Reprint from

SURGERY, GYNECOLOGY AND OBSTETRICS

November, 1906, pages 661-671

28A

SACROCOCCYGEAL TUMOR (TERATOMA)[1]

With an Opening Entirely through the Sacrum, and a Sinus Passing through This Opening and Communicating with the Rectum, the Sinus Resembling a Bronchus

By W. W. KEEN, M. D., LL. D., Philadelphia
Professor of Surgery, Jefferson Medical College

and

W. M. LATE COPLIN, M. D., Philadelphia
Professor of Pathology, Jefferson Medical College

SURGICAL REPORT

By DR. KEEN

HELEN W., aged 2 years, was first seen on October 1, 1902. When born, a moderate sized tumor was noticed in the lumbosacral region. The tumor has grown considerably, so that now its dimensions are 21 x 12 cm., and 5 cm. in thickness. (Fig. 1.) There is a slightly developed hypertrichosis over a considerable area, suggesting a spina bifida occulta. Just below its center there is a small fistulous opening in the skin, but no inflammatory redness around the opening. The mother is quite sure that on two or three occasions, when she gave the child an enema, the fluid escaped from the rectum through the small opening. In addition to that, she thinks she has observed more than once a fæcal odor. There is a very small amount of constant mucous discharge. There has never been any attack of illness with discharge of pus from the rectum which could be interpreted as the bursting of an abscess posterior to the rectum by which the opening in the rectum could have been established after birth. It is therefore reasonable to conclude that the complete sinus was congenital. No other deformity exists.

October 6, 1902. In order to make a careful examination, I etherized the child to-day. Posteriorly, in spite of the difficulty caused by the large tumor, I was able to feel, though somewhat obscurely, the edges of an opening in the sacrum just above the fistulous opening. On inserting a finger into the rectum, I readily found an opening in the sacrum anteriorly, an opening which would admit the end of my forefinger. A probe, which could be introduced 6 cm. into the fistula, but no farther, could be felt through the posterior wall of the rectum, but at no point was I able to detect any communication with the gut by this stiff metal probe, though I suspected its existence. I determined, therefore, to use a liquid flexible probe, i. e., an enema of salt solution. Almost instantly the fluid began to flow through the fistulous opening. It was clear, therefore, that there was a perforation completely through the sacrum, and that the fistula passed directly through this perforation in the bone from the rectum to the skin.

As the tumor was growing, I recommended immediate operation, with the view of closing the fistulous opening as well as removing the tumor.

OPERATION

November 4, 1902. I decided to isolate the fistulous tract (identifying it by means of a

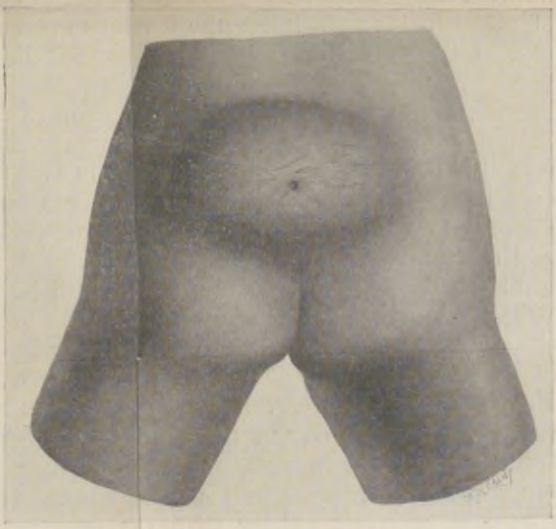

Fig. 1. External appearance of the tumor. Notice the opening of the fistula and the moderate hypertrichosis

[1] Read before the College of Physicians of Philadelphia, October 3, 1906.

28a

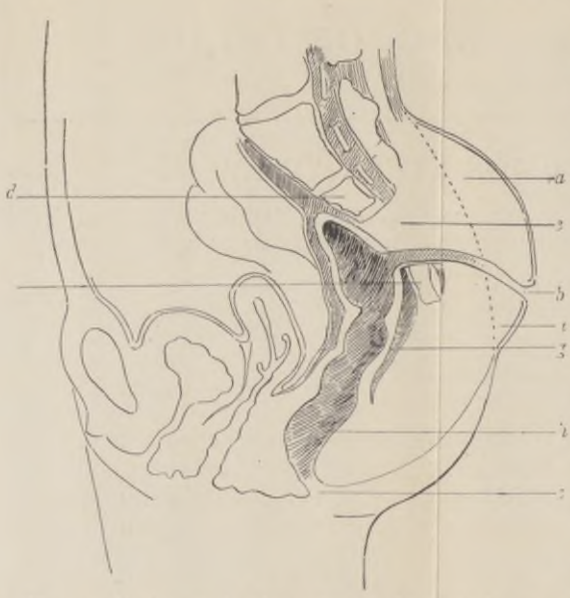

Fig. 2. Diagrammatic scheme intended to show the supposed relation of the tumor, sinus, spine, and the rectum. *aa*. Tumor. *b*. Mouth of sinus, communicating with the rectum. *c*. Anus. *d*. Second sacral vertebra. *e*. Defect in sacrum. *f*. End of sacrum. *g*. Retrorectal connective tissue. *h*. Rectum.

probe passed into it and kept in place) so that it would be like an appendix, and then after removing the tumor, to attempt to invert the tubular sinus as one would the stump of an appendix. Accordingly, I made a transverse elliptical incision, inclosing the fistulous opening. I then dissected back the flaps of skin to the periphery of the tumor. Beginning at the margin, I separated the tumor from the tissues anterior to it by blunt dissection, and was able to recognize readily the normal tissue, even the normal fat, as distinguished from the abnormal fatty tissue of the tumor, and to dissect the tumor from the deep fascia all around, until the fistula was left as a sort of pedicle. When I had dissected the tumor nearly to the fistula, I readily discovered the opening in the posterior wall of the sacral vertebræ. Having separated everything down to the fistula, I then dissected this out, leaving it protruding about 2.5 cm. posterior to the sacrum. On cutting it across I found it to be a tube of considerably larger caliber than the opening in the skin. The external opening would barely allow an average sized pocket-probe to enter. The caliber of the tube internally was about 4 to 5 mm., and clearly lined with mucous membrane. On account of the rigidity of the wall of the sinus, however, I succeeded very imperfectly in inverting it, but after closing it with sutures as well as I could, I tied two ligatures of silk around it, in the hope that the resulting raw surfaces would adhere sufficiently to close the tube.

Next I examined the spina bifida. I found its upper posterior margin was at the second sacral vertebra. It measured vertically about 3 cm. and in width about 1.5 cm. No relation of the sinus to the spinal cord or to the subdural space was evident. On the contrary, instead of being lined with endothelium, it was lined with an epithelial mucous membrane. Fig. 2 represents diagrammatically the anatomy of the parts in section. A small bit of iodoform-gauze was packed over the site of the stump of the sinus. Only three or four ligatures were required, chiefly to a few large veins which emerged through the opening of the spina bifida. What relation the sinus held to the spinal cord I could not determine, saving that it did not communicate with the subdural space. The wound was now closed with interrupted sutures and dressed with suitable pressure-pads to eliminate any dead space.

After the operation, her temperature rose to 102.8°. It gradually fell to the normal by the sixth day, but from that time on for two weeks varied between 99° and somewhat over 100°. The margins of the flaps sloughed for nearly a centimeter in the center. This was followed, naturally, by a slight local infection. Undoubtedly her persistent slight rise of temperature was due to that. The wound healed slowly, and was not entirely well till January, 1903. The sinus at first gave no trouble, and was apparently completely obliterated. It soon, however, reopened and persisted. The specimen was given to Dr. Aller G. Ellis, whose report is appended.

PATHOLOGICAL REPORT ON THE FIRST SPECIMEN

By ALLER G. ELLIS, M.D.
Associate in Pathology, Jefferson Medical College, Philadelphia

SPECIMEN. Fatty tumor removed from sacral region.

The specimen, received in ten-per-cent formalin solution, is an ovoidal mass of tissue 15 cm. in length and possessing a maximum width of from 6 to 7 cm. It

is 3.5 cm. thick and weighs 147 grams. The specimen presents for description two surfaces and a margin.

The first surface (Fig. 3) is convex in shape, and is made up wholly of fat, except at one point. (Fig. 3, *A*.) At this point, which is about equally distant from the ends of the specimen and near one margin, is situated a narrow strip of skin 4.5 cm. in length and having a maximum width at the middle of 0.8 cm. It tapers to a point at each end. Two centimeters from one end, and very near one edge of this strip of skin, is a depressed area surrounding an opening about 0.3 cm. in diameter. The margin of this opening is formed by infolded, wrinkled skin which is brownish in color. Projecting from the margin are a few short, bristly hairs. A probe passed into this opening traverses a sinus about 2.5 cm. in length and emerges on the second surface at a point shown in Fig. 4, a short distance from a cyst to be described later.

The second surface is concave in general appearance, this being especially true of one half, the depression in the other being partially filled by a projecting mass of tissue, the most prominent portion of which is on a level with the edges that limit the concavity. This surface is also formed mostly of fat, but shows here and there small areas of muscle. At a distance of 7 cm. from one end, practically the midpoint of the specimen, and 2.5 cm. from the margin nearest the piece of skin on the first surface, is situated what is apparently a small, thick-walled cyst (Fig. 4, *B*) that has been divided into two unequal parts by an incision that extends almost through it. The incised surfaces of this cyst are approximately circular in outline, the larger being 1.2 cm. in diameter. The surfaces are grayish in color, have a glistening appearance, and are moderately cupped, showing that the concavity of the cyst had been small. On removing the smaller part for microscopic study, the knife encounters what is apparently cartilage, and investigation shows that the greater portion of the wall, which is quite thick, consists of that tissue, the soft lining being very thin. Underneath the cyst, and extending for some distance into the projecting mass described, is an area that is shown by puncture to have this same cartilaginous consistence. Toward

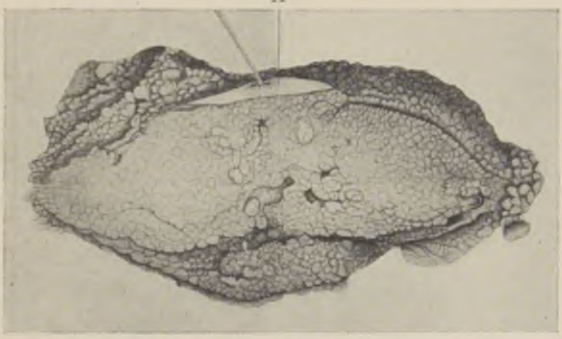

Fig. 3. *A*. Skin on posterior or external surface of specimen after removal. Just below the leader from the letter *A* is a probe engaged in the sinus, the direction of which is thereby shown. One third size. From Report 2002.

Fig. 4. Anterior or internal surface. *B*. Small thick-walled cyst. The internal opening of the sinus contains the tip of a probe inserted from the opposite end of the sinus shown in Fig. 3. One third size. From Report 2002.

the opposite extremity of the projecting mass is an irregular, trough-like depression about three centimeters in length, one side of which shows a number of narrow, transverse folds or rugæ. The tissues of this area have the same general appearance as the other superficial portions of this surface. Less than one centimeter from the cyst is the opening of the sinus previously referred to. This opening is surrounded by an irregular mass of muscle and fat which practically obscures it when the probe is withdrawn.

A piece of tissue 1.5 cm. long and wide, removed from the specimen in such a way as to include a part of the sinus near its middle portion, shows it to be bounded by a clearly defined, rather dense wall slightly more than 1 mm. in thickness.

The margin of the specimen is fairly regular in outline, is formed by the junction of the two surfaces described, and presents nothing worthy of note.

The piece of tissue including part of the sinus, to be known as *A*, and the removed portion of the cyst *B*, were hardened in alcohol, infiltrated with paraffin, and sectioned. Sections from each were stained by the usual laboratory methods, and on microscopical examination they show the following histological structure:

A. The wall of the sinus is composed of smooth muscle, fat, and fibrous tissue, the latter being arranged, for the most part, as a band surrounding the sinus. The lining of the sinus is of two distinct types:

1. The greater portion is formed of many layers of epithelial cells, the superficial being thin and of the squamous variety, the deeper layers being more polyhedral in shape. In some parts the squamous layers are practically all that are present. In others, a few of the deeper layers are of the polyhedral type, while at a few points the polygonal cells are very numerous, dipping down, and giving the structure a resemblance to normal skin. At these points, however, a rather indistinct structure resembling a basement membrane is present. The deeper cells present no special characters distinguishing them from those more superficially placed. Many of the superficial layers possess the usual microchemical reactions of keratohyalin.

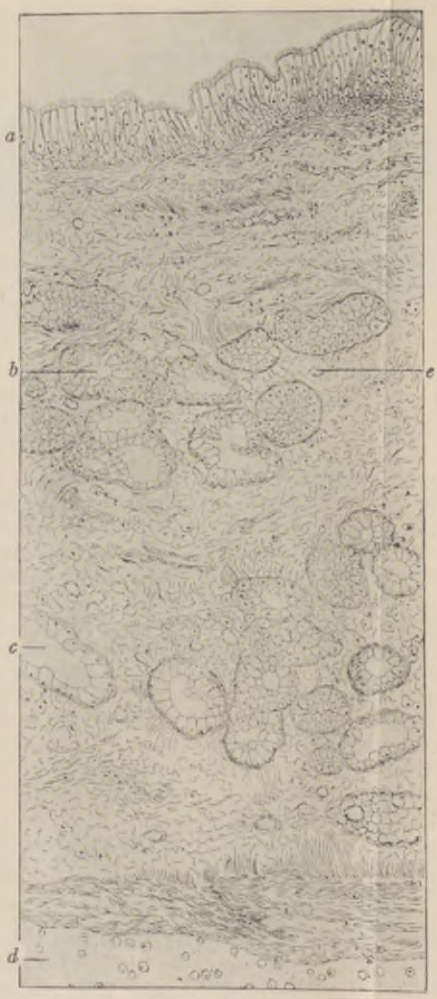

Fig. 5. Section of cyst-wall. Showing trachea-like structure in tumor. *a*. Ciliated epithelium. *b*. Gland. *c*. Gland. *d*. Cartilage. *e*. Vessel.

2. At points this stratified squamous epithelial layer changes abruptly, and without apparent change in the underlying tissue, to a layer of tall columnar ciliated cells placed on a fairly well-marked basement membrane. In some of the sections this columnar cell-lining occupies one third or more of the circumference of the sinus; in others, but a small part. Small areas show some desquamation of the columnar cells, especially near the junction of those with the stratified squamous variety. At points these epithelial cells dip down, forming either tubules or the openings of glands. (Fig. 3, *A*.)

Immediately beneath the epithelial lining of the sinus is a zone, varying in width, of loose areolar tissue, very rich in cells, of the mononuclear or lymphoid type, and of various sizes. At a few points, collections of these cells extend for some distance into the underlying tissue. The densest accumulations of them are situated beneath the columnar cell areas described, and at two or three points extend through the latter to the free surface, the columnar cells being absent, or displaced or fragmented, as though the mononuclear cells had infiltrated that area.

At one point (Fig. 2, *a*) deeper in the wall of the sinus can be identified two areas of cells that have no apparent connection with those just described. The one area is irregularly circular in outline, and is made up of cells and matrix closely resembling the zone immediately beneath the epithelial lining of the sinus. Several points in this area appear to be cystic, as there are distinctly outlined cavities bordered by the loose areolar matrix, in which the cells of this area are placed. The second area of cells, ovoidal in outline, is closely approximated to and continuous at one point with the first area. The periphery of this area is formed of cells apparently identical with those in the first area, but in the central portion the nuclei are much flattened and elongated, assuming in some instances a spindle shape. Slight, if any, cystic change is present at this point.

The outlying portions of the sections, including the sinus, are made up mainly of muscle and fat. Some of the muscle bundles apparently have been the seat of a degenerative process of some sort, as they present a homogeneous, colloid appearance and manifest unusual tinctorial reactions. Blood-vessels are fairly numerous throughout the wall.

Cross-sections of branched tubular mucous glands are also seen. (Fig. 3, *A*.) As these are identical with glandular elements in the cyst-wall, they will be described later.

B. Examination of sections from this portion of the specimen passing through the entire cyst-wall at once shows it to have a histological structure which resembles in the most striking manner that normally found in the wall of a bronchus. (See Fig. 5.) There are present in succession, from without inward, muscle and areolar tissue, a broad band of apparently normal hyaline cartilage surrounded by its perichondrium, submucosa containing transverse and longitudinal sections of mucous glands, and a distinct basement membrane surmounted in areas by tall, columnar, ciliated epithelial cells. These cells show varying degrees of desquamation, being entirely absent from the greater extent of the surface. At points in some of the sections showing no epithelium, there are on the surface projections or fragments of muscle and loose areolar tissue containing blood-vessels. The submucosa shows some cellular infiltration, and at areas the epithelial cells lining the tubules of the mucous glands are desquamating and exhibit fragmentation.

The glands referred to as present in the submucosa of parts of the sinus and in the cyst-wall are histologically fairly uniform. Sections so fortunate as to show clearly duct openings on the free surface have not been obtained, but dipping-points with changes in the superficial epithelium, such as are usually noted in the mouths of ducts, have been encountered frequently. In the submucosa the glands show lobulation with a varying degree of clearness. In suitable sections such lobules are composed of twenty to thirty or forty units,

each unit being a transverse section of a tube and composed of a membrana propria, upon the inner surface of which is placed the usual glandular epithelium, and beneath which is the interstitial connective tissue carrying the blood-vessels. The epithelium in different areas, or even in different parts of the same tubule, sometimes shows the characters of fully distended secreting cells, while at other points these bodies are empty. In many of the lobules branching tubules can be seen. The intralobular ducts are of the type usually seen in mucous glands.

made because there is some doubt as to this portion of the specimen being distinct from the sinus. Although there is no direct connection between the two in the specimen as received, we are inclined to the view that there was such connection originally. This view is supported, to some extent, by the proximity of the two structures, and more strongly by the presence in the sections containing the sinus of tubular

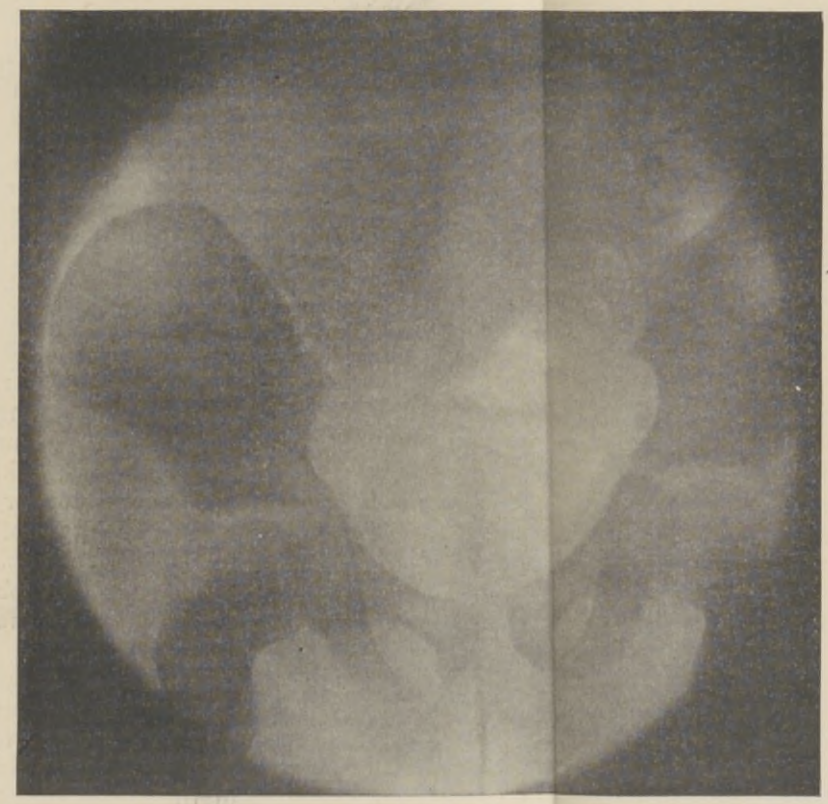

Fig. 6. Skiagraph of the pelvis. Showing in the middle line, on a level with *A*, the oval opening in the sacrum, and the apparent absence of the lower sacrum and the coccyx.

DIAGNOSIS

The histological findings in this specimen indicate that it is a variety of the congenital sacrococcygeal malformations that include dimples, sinuses, and cysts. The sinus in this instance is lined partly by elements resembling skin and partly by a structure resembling mucous membrane, the formative attempts having proved more or less abortive in each case. The cyst, if such it is, has an almost perfect bronchial structure. This qualifying statement is

glands having the same structure as those found in the wall of the so-called cyst.

The point raised is not of great importance, except as determining whether the condition present is that of a sinus and a cyst, or is simply that of a sinus, the so-called cyst being its terminal point. In either case, the teratomatous tendency of these formations is evidenced in this specimen by the different structures lining the sinus and by the presence of a wall presenting the histologic characters of a bronchus.

October 26, 1903. Her general health since the operation, nearly a year ago, has been very good, and she has grown rapidly, but the small sinus which reopened still persists at the site of the original opening. As far as the probe can detect, it is about 3 cm. deep. I advised another operation, believing that most probably, as there had never been any escape of gas or any fæcal odor, but only a few drops of pus each day, it was probably now merely a blind sinus terminating in a cul-de-sac.

SECOND OPERATION

October 29, 1903. Before beginning the operation, in order to determine whether the sinus opened into the rectum, as before, I repeated the maneuver preceding the first operation; i. e., I injected into the rectum a considerable amount of salt solution. Before the first operation, this immediately welled out through the sinus; to-day not a drop appeared, though I injected a much larger quantity than at the first operation.

A probe was first passed as far as possible into the sinus. An elliptical incision was then made transversely above and below the probe, and the tissue all around the sinus dissected out. I finally divided the foot-stalk or pedicle by which it seemed to be connected to the deeper tissues; and after the operation, on splitting open the sinus, I was pleased to find that I had passed about 5 mm. below the apparent end of the cul-de-sac. A finger could be inserted from the back into the opening in the sacrum and felt by a finger in the rectum. No sinus could be detected by the probe or otherwise. The wound healed without any reaction in a few days, and has never reopened.

PATHOLOGICAL REPORT ON THE SECOND SPECIMEN

By DR. A. G. ELLIS

Specimen. Tract of sinus removed from sacral region.

This specimen consists of a small piece of tissue 3 x 3 cm. and 0.8 cm. thick. The surfaces are rough and grayish in color, except a narrow groove near the center, which partially divides the specimen into two nearly equal portions. This area is slightly pinkish in color and a trifle less dense than the surrounding tissue.

The specimen was fixed in ten-per-cent formalin, dehydrated, cleared and infiltrated with paraffin. Sections were cut, and, after staining by the usual laboratory methods, show the following:

The findings, in general, are those described in detail in the report upon the specimen removed from the same patient at the first operation (*vide ante*), though the arrangement of the tissues does not so nearly correspond to that of certain typical structures, bronchi for instance, as in the previous specimen. A small part of the border of some of the sections shows stratified epithelium similar to that of skin, hair follicles, and sebaceous glands, both of which are very rudimentary in type. Other portions of the sections contain small areas of cartilage, in proximity to which are areas of tubules lined by epithelial cells, the latter areas conforming to those of mucous glands. Near these are deep infoldings or tubules lined by columnar epithelial cells bearing distinct cilia. These structures, instead of being placed in proper order to form the histological structure of a bronchus, are scattered indiscriminately in the fibrous tissue which forms the larger part of the sections, none of them, however, being at any considerable distance from the others. Certain parts of the sections contain striated muscle which shows chronic productive interstitial change and atrophy of the fibers.

Diagnosis: Sacral teratoma.

REMARKS

This case is evidently a very rare form of teratoma. The notes and drawings have been submitted to Drs. George S. Huntington and Francis H. Markoe of New York, George A. Piersol of Philadelphia, Professor Martina of Gratz, and Professor Ballantyne of Edinburgh, all of whom concur in this opinion. The skiagraphs were more recently obtained, and have not been submitted to the gentlemen named.

So far as I have been able to search, I have not found any case which is precisely similar to it. It seems to be unique by reason of the sinus, which, beginning at the skin, perforated the sacrum and the spinal canal and terminated in the caliber of the rectum, the sinus itself resembling an imperfect trachea of bronchus in structure.

At the time when the child first came under my care, for certain reasons unnecessary to state I was not able to obtain a skiagraph. Even had I been able to do so, it is doubtful whether it would have been of much assistance, because at two years of age the ossification of the bony structures is so much less advanced than at present. Recently, at six years of age, I have obtained two skiagraphs. Compare these also with Fig. 2. One of the pelvis alone (Fig. 6) shows very well (and, of

course, much better in the original skiagraph than in any reproduction), in the middle line, at the level of the letter *A*, the existence of an oval opening in the sacrum. In the original skiagraph this opening shows quite distinctly

When I examined the child's pelvis by the finger in the rectum at the time of the first operation, I did not suspect the absence of bony development in the lower part of the sacrum and coccyx, nor would it have been suggested

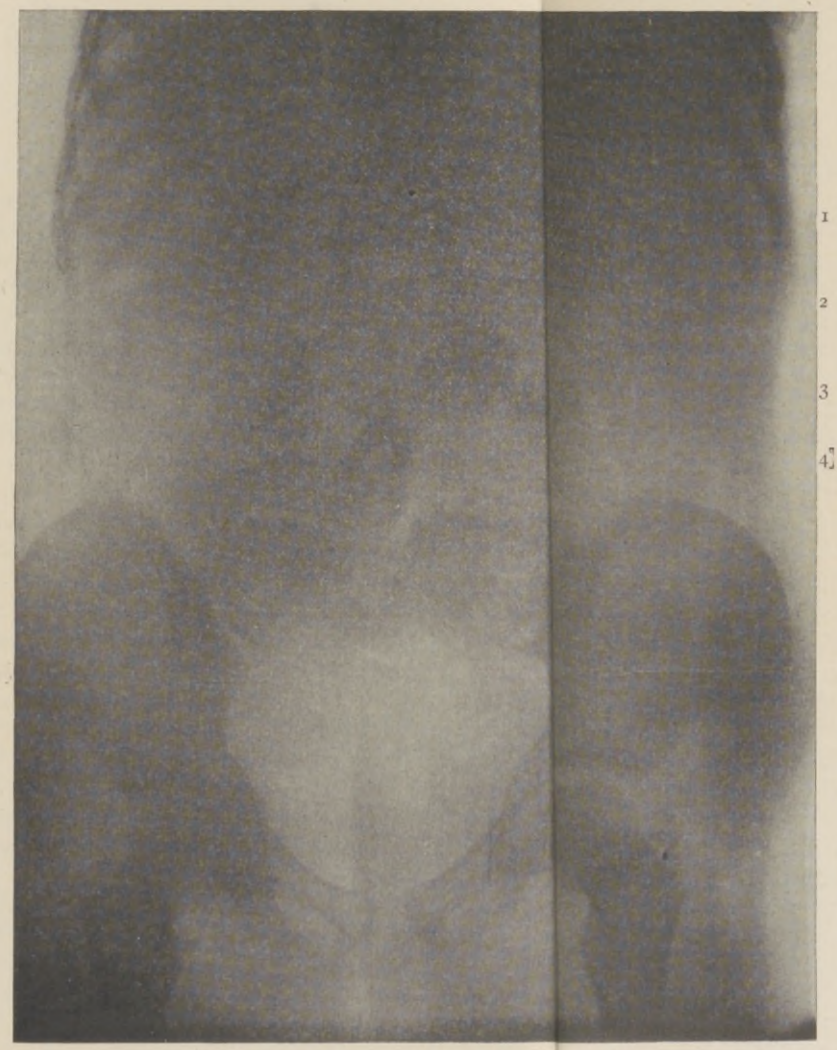

Fig. 7. Skiagraph. Showing the attempted bifurcation of the lumbar spine, and a possible rudimentary arm, on a level with 4, as well as the points noted in Fig. 1. 1, 2, and 3 are placed at the level of the first, second, and third lumbar vertebræ.

an upper and a lower portion, with a slight constriction in the middle, so that it resembles the figure 8, with the cross line in the middle partially obliterated. There is apparently, also, no bony sacrum below the second or third sacral vertebra, and no bony coccyx.

recently, when I re-examined by rectal touch, except for the finding of the skiagraph. Evidently these bones either still consist of quite firm cartilage or else of fibrous tissue so firm as not to have led me to suspect that it was not bone. The fact that the opening is so long

vertically was not discovered, because only the lower part of the opening could be reached by my forefinger. This was engaged by the lower half of the 8, and as I could not reach the upper half of the 8, I did not know that it existed. Even at the recent examination, after I knew the shape and size of this opening, I could not reach its upper portion.

The second skiagraph (Fig. 7) shows this same fibrous tissue, but reveals a second fact of great importance; namely, that the body of the first lumbar vertebra (on a level with 1) is broader than normal; that of the body of the second (2) is still broader; the body of the third lumbar (3) is nearly twice the breadth of a normal vertebra, and shows a marked tendency toward separation into a right and a left half. The bodies of the fourth and fifth lumbar form a confused mass which is very broad, and about in the center is a light space which apparently marks their irregular but complete division into a right and a left half. Just below this is the oval opening already described in the sacrum. Just below the level of the body of the third lumbar vertebra, and on a level with 4, is a dark shadow, which suggests the possibility of an abortive humerus, forearm, and possibly a hand.

In the light of these skiagraphs, examination of the back showed also this great breadth of the lumbar spine (13 cm. at the level of the iliac crests), so that, as will be observed (Fig. 7), the lumbar spine forms a pyramid, the base of which is apparently at the junction with the iliac bones.

The attempted duplication in the spine and the imperfection of the lower sacrum and coccyx may or may not be indicative of any imperfection or duplication in the pelvic organs. The rectum, we know from repeated examinations, is normal, excepting the fistulous opening, which passed from the rectum through the sacrum, the spinal canal, and the tumor to open just below the center of the tumor. Whether the vagina, and possibly also the uterus, either or both, are double is uncertain. In so young a child, of course, I would not be justified in making any vaginal examination.

I have explained the whole matter to the parents, and advised that when she is 15 or 16 years of age, and before the question of possible marriage would arise, she should be reexamined, and if necessary for a thorough examination, she should be etherized. The condition of the vagina and the uterus should be ascertained and the pelvis carefully measured, so that if there is any bar to marriage this should be known and suitable measures taken. Whether the pelvis will develop normally or abnormally, and so influence parturition favorably or unfavorably, would be of the utmost importance to her future welfare.

Dr. Coplin will discuss the case more fully from the pathological and embryological standpoint. Certainly it is a bigerminal teratoma, the duplicated spine representing the imperfect fusion of the two normal centers of ossification of the vertebral bodies, while the tumor, with its curious development of the sinus representing an imperfectly developed trachea or bronchus, and possibly, as shown in Fig. 2, a rudimentary arm may represent a second ovum.

From the surgical standpoint, there are only two points to be considered: 1. Before operation I was in great doubt whether, in order to obliterate the canal leading from the skin into the lumen of the rectum, it might not be necessary to make a considerable opening in the sacrum, and thus gain direct access to the posterior wall of the rectum for the purpose of inverting the opening in this wall. This would have involved great danger of opening the subdural space, with possible injury to the nerves of the cauda equina. 2. A second method of attack I had considered was the perineal route between the sacrum and rectum, practically a Kraske operation.

Fortunately, I did not have to attempt either of these routes, inasmuch as I found, during the operation, that I could dissect away the entire tumor until it was attached only by the tubular sinus as a pedicle or foot-stalk, and I was able to obliterate the rectal portion of the sinus at the first operation; while the persistent distal portion was removed and obliterated at the second operation. Had I failed, the two possible modes of attack above mentioned could have been attempted.

It is now almost three years since the last operation. The child is well developed and healthy. When she walks there is a little

waddling gait, due, I presume, to the unusual breadth of the spine and pelvis, or possibly an instability in the lumbar spine. There has never been the slightest trouble from any fæcal accumulation in what must still exist, — the diverticulum from the lumen of the rectum to the point of obliteration of the tubular sinus at the posterior surface of the sacrum. It is worthy of note, also, that in spite of the large oval opening in the anterior portion of the sacrum, there has never been any tendency to the formation of that rare anomaly, an anterior sacral meningocele.[1]

SACROCOCCYGEAL NEOPLASMS A SUMMARY

By DR. COPLIN

The sacrococcygeal region shares with other parts of the body in the distribution of new growths. To many neoplasms the area is but slightly liable, and, as in several anatomical fields, is subject to morbid growths in many respects somewhat unlike those occurring elsewhere. In the sacrococcygeal field the common tumors of wide distribution are rare, and the rare tumors of other parts are here more frequent. Of the histoid growths, fibromata, chondromata, osteomata, and lipomata are infrequent; carcinoma and sarcoma, rare; papillomata of various types are probably the most common of the simpler neoplasms involving this region; angeiomata and endotheliomata have been observed. It is not unlikely that the gliomata described have been types of teratoma in which a single element overshadowed other structures. Sebaceous cysts and hygromata, the latter probably of bursal origin, deserve little more than mention.

The important neoplasms of special interest to the practitioner because of their practical importance, and to the oncologist by reason of their histogenesis, are now conceded to be derived from developmental errors frequently arising early in the evolution of the embryo. Perfect evolution of the tissues embraced in this part of the body is beset by many narrow escapes, and the sharply drawn lines which nature must follow render many and varied deviations so easily possible that it is a wonder that disaster is not of more frequent occurrence. It is in this area that the caudad termination of the primitive streak must most accurately attain its evolution and involution, the neurenteric canal develop and disappear, the anus complete the intestinal tube, the posterior fissure properly close, coccyx and sacrum develop, the inferior extremities symmetrically adapt themselves to the trunk, possibly the caudal appendages of our ancestors bud and disappear, and all of these, and other processes of development, must keep uniform pace, so that none may outstrip another in the relatively rapid growth of all the parts involved. Added to these, it must be remembered that in the embryo, within a few millimeters of the area under consideration, is progressing the complicated evolution of the genito-urinary organs, themselves marvels of intricate complexity. Inaccurate coaptation of fissural margins, delayed closure of clefts, ectopia of fragments, sequestration of cell groups, delayed disappearance of structures that normally atrophy, and inclusion of contiguous elements, are possibilities, any one of which constitutes a deviation from the normal, which, later, during intrauterine life or post-natal existence, may jeopardize the well-being of the individual.

Within recent years few groups of tumors have attracted the attention of investigators more than new growths of the sacrococcygeal region; to review, even briefly, any large part of the literature of the subject would exceed the proper limits of this paper. Borst (1) has summarized the known facts and promulgated theories to 1902, and little has been added by the more recent communications of Martina (2), Bartel (3), Chiari (4), Féré (5), Kirmisson and Bize (6), Tedenat (7), Graff (8), and others (9), all of whom have recorded important observations on the subject. Aside from dermoids due to the inclusion of bits of skin, the all-important feature (oncological) of these studies has been the attempted elucidation of the histogenesis of teratomata. The term "teratoma" is here used in a sense similar to "embryoma," preferred by Wilms and others for similar tumors arising in the ovary. The discussion has been warmest over the questions of the monogerminal or the bigerminal origin of these growths, and here — as is often the

[1] Nieberding, *Münch. med. Woch.*, 1904, li, 1384, in an excellent paper, has collected the nine then recorded cases of such an anomaly.

case in medicine — confusion has arisen because of inaccurately drawn distinctions rather than essential differences in opinion. Certainly many of these tumors may be considered monogerminal — that is, they have arisen from a single impregnated ovum. Of course, the simpler inclusion dermoids are of such origin, and a like explanation is reasonable for other cysts or neoplastic masses resulting from developmental errors, excessive hyperplasia of one or more elements, ectopia of contiguous tissues, and similar errors in development.

After the discovery of Kowalewsky, Middeldorpf sought to explain certain of these tumors as arising from structures forming the neurenteric canal, but such an explanation fails in cases where definite organs, or parts of organs, as limbs partly or wholly formed, nerves, parts of eyes, pulmonary structures, renal tissue, mammæ, bone, etc., are present. Tourneux and Hermann attributed the presence of nerve tissue intercallated between epithelial and connective-tissue elements to persistence of medullary coccygeal vestige. Borst suggests that brain-like structures may be of this origin. The view that Luschka's body may give rise to sacrococcygeal growths is now abandoned. None of the origins suggested offer an adequate explanation for the presence of definite organs, or demonstrable parts of organs, in such tumors.

I cannot discuss in detail the views of Marchand and of Bonnet as to the possibilities of such tumors arising from impregnated polar bodies or wandering blastomeres. On the assumption that but two polar bodies are present in most vertebrates, the explanation fails in cases like that recorded by Sänger, in which a single ovary contained five embryomata. The preliminary divisional products of ovular segmentation — blastomeres — at a stage prior to the formation of the blastodermic layers must possess potentialities which, theoretically, and according to Bonnet demonstrably, would be adequate to continued existence and more or less perfect independent development if separated from the original mass. On this theory it becomes possible to conceive that should blastomeres be detached and continue proliferating, they might give rise to a more or less successful attempt at the reproduction of tissues normally developed from properly constituted blastomeres, and in this way give rise to a structure containing the many tissues observed in teratoma. This ingenious explanation of ovarian teratomata — preferably embryomata — has proven very enticing, but falls short of the satisfactory when weighed against the many facts that might be adduced.

The bigerminal[1] origin of many sacrococcygeal tumors is most in accord with the known facts concerning embryology and neoplastic development. It is the commonly accepted explanation, for duplicated or parasitic monsters include the attachment to or implantation of one fœtus upon or within another. This theory places all tumors containing evidence of such highly organized bodies as could best be explained by the assumption that they originated from an impregnated ovum directly in line with the grosser developmental anomalies represented by autosite and parasite. Nearly a half-century ago, Virchow maintained the bigerminal origin of such tumors. In the neoplasms the impregnated ovum runs its course of aborted growth within the tissues of the autosite or host; in the other case it is attached to the otherwise more or less perfectly developed child; in one instance it is represented by the neoplasm, in the other by the parasite.

An interesting field of experimentation considered by Féré, Chiari, and also by Lucene and Legros, is the artificial production of tumors of this type by grafting young chick embryos into the cavities or tissues of hens. Space is not available for detailed discussion of their recorded observations, except to say that a certain measure of success has crowned their efforts; after seven weeks, skin with attached feathers has been found in the abdominal cavity. The grafts have developed cartilage and bone, the latter showing epiphyseal formation. Trachea-like structures, and even retinal elements, have been produced. About one year ago, my associate, Dr. Rosenberger, at my suggestion, began a series of experiments along the line indicated. So far the results have not been encouraging; apparently, they are negative. We hope, however, to re-

[1] For essentially the same purpose, Ballantyne prefers the terms "monosomatous" and "bisomatous" to the terms "monogerminal" and "bigerminal."

port them in detail in the near future. The question of homologous hen and chick, and other factors that need not be mentioned at this time, renders success largely a matter of accident, but failures, in the presence of recorded results, must not be taken as controverting the reported facts.

In the case reported by Dr. Keen the presence of such highly organized structures as trachea and glands, the separation (duplication?) of the sacral vertebræ and the suggestion of a structure — possibly a limb — shown by the X-ray — all point to the bigerminal origin of the growth.

I cannot close without saying something on the question of malignancy. As a rule, these tumors show no tendency to metastasis. Graff, Krukenberg, Montgomery, Czerny, and others have reported dermoids undergoing carcinomatous transformation, but that is something different from a primarily malignant tumor. The earlier writers thought malignancy the rule, and in some cases the microscope has indicated sarcoma; in one instance adenosarcoma, in another adenocarcinoma. Goldschmidt called his tumor a chondromyxolymphadenosarcoma. While complete removal is highly desirable, Phocas, Cazin, and Broca have shown that it is not absolutely necessary, as imperfect ablation may not be followed by recurrence. Thedenat calls attention to the great danger resulting from injury and infection; of the unoperated cases — when the tumor was present at birth — three fifths of the children die before the third year. The location favors injury, and slight trauma is often followed by disastrous infection, which, in turn, is most difficult to arrest. A sudden enlargement may be due to a mild infection that does not frankly manifest itself, and could easily be taken for excessive neoplastic activity. Hoppe states that in but two recorded cases is metastasis suggested, and his investigation of the reports (Beatson, Frank) leads him to discredit either the diagnosis of the primary tumor or the accuracy of the observer. Stolper, and also Calbet, thinks sacrococcygeal growths intrinsically benign. It is well to remember, however, that undescended testicle, supernumerary mamma, and other abnormalities or ectopias, are not infrequently starting-points for rapidly fatal malignant tumors. Excision is clearly wise. Another important point is the recently established relation between teratoma of the testicle and syncytioma malignum, the dangerous character of which is well known. I have one such specimen, the patient dying in a short time after the appearance of symptoms. Scott and Longcope (10), and also Frank (11), have recently recorded such cases.

BIBLIOGRAPHY

1. Börst. Die Lehre v. d. Geschwulst, Wiesbaden, 1902.
2. Martina. Deutsche Zeitschr. f. Chir., bd. lxxx, 1905.
3. Bartel. Wiener klin. Woch., Feb. 25, 1904, p. 213.
4. Chiari. Centralbl. f. allg. Path. u. path. Anat., Dec. 31, 1904, p. 987.
5. Féré. C. R. Soc. de Biol., tome v, p. 345, 1903.
6. Kermisson and Bize. Rev. d'Orthopédie, March 1, 1906, p. 141.
7. Tedenat. Assoc. Françias de Chir., xvi Congrès, 1903.
8. Graff. Deutsche Zeitschr. f. Chir., lxiv, p. 526, 1903.
9. Lucene and Legros. Bull. de la Soc. Anat. de Paris, 1902, No. 8, p. 764.
   Engelman. Arch. f. klin. Chir., bd. lxxii, h. 4, 1902.
   Winter. Inaug. Diss., Königsberg, 1905.
   Lissena. Beit. z. path. Anat. u. z. allg. Path., bd. xxxii, 1902.
   Hoppe. Deutsche Zeitschr. f. Chir., bd. lxvi, 1903.
   Anspach. Proc. Path. Soc. of Philadelphia, Nov., 1903, p. 209.
10. Scott and Longcope. Bull. of the Ayer Clin. Lab. of the Penn. Hospital, Jan., 1905, No. 2.
11. Frank. Jour. Amer. Med. Assoc., Jan. 27 and Feb. 3, 1906; full list of other cases.