

## BENIGN CYSTIC TERATOMA OF OVARY CONTAINING A HOMUNCULUS

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### Introduction

Mature cystic teratoma is a common type of ovarian tumor, accounting for 20% of all ovarian tumors<sup>9)</sup>. Histologically, the tumors is composed of well differentiated tissues of ectodermal, mesodermal and endodermal derivatives. Stratified squamous epithelium, sebaceous glands, hair follicles, sweat glands, teeth and smooth muscles are often observed<sup>2)9)</sup>. These various tissues, derived from the three germ layers, only rarely show an orderly organoid arrangement forming pituitary<sup>4)</sup>, prostate<sup>6)</sup>, developing eye<sup>7)</sup>, lung<sup>2)</sup>, breast tissue<sup>3)</sup> and even a part of a miniature human body (homunculus)<sup>1)11)</sup>. The present report is an additional case of a benign cystic teratoma of the ovary containing a structure resembling a human body.

### Case Report

A 33-year-old, gravida 2, para 2, Japanese woman visited our out-patient clinic on November 30, 1982, because of left lower quadrant abdominal pain. Her menstruation had followed a regular 30-day cycle since menarche at the age of 12 years. Her last menstrual period was 8 days from November 12, 1982. Her past and family medical history was unremarkable.

Pelvic examination revealed a freely mobile cystic mass in the left adnexal region. Abdominal ultrasound scan demonstrated a newborn head-sized cystic tumor containing a goose egg-sized solid mass in the small pelvis. Preoperative laboratory determinations, including blood counts, urinalyses, serum electrolytes, BUN, creatinin, liver enzymes, chest X-ray and electrocardiogram were all within normal limits. Immunological pregnan-

cy test was also negative. Her blood type was O and that of her husband type AB.

Laparotomy was carried out on December 16, 1982. The left ovary was replaced completely by a newborn head-sized cystic tumor which grossly appeared to be a dermoid. No pathological abnormality was found in the right ovary, uterus or other intraabdominal organs. The tumor of the left ovary was removed with the left tube. Postoperative recovery of the patient was uneventful.

### Pathological Findings

**Gross Findings:** The cystic tumor arising in the left ovary was 11×12×15cm and weighed 665g. Upon opening the specimen, an oval solid mass, measuring 5×6×8cm, was identified within a single cavity filled with sebaceous materials and hair. This mass was attached to the inner aspect of the cyst wall and resembled a miniature human body (homunculus) (Figs. 1A, 1B and 2). It was entirely covered by normal skin with fine hair. A head-like structure with long hair occupied the superior portion of the homunculus. Two pairs of appendage resembling rudimentary limbs protruded from both sides of the inferior portion appeared to represent the abdominal, lumbar, and sacral parts (Figs. 1A and 1B).

**Light microscopy:** The inner side of the cyst wall was composed of keratinizing squamous epithelium with sebaceous glands and hair follicles. Foreign body giant cells were present focally in the lining epithelium. The homunculus was also covered with keratinized squamous epithelium containing sebaceous glands, sweat glands, and hair follicles (Fig. 3). Beneath the normal skin, skull bone occupied the upper part of the homunculus, and contained brain tissues in the cavity, constitut-

ing a head-like structure (Fig. 4). A pair of developing eyes was also found in this region (Fig. 5). In the central axis of the lower part of the homunculus, a bony skeleton was present containing fatty bone marrow where many erythroblasts and granulocytes were admixed (Fig. 6). Two or three small bone were also observed in the extremity-like structures. However, no nails or joints were found. Small bundles of the peripheral nervous tissues and smooth muscles were also interspersed in the subcutaneous tissues of the homunculus. Moreover, an alimentary canal with an inner mucosal layer surrounded by two layers of the smooth muscle, was identified in the abdomen (Fig. 7). No chorionic villi, immature malignant tissues or other organoid structures were seen in any of the sections examined.

Cytogenetic analyses. Blood type of the hair and body fluid of the homunculus was O as determined by the adsorption-elution method<sup>10)</sup>. X-chromatin was present in 20% of the tumor cells by the cresyl echt violet staining method<sup>5)</sup>. However, Y-chromatin was not demonstrated by the Q-staining method using quinacrine mustard<sup>8)</sup>.

### Discussion

The recent review by Weldon-Linne and Rushovich described 19 cases of ovarian cystic teratomas containing homunculus in the English literature. The gross findings of these cases do not demonstrated a miniature whole but only a part of the human body such as upper and lower extremities, head and/or pelvis. In most of the homunculi, there was a predominance of lower extremities and pelvic structures in contrast to the ordinary dermoid cysts, where supra-diaphragmatic tissues such as teeth, brain, respiratory epithelium and thyroid are common<sup>2)9)11)</sup>. Visceral organoid tissues are generally absent<sup>11)</sup>. However, the present case was composed of structures suggestive of a head, four extremities, trunk, and pelvis, showing a close resemblance to a miniature human body. Microscopically, the homunculus which was covered with normal skin and its appendages, contained brain tissues within skull bone, rudimentary eye, bony skeleton, nervous plexus and intestine. Although such tissues appeared sporadically in ovarian teratomas<sup>2)7)9)</sup>, no homunculi combining them normotopically have been reported in our

knowledge.

Histogenetic consideration of a homunculus in ovarian teratoma must evoke great interest among gynecologists and pathologists. Its occurrence may be due to an ectopic pregnancy, included monozygotic twin or parthenogenesis<sup>11)</sup>. Although chromosome analysis of the cultured tissue from the homunculus was not performed, we demonstrated the presence of X-chromatin in 20% of the tumor cells. The blood type of the homunculus was O, while that of the patient O, and her husband AB. Moreover, the patient had a normal menstrual cycle and a negative serological test for pregnancy. No placental tissue was identified in the tumor. These findings suggest a parthenogenetic or monozygotic twin origin. However, true instances of fetus in fetu generally involve monogonadal midline structure in young infants<sup>11)</sup>. Consequently, the present case might be of a parthenogenetic origin and show an extreme level of differentiation of which a teratoma-stem cell is capable, although whether it originated from a somatic cell or germ cell still remains to be investigated.

### Acknowledgement

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### Captions for Figures

Fig. 1A. Anterior view of homunculus (right) and dermoid cyst (left).

Fig. 1B. Lateral view of the homunculus.

Fig. 2. Cut surface at midline of the homunculus, showing skull bone(A), brain tissue(B), eyes(C), bony skeleton(D), pelvis(E) and alimentary canals(F).

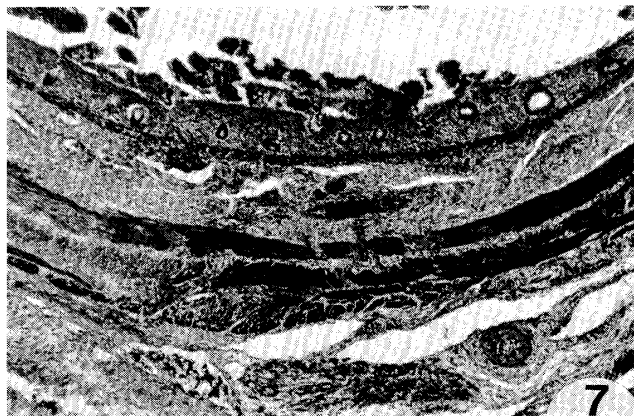
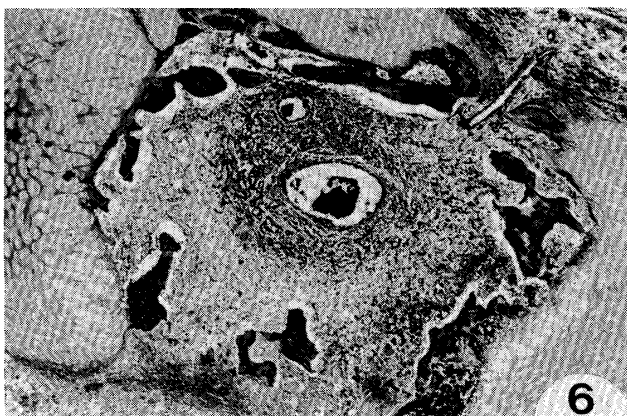
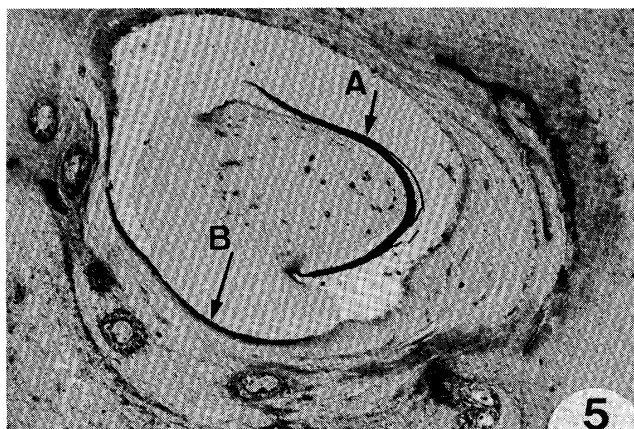
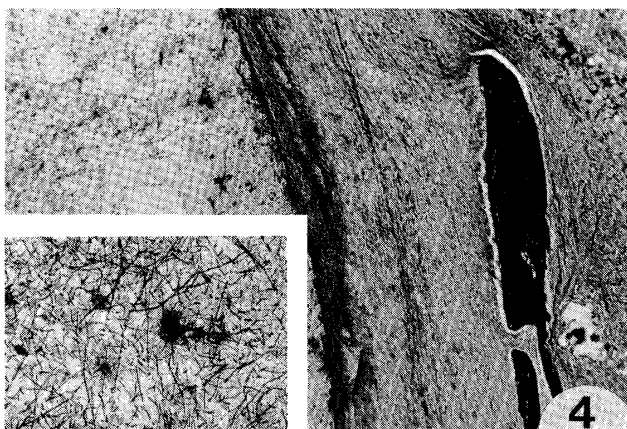
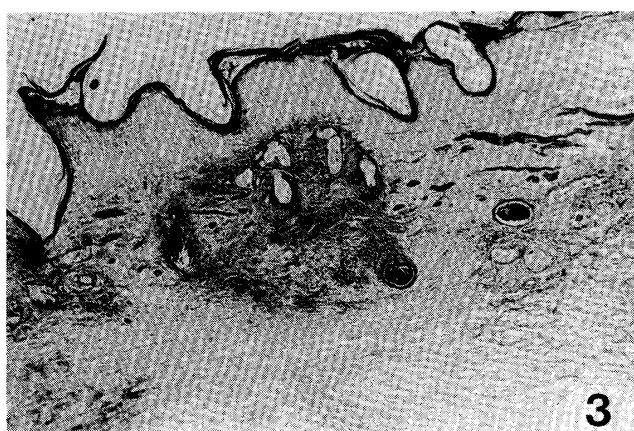
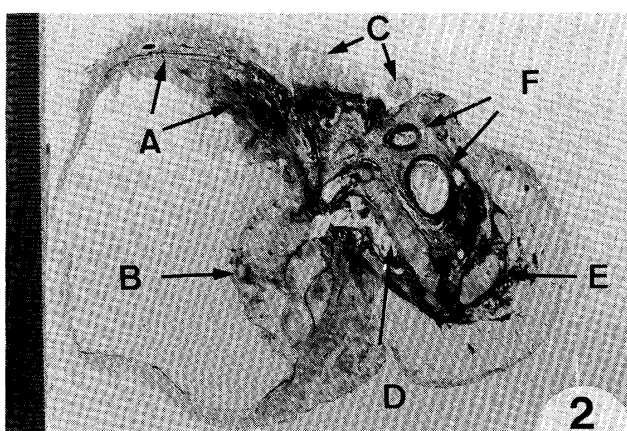
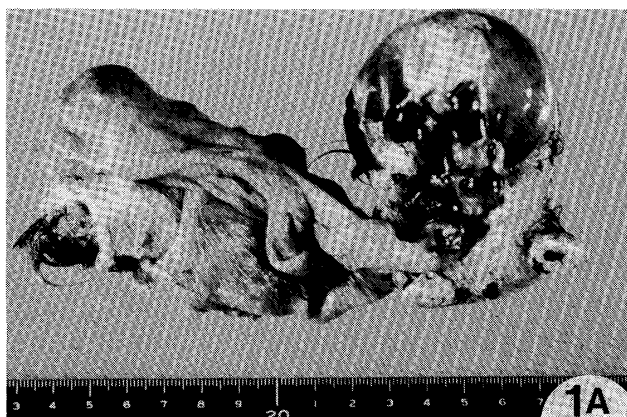
Fig. 3. Keratinized squamous epithelium with hair follicles and sebaceous glands. Hematoxylin & eosin  $\times 15$ .

Fig. 4. Brain tissue (left) in the cavity surrounded by bone (right). Hematoxylin & eosin  $\times 15$ . Inset; High power view of the brain tissue. Plexus of nerve fibers and neural cell bodies are seen. Phosphotungstic acid & hematoxylin  $\times 100$ .

Fig. 5. Developing eye. Rudimentary lense(A) and pigment epithelium(B). Hematoxylin & eosin  $\times 15$ .

Fig. 6. Bone marrow surrounded with bone.

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Hematoxylin & eosin  $\times 15$ .

Fig. 7. Alimentary canal. Hematoxylin & eosin  $\times 20$ .

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