A TERATOMA OF THE ABDOMINAL CAVITY.

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PLATES XIX-XXII.

Teratomata have always attracted attention by their strange appearance, but of late years they have given rise to much investigation on account of the promise they hold out of elucidating the etiology of the true tumors.

The tumor to be here described was a solid teratoma of the abdominal cavity, in itself a sufficiently rare occurrence, and, besides, was remarkable on account of the great variety of tissues which it contained. Clinically also it took an unusual course, for, after being operated on and apparently entirely extirpated, it rapidly recurred.

In May, 1893, Dr. W. F. McNutt was called by Dr. J. S. Stone to see a patient, a girl, 12 years of age, who was suffering from an abdominal tumor. The patient herself had noticed ten months previously that she was growing stouter and that her clothes no longer fitted her. The rate of growth had at first evidently been slow, but latterly very rapid, as Dr. Stone for a few days previous to the consultation had been able to make out an increase in girth of one and one-half inches a day. Both previous to and after the enlargement of the abdomen the patient had always enjoyed good health. There was no loss of flesh and she had never suffered any pain. There was nothing in the family history to
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indicate any hereditary inclination to the formation of neoplasms. One grandfather had died of tuberculosis. There was no history of multiple pregnancies on either side of the house.

On physical examination the abdominal veins were found dilated and the abdomen was considerably enlarged. The swelling was general, and much of it was evidently due to ascites. By palpation and percussion a solid mass could be made out in the right side of the abdomen.

On May 27, 1893, the abdomen was opened, and the tumor, together with about two gallons of slightly bloody ascitic fluid, was removed.

When Dr. McNutt brought me the specimen he said that the tumor was found on the right side of the abdomen and in the peritoneal cavity; that its general shape, as it lay in the abdomen, was long, narrow and curved, and that the shorter arm of the curve lapped around the caputcoli, while the longer arm extended up along, and was attached to, the outer side of the ascending colon throughout nearly its whole length to a point nearly as high as the liver; that it shelled out easily, and when it was taken away the operation seemed to be complete, as there was no appearance of any other diseased tissue in the abdominal cavity. Dr. McNutt also told me that in removing the tumor no large blood-vessels were encountered, the vessels being only such as run in brittle adhesions; and while that part of the peritoneum from which the growth was detached was of course raw and bleeding, the rest of the peritoneal surface, as far as could be seen, was perfectly smooth, glistening, and normal. After the operation the abdominal cavity was closed and the patient made a speedy and uneventful recovery for the time being.

A photograph of the tumor is shown in Plate XIX, Fig. 1. When I first saw the original tumor, a few hours after the operation, it lay in a basin, a formless, quivering, semi-fluid, jelly-like, shiny mass, with white, greasy patches on its surface, and small pieces of bone, the size of a silver half-dollar and less, scattered in its substance. After being in alcohol for some time the tumor became firmer and assumed more definite outlines. It weighed two pounds, and resembled very strikingly a cerebral hemisphere. Like this it was elongated and lobulated, and all the lobules were smooth and grew out in one direction as if they sprang from a common elongated base. Two of the fissures cut into the tumor so deeply that they divided it into three parts, a large central portion with a smaller portion at each end. These three lobes, besides being united at their bases, were con-
nected together with strings of tissue thrown across the fissures. On cutting into the tumor it was soft, and in many places honeycombed with minute cysts, many of which were so large that they could be seen with the naked eye. The largest cyst was about the size of a cherry; then there were a number of pea-sized cysts, but by far the greatest number were very minute and could only be made out with a lens or a microscope. In spite of this cystic structure, however, the tumor, looked at as a mass, was a solid growth, a point of a good deal of importance considering the rarity of solid dermoids in the interior of the body.

The term solid teratoma or dermoid does not mean that the tumor does not contain cysts. In fact such teratomata contain large numbers of cysts, but these cysts do not interfere markedly with the general solid appearance of the neoplasm, while in the so-called cystic neoplasms of this class the hollow character of the tumor is the main feature.

Almost the whole of the outer surface of the tumor was covered with a distinct epithelial membrane. In many places this covering was clearly skin (Plate XIX, Fig. 2), being provided with fine downy hair and sebaceous glands. In other situations it was devoid of hair and looked more like the mucous membranes that are covered with flat, stratified epithelium. This epithelial covering, provided or un-provided with hair, followed the irregularities of the tumor down into the deepest fissures. On some parts of the free surface evidences of an epithelial covering were not distinct to the naked eye, but even in such places on microscopic examination there could be made out occasional indications of a papillary layer, or groups of cells indicating probably the remains of an epithelial covering.

In most dermoids the skin lines one or more of the cysts of which the tumor is composed; in this tumor, however, nearly all the cutaneous integument was found on its external surface, and only one small cyst was discovered lined by skin.

The extent of this cutaneous envelope, its position on the outside of the tumor, the ease and apparent completeness with which the tumor was shelled out of the peritoneal cavity, were noteworthy facts,
making it seem probable that this teratoma was an independent individual enclosed, to at least a great extent, in its own skin.

Although a number of small pieces of bone were present in the tumor, none of them resembled at all clearly in shape any definite human bone. Nor did the islands of cartilage here and there met with (Plate XX, Fig. 4) show a form recognizable as that of any of the cartilages or bones of the body. There was nothing in the tumor therefore which could justly be called a skeleton, nor could it be made out, as is sometimes possible in teratomata, that the bones were arranged according to the conformation of the foetal skull. But, as will be shown further on, one of the pieces of bone may have represented a fusion of some of the bones in the neighborhood of the ear. All the pieces of bone, as well as the islands of cartilage, were bound in the tumor by strong attachments, so that it was difficult to free them from the surrounding tissues. The largest piece of bone had a hollow in it enclosing an empty tube about half an inch in diameter. The total weight of the pieces of bone amounted to about one ounce. No teeth were found anywhere in the growth.

As has been said, in many places the tumor was honeycombed and spongy with minute cysts, but in other regions there were very few cysts, and in still other areas of considerable size there were none at all. Some of the cysts communicated with one another. The majority were filled with a mucoid substance, and none of them were filled with sebum. No polypoid or warty projections were found jutting into the cysts. Some of the cysts had a refractive yellowish lining as smooth as peritoneum, and one of them was found lined with skin.

The appearance of the tissue in among the cysts and between the bones and masses of cartilage varied considerably. In some places it was fairly dense and tough and apparently fibrous; in others, much softer and even jelly-like. In many parts of the stroma there were streaks of black or brownish-black pigment, and in one place a sheet of tissue, measuring 1 x 2 cm., covered on one side by a thin, continuous, even, black pigment layer, was found attached by one of its edges to a bone in the centre of a large lobe. The other relationships
of this sheet of tissue could not be determined owing to the disturbance of the parts in getting out the bone. This sheet of tissue is interesting because, on microscopic examination, it was shown to be a part of an eye.

Pieces of the tumor, hardened in alcohol, were embedded in celloidin, sectioned and stained by various methods. A study of the finer structure of the growth revealed even more complicated relations than the gross appearances had indicated. The growth contained tissues and portions of organs corresponding in embryonic origin to all three of the germinal layers. Corresponding to the epiblast there were skin with cutaneous organs and appendages, central nervous system, peripheral nerves, and rudiments of eye structures. The hypoblast was represented by mucous glands, tubes and cysts with epithelial lining and surrounded by smooth muscle. The mesoblastic tissues consisted of bone, cartilage, white fibrous tissue, yellow elastic tissue, mucoid connective tissue, adipose tissue, smooth muscle fibre, and blood-vessels.

The best developed organ in the teratoma was the skin with its hairs and attendant sebaceous glands. This is a regular phenomenon in such tumors, and it is usually such a prominent feature as to give them the name dermoid. This predominant participation of the skin is supposed to be due to the fact that the ectoderm has the start of the other fetal layers in development. The skin in the case under consideration was so far advanced that there was at any rate a commencement of defluvium of the hair (Plate XX, Fig. 6). The embryonic nervous system, the eye-structures, and structures belonging to the intestine were also remarkably developed. Microscopic examination of the skin of the surface of the tumor showed the presence of nearly all the constituents of normal skin—epidermis, corium, tela subcutanea, hairs with hair follicles, sebaceous glands, and blood-vessels. In one section of skin a definite bundle of mature medullated nerve fibres was found. As so frequently happens in dermoids, no sweat glands were discovered.

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system in dermoids seems to be a common occurrence, for of nineteen consecutive cases of dermoid cyst of the ovary examined by him he found such tissue in eighteen. Professor W. H. Welch first drew my attention to the presence of tissue of the central nervous system in this tumor, and its description will be left to Dr. Lewellys F. Barker (p. 284 et seq.).

It has been previously mentioned that a sheet of tissue was found covered by a thin, continuous, even, black layer, and attached by one of its edges to a bone in the centre of a large lobe. Transverse sections of this sheet of tissue showed the black layer to correspond to the pigment epithelium of the retina. There were flat polygonal cells filled, excepting the nucleus, with dark brown pigment, and these cells were so placed as to form a mosaic (Plate XXII, Fig. 11). This epithelium lay on a slightly pigmented connective tissue that was much firmer than the soft connective tissue usually encountered in this tumor. No definite tunica vasculosa (chorioidea) could be made out. The pigment epithelium could have been nothing but the lamina pigmentosa retinae, as there is no other structure in the human body that resembles it; and the firm, slightly pigmented, in places distinctly laminated connective tissue on which it lay was probably the sclera.

Structures corresponding evidently to fetal intestine were found. This could be seen as an oval tube cut transversely and lined with exquisite cuboidal epithelium. Numerous villus-like processes projected into the lumen. Beneath the epithelium there was a layer of cellular connective tissue, and the whole tube was surrounded by a definite tunica muscularis consisting of bundles of smooth muscle fibres, cut chiefly obliquely, but in part transversely (Plate XXI, Fig. 8).

Another tube lined with cylindrical epithelium lay close to a small mass of cartilage. This might have been embryonic trachea, though no smooth muscle could be made out in its wall, nor did the lining epithelium bear demonstrable cilia. Cysts with mucus in the lumen and lined by ciliated columnar epithelium with beautiful goblet cells were, however, also found, and doubtless corresponded to some part...
of the respiratory tract. Nothing recognizable as liver or pancreas
was found.

The tube, which has already been mentioned as having been re-
moved from a concavity in the largest piece of bone, and which was
about 1 cm. long and 7.5 cm. in diameter, was examined microscopi-
cally. The wall of the tube contained a great deal of elastic tissue, the
delicate fibrils of which were well brought out by the orcein stain.
In some places the elastic tissue was arranged in the form of alveoli
containing cartilage cells. There were also numerous large hairs in
the wall of the tube. This tissue was therefore elastic cartilage, and as
it was a tube situated in the hollow of a bone, and as there were also
large hairs in the wall of the tube, this elastic cartilage may have been
from the external ear. It is also of interest that this hollow bone
with its tube containing elastic cartilage was situated near the sheet
of tissue containing the flat pigmented epithelium of the eye already
described. The bone was too large for the quadrate bone, but it may
have represented a fusing together of several bones in the neighbor-
hood.

The stroma of the tumor, made up of mesodermal structures, was
not inferior to the epithelial elements in point of variety. There were
little masses of hyaline cartilage showing concentric arrangement of
cells toward the periphery, and each mass was surrounded by a well
marked perichondrium; bone with bone corpuscles and canaliculi;
mucoid connective tissue; white fibrous tissue; yellow elastic tissue;
adipose tissue; embryonic connective tissue, resembling round-celled
and spindle-celled sarcomatous tissue; and long bands of smooth muscle
fibre. Then there was a great deal of intracellular and diffuse (inter-
cellular) dark brown, granular pigment; and the tumor was well sup-
plied with small blood-vessels, arteries, capillaries, and veins, having
for the most part embryonic walls. No trace of a heart was found.
The blood in the blood-vessels had all the characteristics of adult blood.
No nucleated red blood corpuscles were seen. The white and red
blood corpuscles appeared to be present in normal proportions.

No striated muscle fibre was found in the tumor. All the struc-
tures in the interior of the tumor were shuffled up together without
any ascertainable law or order. There was an immense variety in
the kind of tissue lining the cysts; some, as said, were clothed with
flat, stratified epithelium; others with simple columnar epithelium;
others with stratified columnar epithelium; others with ciliated column-
ar epithelium; and still others with goblet cells (Plate XX, Fig. 3).
There were also many irregular cysts or alveoli containing stratified
columnar epithelium forming wild-looking figures and broken fes-
toons scattered about in a disorderly fashion in the alveoli, and without
a basement membrane, as in adeno-carcinoma.

The cyst situated near the choroidal structure, before mentioned,
was lined with skin provided with hair and sebaceous glands. This
was the only unequivocal dermoid cyst within the tumor, as all the
rest of the skin was on the outside of the tumor instead of lining a
cyst. There were other cysts lined with flat stratified epithelium, but
this, of course, might have been mucous membrane and not skin.

In many places there were masses and strings of round or polyhedral
epithelial cells forcing their way between the fibres of the stroma of
the tumor as in ordinary carcinoma (Plate XX, Fig. 7). There was
also the formation of epithelial pearls and of onion-like bodies, which
so frequently happens when flat epithelium grows in a confined space,
as notably in flat-celled epithelioma.

Everything went well after the first operation for a few weeks, and
the child appeared to enjoy her usual good health, but in July, a
little over a month after the first operation, the abdomen was seen to
be enlarging again.

A second operation was performed July 15, 1893, when a mass
about as large as that removed at the first operation was taken away.
At the first operation the tumor shelled out easily and the operation
appeared to be complete. The second operation, however, Dr. McNutt
told me, was manifestly incomplete. The friable, glairy, recur-
cent tumor, which was located mainly on the right side of the peri-
toneal cavity, had to be removed in pieces and could not be enu-
cleated, and much of the diseased tissue could not be removed at all,
as it had spread out laterally and involved a much larger surface of
the peritoneum. At this operation a considerable quantity of bloody serum was found in the abdominal cavity, but not nearly so much as at the first operation. The abdomen was closed, and the patient died July 18, 1893, 51 days after the first operation.

After death Dr. McNutt made a simple incision into the abdomen and found that the diseased tissue had spread as a continuous layer of glairy, friable tissue over the visceral peritoneum of the lower part of the abdomen behind the bladder and nearly across to the left side of the abdominal cavity. The parietal peritoneum showed no evidence of disease. There did not seem to be any involvement of the intestinal wall excepting the growth of the neoplasm in the peritoneum covering the intestine. The abdomen only was opened and none of the viscera were cut into; the liver, the spleen, the kidneys, the uterus and ovaries were not incised, but they all appeared to be normal as far as could be judged from the external surface. The thoracic cavity was not examined. Very little fluid was found after death in the abdominal cavity.

The quick recurrence of the tumor in loco, its rapid growth, its spread by lateral extension, the appearance of the soft, glairy, brittle tissue removed at both operations, as well as the occurrence of bloody fluid in the peritoneal cavity in both instances, were enough to establish the identity of the recurrent growth and the original tumor, and the microscopical examination was confirmatory of this in every respect.

The general character of the recurrent growth, like the original, was that of a solid tumor richly studded with cysts, most of which were microscopic in size. A great many of the cysts were, however, as large as a pea, and some of them of the size of a walnut. As in the original tumor there was a great variety in the kind of tissue lining the cysts, and one cyst was sometimes clothed with many varieties of epithelium; for instance, columnar epithelium with nuclei far from the base of the cell was continuous with columnar epithelium with nuclei near the centre or near the base. This in turn was continuous with ciliated epithelium, which was finally continuous with round or polyhedral epithelial cells. As another example, an irregular cyst
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was lined on one side with stratified columnar epithelium, and the opposite side was covered with stratified flat epithelium, while a great number of flat epithelial cells were desquamated into this cyst cavity. In another cyst, stratified columnar epithelium tapered off into a single layer of cuboidal epithelium. There was a great number of well formed tubes lined entirely with stratified columnar epithelium. Many cysts were lined either partly or entirely with a single layer of beautiful columnar epithelium, each cell of which had a single nucleus placed far out at its distal extremity. Examples of this kind of epithelium were found both in the original and in the recurrent tumor, and were at first thought to represent the membrana adamantina of the fetus, but a hair was discovered in the original tumor, in which the outermost cells of the external root sheath had exactly these characteristics; the cells formed very long columns, much longer than they usually do, and each cell was provided with a single nucleus that lay far out at its distal extremity (Plate XX, Fig. 5). Therefore, although the cells that looked like those of the membrana adamantina might really have been so, yet there was a possibility that they might have belonged to hair follicles where the development stopped with the formation of the external layer of the external root sheath. In further support of the pilary origin of these cells many follicles were detected having about the diameter of a hair follicle, and lined with a single layer of columnar cells, with their nuclei far out toward the free extremity of the cell.

The recurrent growth, like the original, contained tissues from all three of the germinal layers. Indeed nearly all the structures met with in the original tumor could be found in various parts of the tissue removed at the second operation. There were large masses of tissue of the central nervous system. The endodermal structures were scattered through the other tissues in considerable amounts, for instance, cysts lined with ciliated and columnar epithelium, and spaces lined with goblet cells.

The mesodermal stroma of the recurrent tumor was similar to that of the original, and consisted of little globes of hyaline cartilage surrounded by perichondrium, mucoid connective tissue, adipose tissue,
well formed fibrous tissue, embryonic round-celled and spindle-celled connective tissue resembling sarcomatous tissue. One small flat piece of bone was removed with the tissue taken away at the second operation, but of this more hereafter.

In the immediate neighborhood of some of the tubes lined with lamellated epithelium, stratified columnar epithelium could be seen to form the wildest wreaths and broken festoons, scattered in irregular cavities much as in adeno-carcinoma. There were also places where masses of round epithelial cells could be seen invading the stroma of the tumor and pushing along between its fibres as in ordinary carcinoma. There was one particularly striking example of the metaplasia of columnar into flat epithelium (Plate XXI, Fig. 9). This was found where several follicles lined with simple columnar epithelium appeared to meet. The epithelial walls of two of these follicles were imperfect, and the epithelium at these imperfect points, as well as that lining the mouth of a third follicle, seemed clearly to have changed into flat epithelium with pearl formation. Of course there was a possibility that an invasion of flat epithelium might have taken place, but any one looking at the specimen would be surely drawn to the view that it was an instance of metaplasia of columnar to flat epithelium. Nests of flat epithelium containing pearls were found in the recurrent as well as in the original tumor (Plate XXI, Fig. 10).

That this tumor was a teratoma there can be no doubt, as is shown by the variety of tissues entering into its structure. It being settled that the tumor was a teratoma, the determination of the exact locality in which it grew is the next point to be considered.

As mentioned by Ernst Schreiber,* teratomata are oftenest found in the region of the head or sacrum. The next most frequent locality for their growth is the genital glands, and therefore the possibility of the tumor under discussion having originated in the ovaries must be carefully considered. But decidedly against this view is the fact that the growth lay high up in the abdominal cavity, and well away from the ovaries, which were found perfectly healthy and in their

* Virchow's Archiv, cxxiii, 165.
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usual situation. Furthermore, no strands of tissue binding the tumor to the ovaries were encountered during the operation for removal.

Another remote possibility presents itself, that, as in Kolaczek's case, tissues from an ovarian dermoid might have been transplanted from the ovary to the peritoneal cavity and have grown there; but in such an event the original dermoid or teratoma would have been quite obvious in the affected ovary, while in the case under consideration the ovaries were expressly stated to have been found healthy.

According to Wilms, very few instances of dermoid have been reported as occurring in the abdominal cavity proper, excluding those of the pelvis, most of which were single cysts, and judging from the cases reported in the literature instances of solid abdominal teratoma must be exceedingly rare. But Meckel, Bonfigli, Marchand and Maydl have each reported abdominal growths of a more complicated structure than simple dermoid cysts. Meckel's tumor was situated on the diaphragm and contained twenty-one pieces of bone, four teeth and some hair. Bonfigli's case was that of a pear-shaped growth occurring in a strand of tissue between the liver and the stomach. It contained a piece of true bone in which were found two teeth, and nineteen more teeth lay free in the tumor cavity. The main cyst was lined partly with cutis. There were other cysts filled with mucus, but they were not further described.

One of the most complicated teratomas of the main abdominal cavity was described by Marchand. It was situated behind the peritoneum, and on the aorta and left kidney, and contained a skull cavity, intestine, male genitals, prostate and cysts lined with ciliated epithelium. This case was therefore very far advanced developmentally, and was analogous to the following reported by K. Maydl: In a well developed but very anemic man, 19 years of age, a tumor

* Virchow's Archiv, lxxv, 39.
† Loc. cit.
‡ Deutsch. Arch. f. Physiol. (1815). Quoted by Wilms.
§ Ref. Schmidt's Jahrb. clxx (1876), 180. Quoted by Wilms.
was found occupying the greater part of the pelvic cavity. Laparotomy was performed and the tumor was seen to lie between two folds of the root of the mesentery, and consisted of a well formed trunk and limbs. The head was absent, and in its place there was a tuft of hair 50 cm. long. The fetus was enclosed in a firm amniotic sac that contained also some oily fluid. A thickening of the amniotic membrane, connected with the superior mesenteric artery, showed histologically the structure of the placenta. The umbilical cord was entirely wanting. The tumor was classed as a dermoid and was assumed to be a rudimentary twin, an instance of foetus in foetu.

My case evidently is to be placed in the same category with the four cases just mentioned, and it is almost unnecessary to draw attention to the parallelism between the two last mentioned tumors, Marchand’s and Maydl’s, and the growth we are now considering. All three have made the impression on those examining them that they were dealing with tumors that were in reality independent individuals. All three were situated in the abdominal cavity; two of them, Marchand’s and Maydl’s, retroperitoneally, and one of them, the case in hand, intraperitoneally.

The age at which teratomata and dermoid cysts develop appears to vary much. Wilms, in his article previously referred to, speaking more particularly of dermoids of the ovary, makes the statement that they may appear at any time of life between intrauterine existence and the seventieth year, although the number occurring in childhood and old age is small, as the majority of cases occur during the second and third decade. In the case here reported the patient was twelve years of age. As the child had not yet begun to menstruate, no connection between a physiological increase of blood supply to the abdominal organs and the growth of the tumor could be definitely proven.

The relatively great development of the ectodermic structures (integument and nervous system) in the tumor is quite in accord with other knowledge, for we have evidence that both ontogenetically and phylogenetically the ectoderm takes precedence of both the endoderm and mesoderm. In speaking of dermoids of the ovary, Wilms says that "the advanced development of the ectoderm as well as the pre-
ponderance of the tissues of the head in these tumors are easily explained by the early differentiation, both of the epiblast and of the cephalic extremity in the fetus." Wiedersheim,* in referring to the relative age of the ectoderm, states that the dermal skeleton is phylogenetically older than the endo-skeleton. Its relative age is shown not only by palaeontology but also by ontogeny, e. g. the young file fish is provided with a complete dermal armor at the time when the ossification of the primordial cranium has hardly begun.

Our case is somewhat remarkable in that it is the third instance on record where the hair of a dermoid has been found growing free into the abdominal cavity. The first case is that reported by Kolaczek,† in which the patient, who was a spinster, 45 years of age, was operated on for a dermoid cyst of the ovary. At the operation a number of lentil-sized yellowish nodules were found scattered over the visceral and parietal peritoneum. Many of these nodules were pierced through their centre by a fine hair, and were regarded by Kolaczek as miliary metastases from the dermoid of the ovary. According to Wilms, Fraenkel‡ has reported a similar case.

Until recently the existence of nervous tissues in dermoids and teratoma has been considered to be rare. Baumgarten,§ writing in 1887, made the statement that only six authenticated observations of the finding of central nervous tissues preceded that of his case. Recently, however, it has been shown that such tissue is really of extremely common occurrence, and one must believe that its supposed rarity in the earlier cases is due to the fact that it was not recognized. Wilms believes that tissues of the central nervous system are almost always, if indeed not constantly, present within ovarian dermoids, for he found it in 18 out of 19 consecutive cases which he examined. He believes furthermore that the finding of such tissue in a teratoma is a good argument for considering the particular tumor in which it is found as an independent individual.

† Loe cit.
‡ Wien. med. Wochenschr., 1883.
§ Virchow’s Archiv, cxxii, 515.
Wilms, in his investigations, frequently ran across corpora amylacea, and he states that they were of great value to him in locating the tissue of the central nervous system. In the very numerous sections of our tumor examined no corpora amylacea were found.

Eye structures have not been encountered very frequently, as up to the present I have been able to collect from the literature records of only fifteen cases where pigmented epithelium has been demonstrated in teratomata and dermoids. These cases have been reported by Marchand,* Baumgarten,† Lazarus,‡ Wilms § (3 cases), Kappeler,¶ Pommer,‖ Rippmann,** Verneuil,** Kümmel,** Benno and Martin Schmidt,** O. Spöndly,** van Duyse,†† and Lovett and Councilman.‡‡ If the case in hand is added to the above mentioned we have a total of sixteen where pigment or pigmented epithelium has been found in teratomata.

The question of the recurrence of the growth and its malignancy has next to be considered. It has seemed to me worth while in this connection, on account of the great interest of this side of the subject, to review with some care the cases of teratoma which have been reported as recurrent or malignant.

The instances where teratomata have been found, either clinically or microscopically, to be malignant, or to have undergone malignant degeneration, are not numerous. This is somewhat surprising when one reflects that teratomata have been considered to be misplacements of foetal tissue, and that true tumors, the malignant ones included, are also, according to Cohnheim’s theory, supposed to originate from misplacements or implantations of foetal tissue. The experiment of engrafting foetal tissue into animals with a view to causing tumor formation has been tried fruitlessly, but, as Grawitz has said, teratomata fur-

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* Loc. cit.
† Loc. cit.
‡ Inaug.-Diss., Glessen, 1888.
§ Loc. cit. and Ziegler's Beiträge, xix.
¶ Inaug.-Diss., Zurich, 1896.
** Cited by Kappeler, loc. cit.
‡‡ Journal of Experimental Medicine, ii (1897), 427.
nish an example of the implantation of foetal tissue better than any artificial implantation could, for in teratomata the implantation of foetal tissue occurs in foetal tissue, while artificially the grafting must be done at least on post-gestational tissue. The instances I have collected in which teratomas have been malignant, or have undergone malignant change, are as follows:

I.—Czerny's case.* A congenital sacral tumor, consisting for the most part of fat, and containing cysts lined in some instances with ciliated, in others with flat, epithelium. The patient was 55 years of age, and the tumor underwent malignant change after repeated traumatisms. Czerny demonstrated the metaplasia of ciliated into flat epithelium, with pearl formation. After extirpation there was local recurrence, followed by metastasis of flat epithelial cancer in the inguinal glands.

II.—Virchow's case.† Teratoma myomatodes mediastini. This was a large, lobulated, and, for the most part, smooth tumor occupying the right side of the chest of a man, 22 years of age. The tumor contained striated muscle tissue, sarcomatous tissue, and masses of hyaline cartilage, surrounded by well marked perichondrium. There was a considerable number of cysts in the tumor, some of which resembled proliferating ovarian cysts. Other cysts were lined with skin, others with columnar ciliated epithelium, and still others with simple columnar epithelium. In the middle, more compact mass of the tumor there was tissue having the structure of carcinoma. A metastatic nodule in the left third rib was cystic (simple cysts), sarcomatous and carcinomatous, and contained striated muscle fibres, and a portion of this metastasis recalled vividly the structure of foetal lung. The structure of nodules in the liver and kidneys was not determined.

III.—Jores' case.‡ Combination of an intrathoracic dermoid cyst with malignant cystosarcoma of the left lung. There was nothing of striking interest in the dermoid cyst itself. Its walls, however, were cystic, and these cysts evidently arose from the sebaceous and sweat glands of the dermoid. The intercystic stroma contained many islands of hyaline cartilage, much smooth muscle fibre, and much spindle-celled tissue. Besides the above mentioned cysts there were others more irregular in shape and lined with cylindrical epithelium. This cylindrical epithelium did not sit on a basement membrane, but sat directly on the

* Arch. f. klin. Chir., x, 894.
† Virchow's Archiv, liii, 444.
‡ Virchow's Archiv, cxviii, 66.
spindle-celled tissue. The metastatic nodules in the right lung had exactly the same structure as the main tumor.

IV.—Virchow and Litten's case.* A case of androgyny with malignant teratoid cystoma of the right ovary and bilateral cystic hydrocele of the processus vaginalis of the peritoneum. A multilocular cystic tumor, apparently of the right ovary, developed in a female, 16 years of age. Here and there in the tumor were small dermoid cysts. Medullated nerve fibres, smooth muscle fibres, and masses of hyaline cartilage provided with perichondrium were present in the stroma. In some places the tumor was sarcomatous (round-celled) and in others myxomatous.

In the metastatic nodules in the liver there was neither epidermis nor hair nor cartilage. There were small and large cysts, like ovarian cysts, and the stroma corresponded to the sarcomatous and myxomatous portions of the original tumor. No carcinomatous structure was seen.

V.—Wernitz's case.† A teratoma of the right ovary occurring in a woman, 25 years of age. The tumor had a very complicated structure. The main part of it was formed of small colloid and dermoid cysts. Some of the cysts were lined with simple epithelium, others with columnar epithelium. The dermoid cysts were lined with stratified horny epithelium, and provided with hair and sebaceous and sweat glands. In many places in the connective tissue stroma there were hyaline cartilage and small lamellae of bone. In other places there was large-celled sarcomatous tissue, and here and there were groups of epithelial nests.

At the autopsy metastatic nodules were found in the peritoneum, lungs, kidneys and liver. These metastatic nodules consisted of richly cellular sarcomatous tissue that resembled closely that of the primary tumor.

VI.—Keller and Kramer's case.‡ The patient was a multipara, 20 years of age, who suffered from a quickly growing, nodular tumor of the right ovary. There was much ascites. The tumor was surrounded by a hard fibrous capsule, and scattered throughout it were many small islands of cartilage and pieces of bone. There were some cysts, one the size of a fist, but the tumor was for the most part solid. Microscopically there were sarcomatous tissue, fat, cartilage and bone. Some of the cysts were lined with stratified epithelium, and others with stratified epithelium that gradually changed into cylindrical epithelium. These cysts were provided with sweat and sebaceous glands and hair. Other cysts were lined entirely with simple or stratified epithelium and had no

* Virchow's Archiv, lxxv, 329.
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sweat glands, etc. Smooth muscle fibre was present, but no striated muscle fibre and no nerve tissue.

After the operation there was a quick return of the ascites and a recurrence of the tumor.

The patient died eleven months after the operation, and the post-mortem examination showed an extensive return of the tumor in the peritoneum and metastases in both lungs. Microscopical examination showed the secondary tumors to be sarcomatous, and in one of them, situated in Douglas' sac, islands of cartilage were found.

VII.—Emanuel's case.* A girl, 15 years of age, was operated upon, January 30, 1892, for a tumor of the left ovary, the size of a man's head. A quantity of brown ascitic fluid was found in the peritoneal cavity. There were a great number of small cysts scattered throughout the tumor, some lined with skin provided with hairs and sebaceous glands, others had no lining membrane, others were clothed with stratified cylindrical epithelium, and others with simple cylindrical epithelium. Some cysts were lined with cylindrical epithelium that gradually changed to cuboidal or flat epithelium. One cyst was lined with ciliated stratified epithelium. The mass of the tumor consisted of round-celled sarcomatous tissue divided off into fields by strands of connective tissue. There were also smooth muscle cells, small oval or round masses of hyaline cartilage provided with perichondrium, fat, mucoid tissue, and black and golden-yellow pigment partly intra- and partly intercellular.

The recurrence was speedy, for in five weeks there was a demonstrable ascites, and in a fortnight more there were unequivocal symptoms of the presence of an abdominal tumor. The patient died, June 2, 1892, four months after the operation.

There was a recurrence in loco, and the right ovary, that at the time of operation seemed to be perfectly healthy, was now changed into a tumor the size of two fists. There were metastatic tumors in the subcutaneous cellular tissue of the abdomen, on the parietal and visceral peritoneum, on the under surface of the diaphragm, in both omenta, the suspensory ligament of the liver, and on the lower surface of the liver. Microscopically, the recurrence in loco and the tumors of the omenta were identical with the original growth. The tumor of the right ovary was also similar, but with the addition of blood cysts. The tumors of the peritoneum resembled the sarcomatous stroma of the original neoplasm. The tumor lying in the subcutaneous cellular tissue of the skin of the abdomen was a surprising structure, and is of the utmost interest.

to us, as it was not simply a sarcoma, but a compound tumor like the
original neoplasm of the left ovary. It had a complete connective tissue
capsule. The stroma of the tumor was, as in the original, composed of
round and spindle-celled sarcomatous tissue, and there were numerous
cysts, lined with various kinds of epithelium, some with simple or strati-
fied columnar, others with flat or cuboidal epithelium. As in the original
tumor, there were solid columns of flat epithelial cells, and some of the
cysts lined with flat epithelium did not have a basement membrane, while
all the other cysts had. Besides these structures small pieces of cartilage
and sections of glands were found.

VIII.—Lazarus' case.* A tumor weighing 9000 grammes was removed
from the right ovary of a woman, 27 years of age. It was decidedly
lobulated; the centre of each lobe was formed of cartilage, and around
the cartilage the tissue was honeycombed with small cysts.

The following tissues were found on microscopical examination: Hyal-
line cartilage merging into fibro-cartilage and mucoid tissue; connective
tissue, some of which was sarcomatous; adipose tissue; myxomatous tissue
with branched cells; elastic tissue; flat epithelium with cholesteatoma
pearls; cysts with distinct flat epithelium; cysts formed from dilated
glands; and cysts lined with skin provided with sweat glands, hair folli-
cles, but without hair or sebaceous glands. In addition to these struc-
tures there were smooth and striated muscle fibres; glands with simple
cylindrical epithelium; typical goblet cells like those of the intestine;
cysts with ciliated epithelium; neuroglia; varicose medullated nerve
fibres; ganglion cells like those of the brain and sympathetic system; and
finally eye-like structures. These last lay in the neighborhood of tissue
resembling gray brain-matter.

No hair nor sebaceous glands nor bones nor teeth were found. The
author, however, believed he discovered indications of foetal tooth for-
mation.

The tumor grew with immense rapidity, and other tumors were found
in Douglas' sac and on the diaphragm which were thought to be im-
plantation growths. The growths in Douglas' sac contained cartilage,
and cysts lined with goblet and cylindrical epithelium; those on the
diaphragm were made up entirely of mucous tissue.

IX.—Biermann's case.† This was a spindle-celled sarcoma of the
ovary, the size of a man's head, and contained a dermoid cyst the size of
an apple.

by Wilms in Ziegler's Beiträge, xix, 369.
† Prag. med. Wochenschr., 1885, No. 21.
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X.—Ewald's case.* From a girl, 21 years of age, a teratoma of the ovary the size of a man's head was removed by laparotomy. Microscopical examination did not reveal any carcinomatous or sarcomatous tissue. Three weeks later there was a recurrence as large as the first tumor; this was removed, and at the same time the opposite ovary was excised, as it was also the seat of teratoid disease. A further recurrence was demonstrable ten days after the second operation, and two weeks later the recurrent tumor had attained to the size of a child's head. After several weeks a third operation was done, but the patient died. The autopsy showed that the tumor in the last instance had arisen, not from the region of the ovary, but from the omentum; the peritoneum was studded over with nodules the size of a pea. In addition, behind the peritoneum and in the mesocolon there were two tumors larger than pigeon's eggs. Neither sarcoma nor carcinoma could be demonstrated anywhere. The nodules in the peritoneum consisted exclusively of embryonic connective-tissue cells. Ewald draws the conclusion that teratoma of the ovary is in itself a malignant tumor, and that metastases can occur without carcinomatous or sarcomatous degeneration and without alteration of the histological structure.

I have not included in the foregoing list cases without recurrence or metastasis where it is stated simply that part of the stroma of the teratoma presented a sarcomatous character or nests of epithelium.

There are still some instances where teratoid tumors have undergone malignant degeneration, but they differ so markedly from the foregoing that they require separate mention. A few cases have been observed where flat-celled epithelioma has sprung from the skin lining the internal surface of what have always until lately been looked upon as simple dermoid cysts of the ovary. It would seem, however, that these presumably simple dermoid cysts are in reality complex tumors, for Wilms found in every case examined by him tissues representing all three embryonic layers. If all the ovarian dermoid cysts were compound tumors, one can justly assume with him that this is always their nature. But if all ovarian dermoid cysts are compound tumors, then all instances of malignant degeneration in such cysts would be

instances of malignant degeneration in compound tumors. Although Wilms has found these dermoid cysts to be uniformly so complex, yet he does not believe that these ordinary dermoid cysts of the ovary should be classified with teratomata occurring in other parts of the body, and would consider them entirely apart and by themselves.

Epitheliomatous malignant degeneration has been reported as occurring in ovarian dermoid cysts by Heschl, Biermann, von Wahl, Pilliet, Tauffer, Himmelfarb, Krukenberg, Veit, Wilms, Klein, Yamagiva, and Thumim, thirteen cases in all.

In speaking of carcinomatous degeneration of ovarian dermoids, Thumim says that in looking over the literature he considers that the cases of Heschl, v. Wahl, Biermann, Himmelfarb, Krukenberg, Tauffer, and the second case reported by Yamagiva are the only undoubted examples of this affection which he has been able to find. Nine other cases that have been reported he cannot accept, partly on account of uncertainty that the disease was primary in the ovary, and partly on account of insufficient microscopical investigation.

In considering whether the adeno-carcinoma-like structures described in the case here reported were really cancerous or not, one must bear in mind that what appears to be identity of structure does not necessarily indicate identity of nature, and that structurally a resemblance to cancer may be simulated when one does not really exist. It does seem, however, that the adenomatous tissue here was malignant and therefore constituted an adeno-carcinoma. The epithelium was thrown about in a most disorderly fashion in alveoli having no basement membrane. Furthermore, these structures were heterolo-
gous, for, normally, glands of this or any other character do not occur scattered at random throughout a living being, and heterology is an important element in the estimation of malignancy. The whole tumor was structurally embryonic in character, and looked as if rapidly growing, and clinically it was a rapid grower and recurrent; and embryonic structure, rapid growth and recurrence are of more weight even than heterology in arriving at a diagnosis of malignancy, but here all these combined in pointing to a malignant nature. Nevertheless it can be objected that metastasis was not proven, that infiltration of neighboring tissue was not a marked feature, and that the embryonic character and rapid growth of this glandular epithelium might simply have been co-equal with the embryonic character and rapid growth of the rest of this parasitic individual.

If, however, it be granted that the adeno-carcinoma-like structures in this case were really adeno-carcinomatous, then it is a very rare event indeed, so rare that Tauffer says no case of adeno-carcinomatous degeneration in a dermoid has ever yet been described.* Some of the structures in Jores' case, mentioned above, seem, however, to have struck the author as resembling adeno-carcinoma, for he describes some of the cysts as being irregular in shape and lined with cylindrical epithelium which did not rest on a basement membrane, but stood directly on the spindle-celled tissue of the stroma of the tumor. Metastatic nodules in the right lung were said to be exactly like the main tumor, and therefore, presumably, had also the above mentioned irregular cysts lined with columnar epithelium. In Emanuel's and in Lazarus' cases, already quoted, there were cysts in the metastases lined in the former with stratified epithelium and in the latter with goblet cells and cylindrical epithelium.

Some might assert their disbelief in the tissue removed at the second operation being a recurrent growth, principally because of the immense variety of structures found in it. They might say, that besides the three obvious lobes removed at the first operation there were probably other original implantation tumors scattered over the peritoneal surface that had not yet attained such a size as to attract attention, and

that the tissue removed at the second operation was the tissue of original implantation tumors that had grown into prominence in the intervening time.

Against this opinion is to be urged what Dr. McNutt told me: that at the first operation the peritoneal cavity was, as far as he could see, perfectly healthy and free from tumor growth, excepting the area from which the original tumor grew; also that the large mass of the tissue removed at the second operation was taken from the site of the original tumor; that at the post-mortem the spread of the tumor had taken place as a continuous layer over the peritoneum; and that there were no indications at any point that this continuous layer of new tissue had been formed by the coalescence of smaller tumors.

The term "recurrent tumor" has been continually made use of throughout this article to designate the tissue removed at the second operation. By "recurrent," however, is meant that some particles of the original tumor, too small to be observed by the operator, were left behind and had grown rapidly so as to form the large tumor met with at the second operation. The probability that some of the original tumor was left unremoved at the first operation was shown by the discovery of the small piece of bone in the tissue extirpated at the second operation, for the time between these two operations seemed too short for fully developed bone to form.

A very small portion of the original tumor left behind and growing again would be sufficient to account for the great variety of tissues found in the recurrent tumor. To illustrate this more fully it is only necessary to relate that, at the suggestion of Dr. W. G. Hay, transverse sections were made of a bundle of the strings connecting the lobes of the original tumor. The diameter of none of the strings was greater than that of ordinary twine, but it was surprising to see the variety of structures, both epithelial and connective, encountered in each of them.

The incomplete removal of diseased tissue and the rapid reappearance of a tumor at the point of operation is, it is needless to say, a frequent and often unavoidable occurrence, and constitutes an important factor in the group of events to which the term malignant is
given. Malignancy in the present instance consisted of rapid growth, recurrence and the property of spreading by continuity. The tumor did not appear to have given rise to metastases, although from its structure this kind of malignancy might have been expected, and has been observed several times in similar teratomata.

Some of the different theories that have been framed in the past to account for the origin of teratomata may now be considered, and an opinion arrived at as to which one of them best fits the conditions found in the present instance.

I.—Many dermoids and teratomata are accounted for by a dipping down or involution of the external skin and subsequent pinching off. In such cases a cyst is formed with the epithelium of the involuted skin lining its inner surface. This theory does not, however, explain the present case, which was a solid teratoma covered by skin.

II.—Many teratomata admit of the explanation that they arise from a simple misplacement of a group of cells during development. The free situation in the abdominal cavity, the integumentary covering, and the great variety of structures in the case in hand would, however, all be against this view of its origin.

III.—Some authors have thought that irritation of a tissue may cause a compound tumor consisting of many varieties of the tissue irritated. This theory may be dismissed without comment.

IV.—What is called partial double formation sometimes takes place, that is to say, the same generative act that is responsible for the main being or individual also gives rise to a double formation of some of its parts. This theory satisfactorily explains the formation of supernumerary limbs, but hardly accounts for a tumor in which such diverse organs as skin, gut, nervous system and eye were found.

V.—At times evidently a complete double or twin formation takes place, with partial growing together or fusion of the twins. Many of the double monsters would appear to originate in this way, but the case in question is not to be so explained, for although there were two beings, no fusion had taken place between them. One individual simply grew inside the other.

VI.—It has been presumed that the unfecundated ovum can proceed
to develop without the assistance of the spermatozoon, and form an
imperfect individual. The spermatozoon also without the concurrence
of the egg is thought by some to have some formative power. This so-called parthenogenesis is supported by numerous examples in
the lower orders of life, where the fecundation of the female is only
required every few generations.

This possibility requires to be carefully weighed in the present case,
as the egg is the only cell that we know of that can give rise to an
individual having all three embryonal layers. And the converse is
also true, that an individual having all three embryonal layers must
arise from an egg. Therefore the individual under consideration
necessarily originated from an egg. If this hypothetical egg came
from the ovary of the child in whom the tumor was found, then this
would be a case of parthenogenesis. This child, however, had not
menstruated, and therefore presumably had not ovulated. It must be
admitted, however, that it is thought that an egg may go on to devel-
opment in the ovary before it has ovulated. But if this had been the
case in the present instance the dermoid would have been found in the
ovary, or would at least have been attached to it by a pedicle. As
before stated the ovary was normal, and there were no traces at all of
a pedicle or of a union between the tumor and the ovary.

VII.—It is held that two individuals may begin to develop from
their respective ova, but before development is far advanced one may
be enclosed by the other, and the included one's growth be so inter-
fered with that it does not continue to develop but lies dormant for an
indefinite time. This is called foetus in foetu, and is the most natural
explanation of the facts in the present instance. It is easy of com-
prehension to suppose that during this little patient's foetal life, and
before the abdominal cavity closed, another egg, and possibly a fecun-
dated one, slipped into the open peritoneal sac. The egg lay there
as in a pocket up to the child's twelfth year, when it began to grow,
but in a disorderly fashion and with all the energy of foetal life. In
this view the tumor would represent the patient's twin brother or
sister.