakushi* Saitō (see Case II).

The patient was a girl, 15 years old, living at or near Kyōto. On March 10th 1875, a vesicle-like protuberance formed itself, without any assignable cause, on the white of the left eye, mid-way between the cornea and the outer angle. Three days after, a physician, Mr. Shingū, examined and found it to be about of the size of the tip of little finger, soft and white, somewhat resembling cod-ovary in appearance. In two hours, he observed an elongated macaroni-like body, which on being slowly pulled out was found to be a worm.

The parasite (in spirit) has been tolerably well preserved. Length, 120 mm; breadth 3–6 mm. It is represented in *fig. 4*, in natural size. The hind end (the lower end in the figure) is not natural, having been apparently torn in the fresh state. Near the head-end, the body is broadened and terminates round, but with an invagination at the apex. A number of transversal and longitudinal rugæ were found as usual. On the greater part of the one surface, we noticed the division into three regions by two longitudinal grooves as described in the worm of Case III. For some distance there was also a distinct median groove in the middle region. The specimen was cut into sections (*Figs. 6 & 8*), of which we shall have to speak later on.

**Case VI.**—*Bothr. liguloides from the subcutaneous tissue of the thigh.*—This case relates to a soldier belonging to the Nagoya Garrison, The parasite was extracted by Mr. S. Nagao, Army Medical Officer, who not only supplied us with the following information but also kindly permitted us to prepare sections from his specimen.

The patient was a native of Toyama, in the Province of Etchiu. In the summer of his fifteenth year of age, that part of the right leg just above the knee-joint on the inner side swelled, without any apparent reason for it. In the interior of the swelling a hard mass

was to be felt. There was no pain. It was somehow treated by a local physician and disappeared in about 10 days. A year after, the swelling reappeared at the same place but again subsided in about the same length of time. From this time until his enlistment in the Nagoya Garrison, the same swelling often recurred, invariably during summer. The patient did not definitely remember if it took place *every* year or if there were years in which it did not occur. The enlistment was in May, 1885. In July of the same year, the usual swelling appeared on the inner side of the lower one-third of the right thigh. It was observed that the swelling shifted its position up and down by itself to a small extent. It caused no trouble and soon disappeared. The next year passed without the appearance of the swelling. But in 1887, at the beginning of July, the swelling manifested itself this time at Scarpa's triangle. It did not at all interfere with the patient's general health and dispersed in a few days. In September of the same year the swelling reappeared on the inner side of the middle of the thigh. As it gave him pinching pain, Mr. Nagao was consulted. The latter found a hard mass of the size of a fist, situated in the subdermal tissue at the above-mentioned spot. It could be shifted to a certain extent. The surrounding tissue was inflamed and swollen. Attempts were made to test if it contained anything obtainable by means of inserted syringe, but in vain. Iodine-tincture was administered for about 40 days. This had no desired effect; on the contrary, the swelling enlarged and the pain increased to such a degree as to make the patient incapable of performing his duties. He was then taken into the hospital. Carbolic-acid water was injected into the swollen tissue and cold wrapper applied. In 5 days there was indication of suppuration and so a warm wrapper was substituted for the cold. In 4 days more the swelling suppurated and was cut open. Together with thin pus the worm described below came out of the pus-cavity.

The latter, situated in the subcutaneous tissue, was traversed by trabeculæ of connective tissue in various directions. The wall of the cavity was at some places smooth, as if lined by serosa.

The worm is undoubtedly Bothr. liguloides. The specimen (in spirit) was about 88 mm. long and 3.5 – 6.5 mm. broad. The head-end is represented in *fig.* 5, twice magnified. The other end was torn. The involution at the front apex was distinct. Numerous irregular furrows, both transversal and longitudinal, were present as usual. Of longitudinal furrows, the two that divide one of the surfaces into three longitudinal areas, were unmistakeably recognizable (see *fig.*). When fresh, the body was soft and translucent.

The removal of the worm, which was undoubtedly the cause of the almost annual swelling, took place just nine years after this occurred for the first time.

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To the above six cases we might add one more which however could not be tested by us. Once during the dissection of a subject in the Anatomical Institute of the University, Dr. Disse found a worm imbedded in the subcutaneous connective-tissue of the left inguinal region. According to our informant, Mr. Takesaki of the pathological Institute, who was the eye-witness of the discovery, the worm was about one foot and a half long and tapeworm-like but unsegmented. It was new to Disse. Takesaki, whom we have shown specimens of Bothr. liguloides, believes that it was the same worm. It is said that the worm was preserved, but unfortunately it could nowhere be found.

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We have then before us *at least* 7 sure cases (the case of Scheube inclusive) of the occurrence of Bothr. liguloides in Japan. The

patient of Scheube was infected presumably in Kiushiu (see Lt.). Of the six cases mentioned by us, two occured at Kyōto and the rest at Osaka, Kanazawa, Toyama and Province Echigo respectively. This justifies us in believing that the parasite has a very wide distribution throughout the whole country. We may further assume that special research in this direction would show that the parasite is by no means so rare as it seems to be.

According to Prof. Leuckart the real seat of the worm is the connective-tissue as was found in Manson's case. This is fully borne out by the case in which the worm was found in the subcutaneous tissue (Case VI) and also by the two cases in which it was located evidently in the connective-tissue space around the eye-bulb (Cases IV & V.)

Leuckart made it highly probable that the worm has the power of changing its position—of moving through tissues to a certain extent. Three of the cases just mentioned put this beyond doubt. In two cases (IV & V) namely, the worm was found to have pierced the conjunctiva and to protrude itself and in one case (VI), the periodical swelling of the thigh, evidently caused by the presence of one and the same worm, was found to vary in position almost everytime it appeared, between the part just above the knee on the inner side and the Scarpa's triangle. It was moreover observed that the swelling changed its position of its own account during its existence.

Under such circumstances, Leuckart's explanation of the exit of the worm from urethra, that it had secondarily bored its way into the urinary apparatus, requires no additional evidence to prove its correctness.

All the known cases, except the two in which the parasite was found in the orbit, tend to show that it is mostly located in the lumbar or the pelvic region. The entrance into the urinary organ

is effected probably after the worm has acquired a considerable size. The wandering into the orbit must have taken place when it was yet of small size, but whether as hooked embryo or as small larva it is difficult to say.

As to how long the parasite may exist in the human body, we call attention to case VI, in which a worm caused the periodical swelling of tissues for nine years. In this connection it is to be noted that Scheube's patient suffered hæmaturia more than five years before the discharge of the worm from the urethra. In cases I and II either the swelling of the scrotum (said to be the consequence of inguinal hernia) or gonorrhœa (?) with hæmaturia occurred five or six years previous to the discharge. It is of course impossible to say what relation, if any, existed between these symptoms and the worm in these three cases.

The preserved and much contracted specimens that came to our view, do not allow anything definite to be stated of the configuration of the larval cestode in question. The head-portion of the worm represented in *fig. 4* has been cut into horizontal sections, one of which is shown in *fig. 6*. The involution of the apex appeared in such sections as a narrow branched indentation, which was not very deep. Neither the papilla-like apical projection seen by Leuckart (the partly evaginated head) nor the two grooves characteristic of Bothriocephalus, were observable in any of the specimens. The broadening near the anterior end is apparently the result of contraction. In general the body seems, to judge from what we have seen, to broaden gradually toward the posterior end (*figs 2–5*).

The numerous wrinkles and folds on the surface are undoubtedly the effects of contraction and of the preserving fluid. Whether the one or two more or less prominent longitudinal grooves or depressions, which we have taken notice of in our description of specimens,

are to be looked upon in the same light (as Leuckart does) or not, we are unable to say.

We have studied the finer structure of the worm by sections cut from five specimens, but we cannot add anything of importance to what is already known from the investigations of Prof. Leuckart.

The cuticula covers the entire surface of the worm. It is homogeneous, thin but sharply defined, and does not stain with carmine.

Like other Vermes there is immediately beneath the cuticula a system of fine circular and longitudinal fibers (*fig.* 7, on the right side). Circular fibers run externally to, but in close apposition with, longitudinal fibers. In both layers, the fibers run isolated and parallel with one another. Longitudinal fibers could distinctly be seen in cross-sections.

With respect to strongly developed bundles of longitudinal muscle-fibers as well as those isolated fibers that run in all directions through the mesenchyma, we have found just the condition as described by Prof. Leuckart, except that we have failed to recognize the especially thick grouping of muscle-bundles along the course of lateral nerve-trunks. Anteriorly the muscle-bundles concentrate themselves toward the circumference of the involuted head (*fig.* 6), just in the same way as we see it in the larva of Bothr. latus.

As was pointed out by Leuckart, the number of excretory vessels to be seen on cross-sections is very great. Those running in the neighborhood of lateral nerve-trunks are of much larger caliber than those in the peripheral or the median part (see *fig.* 8). In sections of the head-part (*fig.* 6), we have met with but a very few number of small vessels.

Larval Cestode (Sparganum) resembling the human Bothr. liguloides was found by us in Inuus speciosus as well as in Mustelus itatsi, but we reserve its description for a future opportunity.

*P. S.*—Since the above notes were in type, we were favored by Mr. K. Takahashi of the Medical College with following informations, which we will here add as :—

**Case VII.**—*Bothr. liguloides from the eye.*—The patient was a girl, eleven years old, native of Ko-aiki Village in the province of Kōzuke. In Spring of last year she suffered from conjunctivitis. From January of this year, the upper eye-lid of the left eye began to swell and redden with intervals of comparative repose. Even during such an interval, the eye-lid seemed to be somewhat thicker than usual. On March 16th, during a school-exercise she felt pain in the eye, so that she was compelled to return home. However the pain soon subsided and the next morning she was able to attend the school. On 19th a swelling was noticed on the eye, which was investigated by Mr. Hagiwara, a physician in the town of Mayebashi. According to him, the swelling was of the size of a small bean, was situated on the eye-bulb beneath the conjunctiva, showed no signs of inflammation and could be shifted for a certain extent. On cutting the conjunctiva open, a worm protruded itself. It was then drawn out by means of a pincette, during which process the patient felt a slight pain. It seems that the worm was originally situated in the region of the Fornix of the upper eye-lid but had changed its position so as to come beneath the conjunctiva bulbi.

The extracted worm, shown us by Takahashi, was about 25 mm. long (in spirit). Without doubt it was only a part of the entire worm. The greater part of the specimen was split lengthwise into two parts. The head-end was present. It had just the configuration as represented in our *fig. 4* or *5*, but measuring only 2 mm. in breadth.

## Explanation of Figures (Pl. V *bis.*)

*Fig. 1.*—A piece of Bothriocephalus liguloides in alcohol (Case I). Nat. size. *a* represents the shape of one of the cross-sections of the piece.

*Fig. 2.*—Bothr. liguloides in alcohol (Case II). Nat. size. Very much shrunken. Above is the head-end.

*Fig. 3.*—Bothr. liguloides in glycerine (Case III). Nat. size. The head-end is above. *a*, supposed shape of its cross-section. Here and there longitudinal cuts on the body.

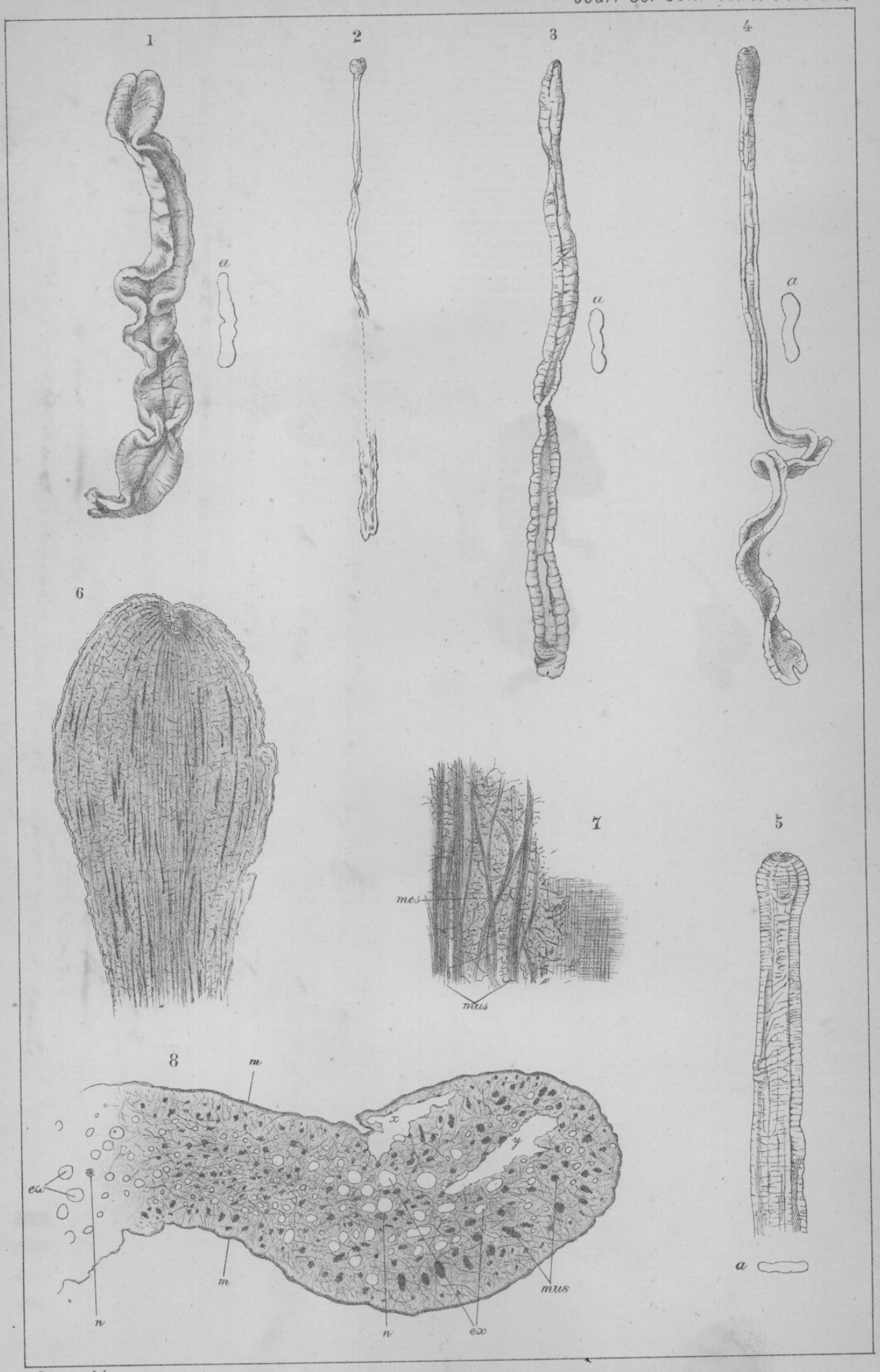
*Fig. 4.*—Bothr. liguloides in alcohol (Case V). Nat. size. *a*, outline of a cross-section near the hind end.

*Fig. 5.*—Anterior part of a Bothr. liguloides (Case VI). Twice magn. Preserved in alcohol. *a*, its supposed cross-section.

*Fig. 6.*—A horizontal section of the head-part of the worm represented in *fig. 4.* Colored by borax-carmine. About 10 times magn. Outlined by means of Camera lucida.

*Fig. 7.*—Peripheral part of the body of Bothr. lig. (*fig. 4*) stripped off, seen from inside. Drawn without the use of Cam. luc. *mes* = mesenchyma, *mus* = bundles of muscle-fibrils.

*Fig. 8.*—Part of a cross-section of the same worm. About 37 times magn.; stained with borax-carmine. *m*, *m* = the median plane. *n*, *n* = the two nerve-trunks in section. *x*, *y* = spaces produced by the tearing of mesenchyma. Cross-sections of numerous excretory vessels (ex) and of strongly colored muscle-bundles (mus) are to be seen.

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