A CASE OF ANTERIOR AND POSTERIOR OR "COMBINED" SPINA
BIFIDA WITH DORSAL HERNIATION OF THE LARGE INTESTINE
AND UNUSUAL ABNORMALITIES OF THE SPINAL CORD AND NERVES.

A Thesis for the Degree of Doctor of Medicine

By

R. L. de C. H. Saunders. M.B.Ch.B.

Department of Anatomy,
Dalhousie University.
Nova Scotia.
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I. FOREWORD

The case forming the subject of this thesis was admitted to the Bradford Royal Infirmary in Yorkshire, England, through the instance of Dr. R. L. Langley of Heckmondwike of that county. During its short life it was there subjected to a careful clinical examination both by Dr Langley and Mr Hamilton Stewart of that hospital.

At death the specimen was forwarded by them to Mr Greig, Conservator of the Royal College of Surgeons Museum in Edinburgh. Conscious of its unique features Mr Greig initiated an inquiry, which was interrupted by his death soon after, when the specimen passed into the hands of Colonel W. F. Harvey, Superintendent of the Royal College of Physicians Laboratory in Edinburgh.

Colonel Harvey feeling that an explanation of its strikingly unusual features should be attempted, very generously placed the specimen at my disposal together with certain illustrations and other data. For this kindness, I should like to express my thanks.
I am much indebted to Dr Langley and Mr Stewart for clinical notes and radiograms, and to Mr John Borthwick of the Department of Anatomy, University of Edinburgh, for photographs.
II. INTRODUCTION

Cases in which there is a cleft vertebral column, or concomitant anterior and posterior spina bifida, with either a union or close relationship existing between the alimentary tract and central nervous system in the region of the cleft, are indeed rare.

In illustration of this, although Cruveilhier in 1824 appears to have been the first to describe such a case, a little more than a century later, Gruber (1926) and Feller and Sternberg (1929) were only able to collect thirty-one such cases between them. Korff in 1937 described a case, but was apparently unable to discover in the literature any further examples that had not already been considered by these workers.

By consulting the Index Medicus and other possible sources of information, the author has been enabled to increase this number to thirty-six. It is perhaps significant that there appears to be only two papers in the English language that deal directly with the subject, namely those of Adelmann (1920) and Bell (1923).
With regard to the present case, its unique character becomes immediately apparent when it is pointed out that it falls into group II of Feller and Sternberg's classification—a group which includes but two cases. The classification itself will be dealt with later, but it may be mentioned here that this group encompasses those cases in which some part of the intestinal tract passes back through a cleft vertebral column, and locally divided spinal cord, to present itself on the back.

The two cases belonging to this group are those of Lucksch (1903) and Adelmann (1920). In the first of these the vertebral defect was in the upper dorsal region, in contrast to the lumbo-sacral defect of the specimen now to be described, and it differed in other ways also. The second, which occurred in a calf, is the only one in which the position and character of the defect corresponded to the present case. It is of interest to note that Adelmann himself reported that he was unable to find an exactly similar case in
the literature of teratology.

In view of this then, the present specimen appears to warrant a detailed description and discussion of its features. The fact that the case lived for some time enhances its interest, and has led to a wide survey of the teratological and clinical characters of this and related types of abnormalities.
III. CLINICAL HISTORY

The case, a female infant, L. R. weighing 5 lbs. 1 oz. was admitted to the Bradford Royal Infirmary when two days old. This was the second child, the first being alive and well. The delivery had been straightforward and up to the time of admission the child had been taking the breast well.

On admission (7.V.35) the child was jaundiced. On examination a rounded red mass, apparently mucous membrane, was seen projecting from the lumbar region of the back. This projected through an opening which palpation revealed was possessed of a bony margin. The opening was one and three-quarter inches in diameter.

At the superior and lateral aspects of the mass, the mucous membrane became continuous with the skin, the junction of these two tissues being at the edge of the opening in the child's back. Between the lower side of the mass and the edge of the bony opening, a small fistula was observed from which meconium exuded. See Figs. 1, 2, 3, and 6.
An interesting and important feature was that a movement, suggestive of a peristaltic wave, was observed sweeping over the mass. Careful examination revealed no other abnormalities. Particular attention was paid to the anus.

The following day (8.V.35), a mixture of undigested curd and meconium was collected from the dressing which protected the mass on the child's back. Later in the day the child passed faeces per anum. Urine had been voided quite normally.

Some days later (10.V.35) the child was X-rayed, and a cleft was revealed in the spine. A catheter was introduced into the fistula below the mass on the child's back, and this emerged at the anus. With Abrodil in the catheter the child was again X-rayed several days later (13.V.35). This confirmed the belief that the bowel communicated with the surface through the cleft which was situated in the lumbar region of the vertebral column. See Figs. 4 and 5.

About a fortnight after admission the child was
still jaundiced. The intestinal mass was now discharging mucus freely and the skin below the mass had become ulcerated.

During the second month of hospital life the mass on the child's back grew steadily larger. Nevertheless, the child appeared to thrive and gain in weight, attaining a weight of seven pounds.

At the end of the third month the child was not taking well and there was a steady fall in weight, attended by fever. The child was now passing faeces both at the fistula and anus.

The beginning of the fourth month found the child emaciated in appearance, with sunken eyes and continuing to lose weight. The child died at the beginning of the fifth month. (9.IX.35)

Death was regarded as due to inanition, the child having refused food and lost weight for almost a month before its demise. The septic focus round the protruded bowel was regarded as the cause of the fever and of the inanition. From the day of admission there
had been a degree of jaundice; sepsis of the ulcerated mass and the discovery of an abnormally placed common bile duct must be regarded as factors contributing to its undue persistence.
IV. ANATOMICAL FINDINGS

The body was that of a well formed female, aged 4 months, presenting no abnormality other than the mass which protruded from the lumbar region of the back. The crown-rump length was 13 inches, and the length over-all 19 inches.

Head and Brain

The head, posterior to the bregma was somewhat flattened from above downwards. In size it appeared to conform with the age and proportions of the child, having a circumference of 37 cms. It was well covered with hair and the face was quite normal.

Following the removal of the brain, the cranium was examined. The occipital and sphenoidal fontanelles had closed. The frontal and mastoid fontanelles, although small, were readily palpable. The average diameter of the frontal fontanelle, as determined by Elasser's method, proved to be 1.05 cms. Data on the post-natal involution of this fontanelle give 2.4 cms. as the average diameter for a child 3 to 6 months old (Abt 1923). Despite this advanced degree of involution, the sutures elsewhere showed no signs of premature obliteration.
The basis cranii looked backward in a marked degree, the clivus being quite vertical. The tentorium cerebelli was in consequence unusually oblique. The falx cerebri passed obliquely backwards toward the right, so that the right occipital fossa was very much smaller than its fellow on the left.

There was a similar disparity in the size of the occipital lobes of the cerebrum, but otherwise no unusual feature presented itself in either the cerebrum, cerebellum or cranial nerves, beyond the fact that the cerebellum appeared to be somewhat small, although normally shaped.

**Thorax**

The thoracic index was 79. Examination of the thoracic cage revealed that ossification was well advanced in the ribs, and that the manubrium and upper three pieces of the sternum each possessed an ossific centre. The thoracic vertebrae and ribs will be discussed later.

The lungs with their lobes and fissures were normal. The thymus was composed of two inferior and lateral masses and a median superior, being of the
trilobar type of Coplin.

The heart appeared to be small, but normal as regards shape and position. No vascular anomalies were noted. Neither the ductus arteriosus nor the foramen ovale was patent.

The diaphragm was normal.

The Back

From the thoraco-lumbar junction a circular protrusion, 67mm. in diameter, rose to a maximum height of 20mm. above the surrounding skin level. It was more rounded and prominent superiorly and to the left, than inferiorly and to the right. (Figs. 2 and 3).

It presented, rather below its centre, a leaf-like protrusion of mucous membrane, which measured 56mm. in its longest diameter, 25mm. in maximum thickness, and had an arc of 113mm. as a free margin. The long diameter was directed obliquely and distally from the left to the right.

The free border of this mucous pad was slightly irregular, of a brownish tint and slightly shrivelled, as if it had become rather dry. Its surface was rugose,
showing diverging striae and between them short transverse linear impressions.

On the superior and lateral aspects of the pedicle of the leaf-like protrusion, the mucous membrane became continuous with the skin surrounding it. At its lower side however, was a small opening or "accessory anus", being the fistula already referred to in the clinical notes.

At the orifice of the fistula, the mucous membrane lining it became continuous above with the mucous pad, and below, with the skin of the back. Internally the fistula communicated with the lumen of a hernial loop of large intestine, which consisted of an entering and a returning limb. Reference to Fig. 6 will make it clear that the mucous membrane, lining the upper wall of this canal, was continuous internally with that lining the dorsal wall of the entering limb, while that of its lower wall became continuous with that of the returning limb. The greatest diameter of the fistula was 4.5mm.

The skin around the leaf-like projection was
mottled and had an irregular white zone which in turn was bounded by skin which was more pink than normal. The white part had a cicatricial or membranous character which suggested the avascularity of a commencing necrosis.

A section taken across the mucous pad, transverse to the spinal axis, showed (i) a covering of intestinal epithelium with corresponding glandular structure, characteristic of the mucosal surface of the large intestine, (ii) submucosa with lymphoid aggregations and ordinary loose fibrous structure, (iii) a muscularis layer which showed circular and longitudinal fibres, and (iv) a large group of Pacinian corpuscles in the mesentery.

These microscopic findings, together with the macroscopic, pointed to this mucous pad or projection being of large intestine origin.

**Abdomen**

On opening the abdomen the striking features were (i) the absence of the transverse colon, and (ii) on drawing aside the coils of small intestine, the presence of a hernial loop of large bowel which made
its way backwards into a large low-lying and medianly placed fossa, which was lined with peritoneum (Fig. 7).

Intestinal Tract and Peritoneum

The stomach, which was J shaped, was placed vertically in the upper left part of the abdomen, to the left of the median plane, its sharply upturned pyloric portion terminating in the middle line at the level of the 12th thoracic vertebra, just above the aforementioned midline fossa.

The upper part of its greater curvature was connected by the gastro-splenic and lienorenal ligaments to a spleen of average proportions, and a large low lying left kidney. An accessory spleen—the size of a small pea—lay in the lienorenal ligament, under cover of the spleen itself.

The lower part of the greater curvature gave attachment to the greater omentum. This omentum was continuous above and on the left with the gastro-splenic ligament, while below and to the left it was attached to the antero-lateral aspect and lower pole of the left kidney. On the right it blended with the left side of
the mesentery. Its lower free margin, which was short, extended, therefore, between the mesentery of the small intestine on the right, and the lower pole of the left kidney on the left.

There being no transverse colon, the posterior layers of the greater omentum ascended directly to the lower border of the pancreas, being adherent to the anterior surface of the large low-lying left kidney.

The proximal part of the duodenum was adherent to the back wall of the abdomen. Although this part passed upwards and to the right, to end on the right kidney at the level of the 11th thoracic vertebra, it preserved the peritoneal and other relations generally associated with the first part of the duodenum. The common bile duct and main (dorsal) pancreatic duct opened on to a well formed duodenal papilla in its upper right part.

The remainder of the duodenum had retained its embryonic form. It was attached to the back wall by a mesoduodenum which contained the head of the pancreas, and lay anterior and to the right of the superior
mesenteric artery, which passed directly into the mesentery.

The mesoduodenum inclined obliquely downwards and to the left, passing insensibly into the mesentery. The mesentery, whose root was very short, and contained very little fat, might be said, owing to its continuity with the mesoduodenum above, to extend from the neighbourhood of the medial side of the left kidney down to the left iliac fossa.

A continuous "common mesentery" thus passed from the fixed proximal part of the duodenum on the right, across and down towards the left kidney and left iliac fossa, thus passing immediately above and then along the left side of the large medially placed peritoneal fossa, already referred to (Fig.7).

The caecum was mobile, and together with the lower end of the ileum was suspended by the distal part of the mesentery. They lay in the medial part of the left iliac fossa, the lateral part of that fossa being occupied by the lower part of the left kidney. The mesentery had therefore an additional border -
a lower border - which passed from the medial side of
the lower pole of the left kidney across to the ter-
minall ileum and caecum.

As already stated, the proximal part of the duo-
denum was adherent to the back wall of the abdomen, and
passed upwards and to the right to form an acute angle
with the remainder of the duodenum which then passed
downwards and to the left, to become continuous with
the jejunum - there being no flexure between these two
parts of the intestine.

The small intestine measured 55 inches (137.5cms)
from the pyloro-duodenal to the ileo-caecal junction.
It lay chiefly to the right of the midline, the left
side of the abdomen being occupied by the vertically
disposed stomach and the large low-lying left kidney.

The terminal part of the ileum entered the right
side of the caecum, owing to the latter being trans-
posed to the left side. The caecum was of the
"dropped cone" type and gradually tapered into the
vermiform appendix.
The ileum and caecum having united, there passed backwards from them, with a slight inclination towards the right, a short segment of large bowel which communicated with the "accessory anus" or fistula lying immediately below the mucous pad on the back. For convenience it will be referred to either as the "ascending colon" or the "entering limb" of the hernial loop of large intestine.

The Midline Fossa

This entering limb of bowel made its way into a peritoneal fossa which occupied the midline in what would normally have been the position of the lumbar and upper sacral vertebrae.

Another short segment of bowel, which was also continuous with the "accessory anus" on the back, emerged from the lower part of this fossa, and there became continuous with the rectum. It will be referred to in future as the "descending colon" or "returning limb" of the hernial loop.

The "returning limb" lay immediately below the "entering limb" and passed from the depths of the
peritoneal fossa with a slight downward inclination toward the left, before it terminated in the rectum. No abnormality was observed in either the rectum or anal canal.

The peritoneal fossa which occupied the midline, was bounded above and below and to the right by a crescentic fold of peritoneum. This fold ran up to join the peritoneum clothing the right side of the head of the pancreas i.e. to the right side of the mesoduodenum (Fig. 7).

The left side of the fossa was less definite, for there the peritoneum passed without interruption from the left side of the fossa on to the left iliac fossa, although, as previously stated, it was flanked above and to the left by the attachment of the "common mesentery" of the duodenum and small intestine.

At the upper and lower limits of the fossa the peritoneum was reflected on to the entering and returning limbs of the herniated loop of bowel. On either side of these limbs the peritoneum extended far back before being reflected on to the intestine, forming a
cul-de-sac on either side, of which that on the right was deeper than that on the left.

Since a blunt probe passed into these culs-de-sac could be felt beneath the skin of the back on either side of the dorsal mucous pad, it was surmised that the peritoneal reflection on to the intestine took place beneath the skin of the back. This surmise was confirmed as dissection proceeded, and it was found that the reflection was separated only by a fibro-fatty mass from the skin of the back. This fibro-fatty mass was partially responsible for the circular protrusion which surrounded the leaf-like mucous mass on the back; the rest of that protrusion being, as will be seen, produced by a dural protrusion and two laminar ridges skirting the vertebral cleft.

Palpation indicated that this midline fossa, which contained the limbs of the herniated loop of bowel, was bounded on all sides by bone. Dissection revealed that it was embraced by the right and left halves of the cleft vertebral column, which were clothed with peritoneum. The exact vertebrae entering
into the formation of the vertebral cleft and forming the bounding elements of this fossa will be described in detail in the section dealing with the vertebral column, but in order to facilitate the further description of the abdominal viscera brief reference will be paid to them here.

The vertebral cleft, as reference to Fig. 8 will show, extended from the 1st lumbar to the 2nd sacral vertebra inclusively. It was bounded on the right side by the right halves of the 1st, 2nd, 3rd, 4th, and 5th lumbar, and 1st and 2nd sacral vertebrae, while, the left side was bounded by the corresponding left halves of these pieces, and by an additional piece bearing a short rib, which lay intercalated between the 12th thoracic vertebra and the 1st left lumbar hemi-vertebra.

**Genito-urinary System (Fig. 9)**

The right kidney was well formed and lay opposite the 11th and 12th thoracic vertebrae and the first four right lumbar hemi-vertebrae. Its lower pole, owing to the angulation of the right hemi-vertebrae lay
immediately in front of the posterior part of the right iliac crest.

The kidney itself was normal in size, shape, and consistence, and showed some slight signs of foetal lobulation. Its ureter descended in a tortuous manner first across the inferior vena cava and then across the right common iliac artery, just at its point of bifurcation, before reaching the side wall of the pelvis.

The left kidney was very irregular in outline and showed marked foetal lobulation. While it possessed an upper and lower pole, it was somewhat prismatic in shape, having a medial, as well as an anterior and posterior surface. A renal artery derived from the left common iliac artery entered a slit on its medial aspect, while the ureter and renal vein emerged from a misshapen hilum on the anterior surface. The kidney was low in position, its medial aspect lying opposite all five left lumbar hemi-vertebrae. Thus its lower part was situated within the left iliac fossa.
The left ureter descended over the anterior aspect of the kidney, and the left psoas in the left iliac fossa, prior to crossing the external iliac artery immediately below its origin. It was accompanied by the left ovarian vessels.

The right supra-renal was normal both in its shape and immediate relations. The left, while semilunar in shape, was somewhat flattened, doubtless due to compression afforded by the body of the pancreas which crossed it. Although the left gland surmounted the left kidney, the lower portion of the posterior surface - normally related to the kidney - appeared to be unduly large, extending from the upper pole of the kidney down to the misshapen hilum on the anterior surface, thereby excluding the pancreas from immediate contact with this kidney - a finding not surprising in view of the low position of the kidney. As regards size, both suprarenals appeared to conform with the age of the child, each being somewhat less than a third of the respective kidney.

The bladder was normal in shape and position.
considering the age of the child, being spindle shaped and mounting up into the abdomen. The vagina was likewise normal but the uterus was bi-cornuate, the left horn being more elongated and slender than the right. The uterine tubes and ovaries lay within the false pelvis.

**Liver and Pancreas**

With regard to the liver, nothing unusual was detected.

As for the pancreas, the head lay within the meso-duodenum below the proximal fixed portion of the duodenum. Its neck, related anteriorly to the lesser sac and pyloro-duodenal junction, was related posteriorly to the portal vein and its tributaries, as well as to a vein, presently to be described, which ran up in the crescentic peritoneal fold bounding the right side of the midline fossa, and terminated in the right side of the superior mesenteric vein. The body of the pancreas passed obliquely upwards, backwards and to the left towards the spleen, passing firstly just above the spinal cleft, and then across the left suprarenal. The latter relationship being due, as shown
above, to the low position of the left kidney.

The dorsal pancreatic duct opened, as did the common bile duct, into the proximal part of the duodenum, at a point 2cms. to the right of the pyloric valve, both however opening independently upon the duodenal papilla. The ventral pancreatic duct joined the common bile duct half a centimetre from the point where the latter duct pierced the duodenal wall.

Here the terms ventral and dorsal have been employed, for while the ventral and dorsal pancreatic elements had undergone fusion, giving an apparently normal pancreas, the ducts had not done so, but had retained the simpler pattern associated with the early stages of pancreatic development.

Abdominal Vessels

The general arrangement of the abdominal vessels in relationship to the spinal cleft is indicated in Fig.9.

The aorta descended in the midline until the 12th thoracic vertebra or upper limit of the spinal cleft had been attained, when it inclined to the right and
lay in front of the right lumbar hemi-vertebrae. It thus passed from a position above the peritoneal fossa to its right side. It divided opposite the 4th right lumbar hemi-vertebra into two common iliacs, both of which at first lay in front of the 5th right lumbar hemi-vertebra.

Thereafter the right common iliac artery continued down in normal fashion, dividing into its two terminals just below the 1st and 2nd right sacral hemi-vertebrae or right sacro-iliac junction.

The left common iliac however, skirted the lower limbus of the vertebral cleft, by passing across the 1st and 2nd right sacral hemi-vertebrae and 3rd sacral vertebra, before it reached the left side. Immediately before it divided into its two terminals just below the 1st and 2nd left sacral hemi-vertebrae, it give off a renal branch to the left kidney. This branch ascended across the left hemi-vertebrae before it entered the medial aspect of the malformed left kidney opposite the 2nd left lumbar hemi-vertebra.

The coeliac artery arose from the front of the
abdominal aorta at the level of the upper border of the 12th thoracic vertebra and appeared at the upper border of the pancreas to break up into its customary terminals. The superior mesenteric arose immediately below it. There was no inferior mesenteric artery.

The superior mesenteric artery after emerging from behind the pancreas, passed into the "common mesentery", to the left of, but on a deeper plane than the free portion of the duodenum, thereby preserving its embryonic relationship to it. Its branches in the mesentery were distributed to the pancreas, small intestine, caecum, and "entering limb" of the hernial loop of bowel, but the mesentery itself contained remarkably few vessels and arterial arcades.

A branch left the superior mesenteric artery, and passed downwards and to the right into the crescentic fold of peritoneum which passed round the right side of the peritoneal fossa, and so made its way to the right side of the rectum. It was accompanied by a vein, which occupied the free edge of the fold, and which, draining the rectum, passed up to terminate
in the right side of the superior mesenteric vein behind the neck of the pancreas. These vessels appeared to take the place of the missing inferior mesenteric vessels, being associated with the "returning limb" of the hernial loop, as well as with the rectum. See Fig. 7.

The inferior vena cava was formed behind the right common iliac artery by the union of the two common iliac veins, at the level of the 5th right lumbar hemi-vertebra. The right and left common iliac veins received their usual tributaries. The left common iliac vein, like its artery, passed below the vertebral cleft, passing from left to right below its companion artery.

The inferior vena cava ascended on the right hemi-vertebrae and right psoas. It was joined by the right renal vein opposite the 12th thoracic vertebra, that vein having ascended obliquely upwards from the rather large hilum of the right kidney, in order to join it at that level.

The left renal vein, emerging from the anterior
surface of the left kidney, ascended in an oblique groove on that surface, to attain the upper pole of the kidney. It then passed just above the upper limit of the vertebral cleft, by crossing in front of the abdominal aorta immediately below the origin of the superior mesenteric artery, and terminated in the inferior vena cava at the same level as its fellow on the right.

The left renal artery, as has been seen, arose from the left common iliac artery. It ascended over the left hemi-vertebrae to attain the medial aspect of the left kidney. The right renal artery arose opposite the 12th thoracic vertebra immediately below the superior mesenteric artery.

**The Vertebral Column**

As stated in the clinical history, palpation had revealed a bony margin surrounding the pedicle of the protruding mucous mass, and led to a radiological examination and the discovery of a spinal cleft. It will be recalled also that the introduction of a catheter into the fistulous opening below the mucous pad, resulted in its emergence at the anus, and the
natural conclusion that this split or cleft conducted the channel which connected the mucous pad and its opening with the bowel.

Dissection revealed a vertebral column, which was normal in appearance down to the level of the 7th thoracic vertebra. Thereafter it showed several irregular vertebrae, and then in the lumbar region proceeded to split into two halves, only to reunite in the sacral region, after enclosing a somewhat piri-form shaped aperture.

Ossification (Harris 1922) commences in the centre of the lower dorsal region at about the 7th week of intra-uterine life, whence it spreads cranially and caudally, reaching the odontoid process and fifth sacral vertebra at the fourth and fifth month respectively. Neural arch ossification commences in the atlas in the 7th week and extends caudally, reaching the third sacral in the 7th month. According to Cunningham's text book the union of the laminae begins in the lumbar region soon after birth, and, spreading cranially, is completed in the cervical region early in
the second year of life, but is deferred in the sacrum till between the seventh and tenth years. After the fusion of the laminae the ossifying process extends into the spinous processes.

The relative size of the vertebral and neural arch centres suggested that ossification had proceeded along these lines. The neural arch or laminar centres showed no signs of fusion, and from, and including the 7th thoracic vertebra upwards, the vertebrae appeared to be normal in every respect.

Below the 7th-thoracic vertebra (see Fig.8) the appearances were such as at first to suggest that little more could be said than that there had been an error of segmentation and divarication of the vertebral halves occasioned by the syngenetic establishment of a notochordal defect, local duplication of the neural tube, and formation of an axial ento-ectodermal connection, as suggested by the hernial loop communicating with the skin of the back. This aspect however will be dealt with later.

In order to arrive at a decision which would permit the computation of a vertebral formula, if only for descriptive purposes, the entire vertebral
column with its contained spinal cord, nerve roots and ganglia, was carefully dissected.

As already stated, the column above the 7th thoracic vertebra was normal. The left half of the 8th was deeper than the right, so that the whole vertebra lay obliquely, with a downward inclination toward the left.

The vertebral piece below this, at first sight, apparently possessed three distinct, ovoid, ossific centres, of which two were on the right and one on the left. On the removal of the periosteum, and after a little gentle scraping of cartilage - a procedure rendered necessary by the fact that the X-ray appearances while suggestive, were not decisive - it was discovered that the upper right centre and the one on the left were in continuity. They were united by a waist, and this, together with their disparity in size, gave them an appearance not unlike the vertebra immediately above - the 8th. They were regarded as representing the 9th thoracic vertebra. The lower right centre of this triad appeared
therefore to be the right half of a deficient 10th thoracic vertebra. It was separated from the upper right centre, i.e. right half of the 9th, by cartilage, which in turn supported the head of the 10th right rib.

The medial border of this lower right centre, or right half of the deficient 10th, lay very close to the left half of the 9th, but was not continuous with it.

If Fig.10 be referred to, it will be seen that each portion of this composite piece had a half arch associated with it, but that there was no 10th left half arch, it being deficient, like the left half of that vertebra.

The belief that this composite piece represented the 9th thoracic vertebra, and the right half of a deficient 10th, is based on the evidence of the vertebrae and their half arches in this region, and the relation of the ribs and spinal nerves thereto.

On the right side, the right half of the 9th thoracic vertebra and its half arch were associated
with the 9th right rib, which was attached both to them, as well as to the 8th thoracic vertebra, and the intervening disc. The 9th thoracic nerve of this side emerged below the pedicle of the 9th right half arch, emerging therefore from the inter-vertebral foramen between the right halves of the composite piece, proving that the upper half must be the 9th, and the lower, the right half of an incomplete 10th.

The right half of the 10th had a half arch, and to these the 10th right rib was attached, as well as to the cartilage between the right halves of the composite piece. The 10th right thoracic nerve emerged from beneath the pedicle of this half arch.

The 11th right thoracic nerve appeared from beneath the pedicle of the 11th right half arch. This half arch and its vertebra gave attachment to the 11th right rib.

Everything therefore on the right side of this region was quite normal and orderly, and the greater regularity of all the spinal nerves, ribs, vertebrae and hemi-vertebrae on the right side of the specimen,
as compared with the less regular arrangement of these structures on the left side, lends support to the belief that the right halves of the composite piece, are, as stated above, the right half of the 9th, and the right half of an incomplete 10th.

Further evidence is forthcoming on examination of the left side of this region. On this side, the left half of the 9th thoracic vertebra and its half arch, supported the 9th left rib, as did the 8th thoracic vertebra and the intervening disc. The nerve, serially the 9th left thoracic, which emerged from beneath the pedicle, of this, the 9th left half arch, was large, and its roots appeared to arise from an area almost equal to that which gave rise to the 9th and 10th thoracic nerves on the right side.

The rib and nerve, immediately below this, serially the 10th left rib and 10th left thoracic nerve, were associated with the half arch of the succeeding piece - the 11th thoracic vertebra as determined by the standards of the right side, which pointed to these being in reality the 11th left rib.
and 11th left thoracic nerve. This was further supported by the fact that this nerve was attached to the cord opposite the 11th right thoracic nerve.

All of which is in accordance with the pattern of the right side, and which suggests that not only is the left half of the 10th thoracic vertebra missing but also its half arch and rib, and that, either the 10th left thoracic nerve is also absent, or that the large nerve seen emerging beneath the pedicle of the 9th left half arch, is in reality the single representative of the 9th and 10th left thoracic nerves.

On the converse of Birmingham's principle (1891), who pointed out that if a vertebra is intercalated there should be no nerve to one vertebra, there should to quote Brash (1915), be two nerves to one vertebra if there is excalation of a vertebra, or to follow his line of thought, two nerves which have run together to form a single representative. This, as a working hypothesis, would explain the missing half of the 10th thoracic vertebra etc., as due to the 9th nerve being the single representative of the 9th
and 10th left thoracic nerves - a supposition, which as will later be seen, derives further support from both the size and mode of origin of the nerve in question.

Had the composite piece, representing the 9th thoracic vertebra and right half of an incomplete 10th, fused with the succeeding piece, the 11th, the result would be an almost perfect mirror image of Brash's case of an anomalous vertebral column. In his case, the 3rd, the left half of the 4th, and 5th cervical vertebrae having fused, it was suggested that the absent half was the 4th, and that the bone condition was secondary to the nerve abnormality, which consisted of a single nerve on the right corresponding to two on the left.

The piece following on this composite representative of the 9th thoracic vertebra and right half of the deficient 10th, was a diamond shaped 11th thoracic vertebra. It has been pointed out that the right side of this vertebra was associated with the 11th right rib and 11th right nerve, while the left side
was attended by what were serially the 10th left rib and nerve but which were regarded as the 11th left rib and nerve.

The 11th thoracic vertebra was followed by a triangular 12th, whose apex was directed downwards toward the cephalic limbus of the vertebral cleft. The right side of the latter was attended by the 12th right rib and nerve. On its left side were the serially 11th left rib and nerve, which were regarded as being in reality the 12th left rib and nerve, both because the nerve emerged from beneath the left half arch of this piece - the 12th thoracic as judged by the standards of the right side - and because the nerve was attached to the cord opposite the 12th right thoracic nerve.

**The Hemi-vertebrae Flanking the Vertebral Cleft**

**The Right Side of the Cleft**

Below the 12th thoracic vertebrae on the right there lay a series of hemi-vertebrae, which, although disposed so as to form an arc convex toward the right, showed no marked rotation in that direction. There
seemed little doubt that this series, which formed the right side of the cleft, consisted of the right halves of the 1st, 2nd, 3rd, 4th, and 5th lumbar, and 1st and 2nd sacral vertebrae. They proceeded downward in quite a regular fashion, and were associated with their appropriate half arches. The lumbar and sacral nerves emerged in orderly sequence below the vertebra of the corresponding number.

Their medial borders presented an even smooth surface toward the cleft. The right halves of the 5th lumbar and 2nd sacral were united medially by a bridge of bone, possibly resultant upon the wedge shape of the right half of the 1st sacral, and the consequent local angulation.

The right aspect of the cleft was in consequence formed by the smooth medial border of these halves, by the right half of the spinal cord enshoathed by its membranes and lying within the right half of the vertebral canal, and more posteriorly, by a ridge formed by the fused half arches of these hemi-vertebrae.
The Left Side of the Cleft

The first piece below the 12th thoracic vertebra on the left, was a hemi-vertebra which had a double centre of ossification and bore a short rib 1.2cms. in length. It has already been pointed out that the 11th thoracic vertebra articulated with the 11th right rib, and what was serially the 10th left rib, but in reality the 11th, there being no 10th left rib. In consequence of which it is evident that the serially 11th left rib articulating with the left side of the 12th thoracic vertebra and its half arch, must be the true 12th rib, and that this short rib must be a 13th rib associated with an intercalated hemi-vertebra, although it is serially the 12th.

It has been noted by Dwight (Brash 1915) that where the last pair of ribs is very short the penultimate are much longer than usual. It is interesting to note therefore that the length (head-tip) of the 12th right rib was 3cms., while that of the 12th left rib (serially the 11th) was 5.6cms. It should perhaps be mentioned that the 11th right rib measured 5.3cms.
In putting forward the evidence in support of the belief that this first hemi-vertebra on the left side of the cleft is an intercalated element, it would be well to bear Birmingham's principle in mind, namely, that if a vertebra is intercalated there should be no nerve to one vertebra.

The piece in question lay immediately over the point where the spinal cord divided into two. It is interesting to note that while the right division of the cord passed off from the right side of this point toward the right side of the cleft, the left side of the cord at this point gave rise to no nerve between the 12th (serially 11th) left thoracic nerve above and a small 1st lumbar nerve, which emerged on the medial side of the left half of the split column, below. This region of the cord was overlain by the intercalated half (See Fig.11).

If Fig. 8 be referred to, it will be seen that no nerve emerged from beneath either the half arch which the intercalated half bore on its lateral side, nor from the "accessory half arch" which lay in
relation to its medial side.

The rest of the hemi-vertebrae forming the left side of the cleft appeared to be the left halves of the 1st, 2nd, 3rd, 4th, and 5th lumbar, and 1st and 2nd sacral vertebrae. The identity of these is suggested both by counting up from the unsplit, although double nucleated, 3rd sacral vertebra, and by the arrangement of the lumbar and sacral nerves on this side.

It is true that no nerves emerged from beneath the half arches of either the 1st or 2nd left lumbar hemi-vertebrae, but they did appear on their medial aspect, thereby permitting their identification. If Fig. 8 again be referred to, it will be seen that while the 1st left lumbar nerve appears to emerge from beneath the "accessory half arch" on the medial side of the intercalated half, it is in reality separated from it by a distinct fibrous band. The 2nd left lumbar nerve is seen to appear from beneath the "accessory half arch" of the 2nd left lumbar hemi-vertebra, and then to cross the left half of the split column to join the 3rd left lumbar nerve.
It will also be noted that the 3rd, 4th, and 5th left lumbar nerves, as well as the left sacral nerves, emerged from their appropriate places, although the 3rd, and 5th lumbar and 1st sacral nerves were supported by small nerves which emerged from beneath the "accessory half arches" associated with the medial aspect of their hemi-vertebrae. These small nerves united and lost themselves in the tissue about the hernial loop.

The hemi-vertebrae and half arches forming the right side of the vertebral cleft are illustrated in Figs. 8 and 10. The simplicity of their arrangement does not merit further description, beyond noting that their half arches entered into a ridge which constituted the right posterior boundary of the cleft.

It is immediately evident that the same cannot be said for those forming the left side of the cleft. Their individual characters are now considered.

The first piece on the left, the intercalated hemi-vertebra, possessed two centres of ossification. The upper of these was small and ovoid, and while
separated by a fibro-cartilaginous disc from the 12th thoracic vertebra, was only separated by cartilage from the lower ossific centre. The whole piece lay very obliquely. Its half arch likewise lay obliquely (Fig.10), and assisted in forming a ridge with the half arches which lay below it - a ridge which formed the left posterior boundary of the cleft. A small "accessory half arch" lay in relation to its medial aspect (Fig.8).

The left halves of the 1st, 2nd, 3rd, and 4th lumbar vertebrae, inclined downwards and somewhat outwards, so that the last of these lay immediately above the left iliac crest. (The piece which occupied a similar position on the right side was the right half of the 5th lumbar.) Their surfaces were directed laterally, and their lateral aspects bore the pedicles of their half arches, which last entered into the formation of the left ridge mentioned above. (Fig.10).

Thereafter this half of the column showed a definite change in direction, passing almost horizontally towards the midline. This horizontally placed portion
consisted of the left halves of the 5th lumbar and 1st and 2nd sacral vertebrae. Their lower borders were fused with the ilium, the union being fibrous in the case of the 5th lumbar, but cartilaginous in that of the two sacral vertebrae. Their half arches also entered into the formation of the ridge constituting the left posterior boundary of the cleft.

The medial or cleft aspect of these left lumbar and sacral hemi-vertebrae, were all, with the exception of the 1st and 4th left lumbar, associated with a series of pedicles. The bony processes associated with these pedicles have hitherto been referred to as "accessory half arches" as that is what they appear to be. By bony and ligamentous union they formed a ridge that extended to and included, the "accessory half arch" of the intercalated piece above, and a bony spur projecting from the cleft aspect of the 3rd sacral vertebra, below. The ridge so formed, was sharp, and served as the anterior boundary of the left half of the vertebral canal.

The left side of the vertebral cleft consisted
therefore (i) of this ridge, (ii) of the left half of the spinal cord ensheathed in its membranes and lying in the left half of the vertebral canal, and (iii) more posteriorly, of the ridge formed by the fusion of the "true" half arches of this side (Figs. 8 and 10).

The Vertebral Arches (See Fig. 10)

As already stated, the neural arch or laminar centres of ossification, down to and including those of the 7th thoracic vertebra, showed no sign of fusion in the midline, the spinous processes being as yet cartilaginous and therefore normal considering the age of the child.

The oblique position of the 8th thoracic vertebra was reflected in its arch, wherein the cartilage-tipped right half arch overlay its similarly tipped fellow of the opposite side.

A similar obliquity obtained in the case of the arch of the 9th thoracic vertebra. Here the cartilage at the end of the 9th right half arch overlapped that of the 9th left half arch, which in turn overlapped the half arch of the incomplete 10th thoracic vertebra.

The 11th and 12th thoracic vertebrae, like the
upper members of the column, had cartilaginous spinous processes.

The arches, or rather half arches, below the 12th thoracic vertebra, were fused into two ridges, which flanked the vertebral cleft. On the medial aspect of these ridges was a rim of cartilage which represented the spinous element. Laterally there was a series of pedicles, which served to indicate the half arches entering into the ridge. At the cephalic end of the cleft, the ridges were united by a fibrous band that passed from the half arch of the intercalated hemi-vertebra on the left, across to the half arch of the 1st lumbar hemi-vertebra on the right. Caudally the ridges tapered, being completed by the sacral half arches.

The half arches of the lower three pieces of the sacrum were united by fibrous tissue, which formed the roof of the "open" sacral canal. Further consideration of Fig. 10 will make it clear that when the spinal cord divided into two, the left and larger portion passed more directly downward than the right, and that both having passed down beneath their res-
pective laminar ridges, united beneath the fibrous roof of the "open" sacral canal.

It will be immediately evident, that although the number of half arches actually bordering the cleft posteriorly, necessarily coincides with the number of half vertebrae bounding it anteriorly, the laminar ridges also included the half arches of the lower three sacral vertebrae. These three vertebrae were unsplit and lay below the vertebral cleft.

It was these laminar ridges that were palpated beneath the skin around the pedicle of the projecting mucous pad (see clinical history). They underlay the more peripheral part of the circular protrusion which lay about the mucous pad, and were largely responsible for its shape.

On viewing Fig.10, it will be seen that the right and left laminar ridges, and the right and left halves of the vertebral canal have been depicted therein as well as the sinuous and fibro-osseous ridge formed by the union of the "accessory half arches" of the pieces skirting the left side of the cleft.
The latter is depicted on the left side, together with the foraminae through which the small nerves, which emerged on the medial side of the left half of the split column, passed. Compare with Fig.8.

The Size of the Vertebral Cleft

The aperture, as may be ascertained from the illustrations, was piriform in shape. Its greatest diameter in the median plane was 26 mm., while the greatest oblique and transverse diameters were respectively 37 and 25 millimetres.

The measurement of 1 3/4 inches mentioned in the clinical notes was perforce the measurement from one laminar ridge across to the other.

Sacrum and Coccyx

It has been seen that the right and left halves of the 1st and 2nd sacral vertebrae lay on either side of the vertebral cleft, and that their half arches entered into the right and left laminar ridges.

The 3rd sacral vertebra formed the caudal limbus of the cleft. It was not split, but possessed two centres of ossification. The larger of these was on
the left, and had a bony spur associated with its cleft aspect. This spur formed the lower end of the ridge produced by the union of the "accessory half arches" of the left hemi-vertebrae.

The 4th piece of the sacrum also had two ossific centres, while the 5th had but one.

The half arches of these lower three pieces of the sacrum constituted the lower ends of the right and left laminar ridges. They also formed the sides of the short "open" sacral canal. The canal was short as the half arches of the 1st and 2nd pieces of the sacrum bounded the cleft, and there were only three unsplit sacral vertebrae.

The sacral half arches were well ossified, and the ossific centres mentioned above are of course those of the sacral centra. In addition to these however, there were two small centres of ossification in both the right and left costal elements of the 1st and 2nd pieces of the sacrum. They lay immediately above the 1st and 2nd sacral nerves, and would appear to corroborate still further the selection of those
pieces as the right and left halves of the 1st and 2nd sacral vertebrae. Neither the costal element of the 3rd or 4th sacral vertebra had such a centre, although it has been stated (Cunningham) that at birth there is one for the costal element on each side in the upper three or four vertebrae.

The sacro-iliac joint on either side was affected by the halves of the 1st and 2nd sacral vertebrae, and by the 3rd sacral vertebra. On the left side a fibrous union existed between the left half of the 5th lumbar vertebra and the ilium.

The coccyx consisted of four as yet cartilaginous pieces.

**The Curvatures of the Column as a Whole**

In the cervical region the column was straight, there being as yet no ventral convexity. The thoracic and lower sacral regions were concave ventrally, as was the region of the cleft (lumbar and upper sacral). The ventral concavity of the cleft region was masked somewhat by the fact that the right and left halves of the split column did not lie in the same plane, the
right half lying on a more anterior plane. Both halves of the split column were however in themselves slightly concave.

As a result of the absence of the left half of the 10th thoracic vertebra, there was, in the region of the composite piece, a slight lateral curvature, convex toward the right, which might have received more emphasis in Fig.8.

The right half of the split column formed a curve which was convex toward the right, and somewhat angulated in its lower third. The left half of the split column was convex toward the left, but its pieces formed a more even curve.

The left hemi-vertebrae were rotated outwards, i.e. laterally, but this was not so marked on the right.

The Vertebral Formula

The vertebral formula, when computed from the right side of the column is C7 Th 12 L5 S5 C4. It is necessarily the same on the left side if the pieces be considered serially, in that the intercalated
hemi-vertebra compensates for the incomplete 10th thoracic vertebra. Actually, however, the formula for the left side appears to be C7 ThI-9*II-13 L5 S5 C4.

Bearing Birmingham's principle in mind, and Brash's converse thereof, we have a plausible explanation of both the intercalated and excavated hemi-vertebra. It is difficult to ignore the influential role which the spinal nerves are said to play in the process of segmentation. Consequently when the disposition of the nerves displayed in this case is considered on the basis of the above principles, there is not unnaturally a tendency to regard the condition as being one in which there has been a suppression of half a segment in the region of the composite piece, and the addition of a half segment in the region of the cleft.

On the other hand, it is interesting to observe that in this case if the left half of the vertebral column be taken serially it appears to have the same formula as the right half; and further, that if the left halves of the 11th and 12th thoracic vertebrae and the "intercalated half" were each to be advanced
cephalically the lower three members of the thoracic column would be "repaired" and the symmetry of the column restored. This might suggest that the 11th and 12th thoracic vertebra are in a sense "ambivalent". For example, what has been described as the 11th thoracic vertebra might possibly be composed of the right half of the 11th and the left half of the 10th, and that in reality no suppression nor intercalation of material has taken place.

Although it is difficult here to reconcile the disposition of the nerves with this idea, the idea is recorded as it appears to be linked with a theory discussed later, which deals with the genesis of such abnormalities. In short it rests on the appreciation of the fact that the presence of a cleft notochord and an axial ento-ectodermal connection (now represented by the cleft spine and hernial loop) would bring about a local separation of the right and left skeletal anlagen and by preventing their proper union would result in a series of hemi-vertebrae. In the event of the axial connection undergoing some
measure of retrogression a disorderly union of right and left skeletal elements might take place at one or both ends of the axial connection. The form and number of the segments on either side would initially be determined by the disposition of the spinal nerves, but the extent of malunion between them would of necessity be qualified by the degree of retrogression of the axial connection.

**Intervertebral Discs**

In order to ascertain whether or not there had been a disturbance of the primitive embryonic axis, i.e. the notochord, intervertebral discs were removed from both above and below, as well as from either side of the vertebral cleft. The discs removed and subjected to a histological examination were those lying between the 6th and 7th thoracic vertebrae, the 4th and 5th sacral vertebrae, and the 1st and 2nd left lumbar and the 2nd and 3rd right lumbar hemi-vertebrae.

All of these possessed a nucleus pulposus. That within the intervertebral disc removed from the right side of the vertebral cleft was considerably larger
than that seen within the disc taken from the left side of the cleft.

In view then of the fact that the nucleus pulposus is a remnant of the notochord, the discovery of pulposal tissue on both sides of the vertebral cleft was taken as evidence that there had initially been a cleft condition of the notochord itself.

The Spinal Cord and Spinal Nerves

The spinal cord and its nerves were normal down to the level of the 7th thoracic vertebra. Opposite the 8th thoracic vertebra, the spinal cord showed a slight increase both in diameter and in the size of its central canal.

The cord descended until the upper limbus of the spinal cleft had been obtained. It then bifurcated behind the intercalated hemi-vertebra on a level with the 1st right lumbar hemi-vertebra. The resultant divisions were unequal in size. At the level of the 3rd right lumbar hemi-vertebra, or middle of the spinal cleft, the right division was 5 mm. and the left 7 mm. in diameter.
Both divisions gave off nerves, but whereas the right division gave off a single laterally disposed set, the left gave off both a lateral and a medial set. The central canal of both divisions was abnormally dilated, and had reduced each division to but a thin wall of nerve tissue.

The left division of the cord, enclosed in a dural sheath, continued directly down into the left half of the vertebral canal. It was flanked anteriorly therefore by the ridge formed by the "accessory half arches" of the left hemi-vertebrae, and posteriorly by the left laminar ridge formed by the fusion of the true half arches of those hemi-vertebrae Figs. 8 and 10.

The cord being cleft asymmetrically, the right division appeared to arise from the lateral aspect of the left. It arose from a base of 14 mm. in its vertical extent, and then passed obliquely across the upper part of the vertebral cleft in order to reach the right half of the vertebral canal. It tapered off rapidly, until at the middle of the cleft, it was, as stated above, only 5 mm. in diameter.
As the right division of the cord passed across the upper end of the cleft, both it and its dural sheath protruded backwards through the cleft, forming a meningo-myelocele. This expressed itself as a swelling beneath the skin somewhat above and to the left of the mucous mass on the back. It was therefore responsible in part for the circular protrusion on the back which surrounded the leaf-like mucous mass. The rest of that protrusion has been seen to be due to the fibro-fatty tissue which covered the two peritoneal reflections taking place beneath the skin of this region, and also to the right and left laminar ridges which skirt the cleft.

The two divisions of the cord passed down on either side of the cleft, each in its own half of the vertebral canal. Each was related medially to the structures transmitted by the cleft, and laterally, as well as posteriorly, to the half arches which formed its half of the vertebral canal. The structures lying between the dural sheaths of the right and left divisions of the cord, were the right and left peritoneal culs-de-sac, and between them, the entering and returning limbs of the hernial loop of bowel.
Immediately below the lower limit of the cleft, behind the 3rd sacral vertebra, the two divisions of the cord, as well as their dural sheaths, reunited. Here the left division appeared to join the right, for the terminal part of the cord, i.e. that part below the junction of the two divisions, appeared to be a direct downward continuation of the right division of the cord. The manner in which the right division branched off from the left at the upper limit of the spinal cleft, only to be joined by the left at the lower limit of the cleft, is reflected in the general disposition of the hemi-vertebrae about the cleft. See Fig.8.

The terminal part of the cord, or conus medullaris, tapered off until it reached the back of the 5th sacral vertebra, when it ended in a bulbous subcutaneous dilatation which lay behind the as yet unossified pieces of the coccyx. The greatest dimension of this dilatation was 8 mm. and it was regarded as a large ventriculus terminalis. The dura accompanied the terminal portion of the cord as far as this dilatation.
The Spinal Nerves

The Brachial Plexus was normal on both sides. It was formed by the 5th, 6th, 7th and 8th cervical, and 1st thoracic nerves and received a small intrathoracic communication from the 2nd thoracic nerve.

There were twelve thoracic nerves on the right side, and but eleven on the left side. The first eight pairs of thoracic nerves were normal. On the right side there were four nerves below that point, but only three on the left. See Fig. 8.

What were serially the 10th and 11th nerves on the left side, were judged to be the 11th and 12th thoracic nerves, both because they emerged from beneath the left half arches, of what were regarded by the standards of the right side to be the 11th and 12th thoracic vertebrae, and because their attachments to the cord were opposite those of the 11th and 12th right thoracic nerves.

From which it will be apparent, that what was serially the 9th nerve on the left side, appeared to correspond to two nerves - the 9th and 10th - on the
right side. This nerve was larger than either the 9th or 10th nerves of the right side. Its posterior root ganglion measured 3 mm. in diameter, whereas their ganglia each measured but 2 mm. in diameter.

It arose, in addition, from an area almost equal in size to that which gave rise to both the 9th and 10th thoracic nerves on the right side, and it pierced the dura exactly opposite the ligamentum denticulatum which separated the 9th and 10th nerves on the right side.

Apart from any inference made from the appearance of the vertebrae, the fact that more radicularia arose from what appeared to be the region of the 9th thoracic segment, than from the region of the 10th segment, suggests that if any nerve is missing, it is the 10th left thoracic nerve.

On the other hand, the size of the nerve and its ganglion, as well as its area of origin, suggest that it is the single representative of two nerves, namely the 9th and 10th left thoracic nerves.

On the right side of the vertebral cleft, the
regular and orderly sequence of the lumbar and sacral nerves permitted, as has been seen, a fair measure of confidence in the numerical identification of the pieces involved. The right lumbar plexus was of ordinary type, but its branches displayed a number of variations which justify a description.

The ilio-hypogastric nerve arose from the 12th thoracic. According to Jamieson(1903) this is not unusual, and he mentions that Schmidt found this arrangement in one out of three or four bodies, where the ilio-inguinal nerve was larger than normal. The ilio-inguinal nerve in this case was large. It arose from the 1st lumbar nerve, and while it emerged from the psoas major and passed down across the quadratus lumborum, it did not pierce the transversus abdominis with the ilio-hypogastric, but continued on across the iliacus to the lateral part of the inguinal ligament, where it simultaneously pierced both the transversus and internal oblique to enter the inguinal canal.

The genito-femoral nerve arose by means of two slender pieces from the ansa connecting the 1st and
2nd lumbar nerves. The lateral cutaneous nerve arose from the 2nd lumbar shortly after it had been joined by the communicating branch from the 1st lumbar.

The obturator nerve arose from the ventral divisions of the 2nd, 3rd, and 4th lumbar nerves, emerged from the medial border of the psoas major, and passed into the pelvis.

The femoral nerve was derived from the dorsal divisions of the 2nd, 3rd, and 4th lumbar nerves. Emerging from the psoas major immediately below the iliac crest, it ran downwards in the groove between the psoas and iliacus and passed behind the inguinal ligament into the thigh, where its branches were quite normal.

This - the femoral nerve - was peculiar however, in that a large nerve which arose from the union of the 2nd and 3rd dorsal divisions, before they joined the dorsal division of the 4th lumbar nerve, passed through and then obliquely outwards over the psoas major to join the medial side of the femoral nerve behind the inguinal ligament.
There were therefore two nerves lying on the anterior surface of the lower part of the right psoas. The medial nerve was the genito-femoral. The lateral was the nerve just described, which emerged from the psoas and crossed it obliquely to join the femoral nerve at the lateral side of the psoas.

This nerve appeared to be an accessory anterior crural (femoral) such as described by Jamieson. In its course it resembled the nerve described by him, while in origin it was not unlike those described by Winslow and Schmidt (Jamieson, loc. cit.).

The 24th nerve on the right side, or 4th lumbar nerve, was the furcal nerve, i.e. that nerve which distributes branches to both the lumbar and sacral plexuses. The lumbo-sacral trunk was therefore formed by the 4th and 5th lumbar nerves.

The sacral plexus was formed by the lumbo-sacral trunk, the anterior primary rami of the first three sacral nerves and part of the fourth. The coccygeal plexus was formed by the anterior primary rami of the coccygeal and fifth sacral nerves together with the
rest of the fourth. These plexuses both in the manner of formation and in the nerves which they gave off, were normal.

On the left side of the vertebral cleft the arrangement of the lumbar and sacral nerves was very complex, and in order to clarify their description, it should be pointed out that they emerged in two sets—one on either side of the left half of the vertebral column. The lateral set were large nerves which arose from the lateral aspect of the left division of the spinal cord, and emerged on the lateral side of the left hemi-vertebrae. The medial set, with the exception of one, were all small nerves. They too arose from the lateral aspect of the left division of the spinal cord, and so their nerve roots embraced the left division as they made their way toward the medial side of the left hemi-vertebrae. This set emerged from beneath the "accessory half arches" of the left hemi-vertebrae.

The lateral set consisted of the 3rd, 4th, and 5th lumbar and the sacral nerves. The medial set was composed of the 1st and 2nd lumber nerves and in
addition three delicate nerves which arose from the cord opposite the 3rd and 5th lumbar and 1st sacral of the lateral set; these last three nerves are, for convenience, referred to as the accessory 3rd and 5th lumbar and accessory 1st sacral. See Fig. 3.

The left lumbar plexus and its branches were therefore but an aggregation of anomalies. Before describing them, it should be noticed that there was on the lateral side of the left half of the vertebral column an area—between the 12th (serially 11th) thoracic nerve and the 3rd lumbar—which was devoid of any nerves, but which was compensated by the appearance of the first two nerves of the medial set on the inner side of that area. These two nerves were regarded as the 1st and 2nd left lumbar nerves, in virtue of the level of their origin in relation to the nerves above and below them, and because one of them participated in the formation of the femoral nerve. They were not "accessory nerves" in the sense of the lower three members of the medial set.

The ilio-hypogastric nerve arose from the 12th
(serially 11th) thoracic nerve. Although at its point of emergence it was separated from its parent trunk by a delicate fibrous band, it rejoined the trunk prior to pursuing its usual course. As on the right side there was no ansa of communication between the 12th thoracic and the 1st lumbar nerve.

The 1st lumbar nerve arose from the left division of the cord just below the point where the right division passed off towards the right. It appeared on the medial side of the left half of the vertebral column, and passed laterally to sink into psoas major. There was no ilio-inguinal nerve on this side of the body.

The 2nd lumbar nerve was the largest of the medial set of nerves. It too appeared on the medial side of the left half of the column, and passing obliquely downwards and laterally across this half of the column, entered the left psoas major to affect a broad union with the 3rd and 4th lumbar nerves in the substance of that muscle. It will be remembered that the 3rd and 4th lumbar nerves belonged to the lateral set of nerves. Both the femoral and obturator
nerves arose from this point of union, and a small accessory obturator nerve arose from the angle between them, and passed through the psoas to cross the pelvic brim.

The 4th lumbar was the furcal nerve and as its descending branch passed down to unite with the 5th lumbar to form the lumbo-sacral trunk, it was joined by a delicate communication from the accessory 3rd lumbar of the medial set of nerves. This communication like the 2nd lumbar nerve, passed medio-laterally through the psoas across the left half of the column.

The lumbo-sacral trunk thus formed, together with the first four sacral nerves, gave rise to the sacral plexus. The coccygeal plexus was formed similarly to that on the right side. The branches of these plexuses were normal.

On this side there was, over and above the absence of the ilio-inguinal nerve, no genito-femoral or lateral cutaneous nerve of the thigh. Despite a careful search in both abdomen and thigh, there was nothing to suggest that either they or their branches were replaced by
other nerves.

The lower three members of the medial set of nerves, i.e. accessory 3rd and 5th lumbar and accessory 1st sacral, united to form a looped "plexus" from which a solitary slender branch passed to lose itself in the tissue about the hernial loop.

A somewhat diagrammatic representation of the manner in which the spinal cord divided into two, and then reunited to form a conus medullaris, is given in Fig. 11. It will be noticed that the left division was larger than the right, and that there was no cauda equina in any correct sense of the term.

The spinal cord at first stretches throughout the whole length of the vertebral column. Toward the end of the third month of intra-uterine life, as the vertebral column and canal proceed to grow more rapidly than the cord, the latter is gradually withdrawn, so that at birth its lower end has attained the level of the third lumbar vertebra. According to Streeter (1919) a posterior root ganglion registers the original position of the segment relative to the vertebral column,
while the point of attachment of the posterior root moves as the cord moves. So that by locating these two points, one can determine the elongation of the nerve roots and thus obtain an index of the relative displacement of the spinal cord as regards the vertebral column.

Now the nerve roots associated with the right division of the cord, were very much longer than those connected with the left. That is to say, the distance between the posterior root ganglion and the point of attachment of the posterior root to the cord, was greater in the case of every nerve arising from the right division of the cord than in the case of those nerves arising from the left division. This measurement was very short in the case of both the medial and lateral sets of nerves belonging to the left division.

This would indicate that the left division of the cord had undergone little or no displacement as regards its half of the column, but that the right division had, as regards its half of the column, undergone some marked displacement. The disparity between
the right and left nerve roots in this region is too great to be due solely to disproportionate growth between the right and left halves of the column. It must therefore be explained as being due to the fact that the right division of the cord bulged backwards through the cleft, participating, as we have seen, in the formation of the meningo-myelocoele.

The abnormal length of the spinal cord, with the termination of the conus medullaris in a terminal vesicle on the back of the coccyx, is not surprising when we consider that the duplicated state of the spinal cord circumventing the vertebral cleft and its contents, precluded any possibility of the cord being withdrawn from its foetal status. In addition the fixation of the cord occasioned by the structures both in and about the cleft, had apart from interfering with its cephalic migration, doubtless dragged the cerebral peduncles caudalwards, thus accounting for the vertic- ality of the clivus and possibly for the smallness of the cerebellar hemispheres.
**Trunk Muscles and Limbs**

The lateral and anterior abdominal muscles were well developed. In the region of the vertebral cleft the psoades and sacro-spinales, as would be supposed, followed the curves of the skeletal defect.

As regards the muscles of the thorax and limbs they too appeared to be well developed. The limbs were well formed.
V. DISCUSSION

(1) Classification

This case falls into the category of an anterior spina bifida, and into the subgroup, to be explained later, of an anterior and posterior or combined spina bifida. It is sometimes convenient also to refer to a deformity of this type simply as a vertebral cleft, after the manner of the German workers who employ, among other terms, the expression "Wirbelspalte". The multiple abnormalities presented by cases of this type render their assessment and classification a difficult task, but a survey of the records appears to warrant the following statements.

In general the condition of combined anterior and posterior spina bifida may be said to occur in any of the four regions of the vertebral column. It is furthermore frequently associated with a cranial defect. When the genesis of this type of spina bifida is considered, it will become apparent that it is a defect which may occur anywhere within the domain of the notochord.
There is a more or less extensive cleft of the vertebral bodies and arches in every case, although of course it is not always immediately apparent.

When there is a gross vertebral defect the halves of the vertebral bodies tend to arch laterally, so that the cleft takes the form of a lyre. The vertebral column in such cases not infrequently presents a marked lordosis in the region of the cleft; when this occurs in the upper reaches of the vertebral column, the neck is very short and the head appears to rest upon the shoulders, thereby imparting a characteristic appearance to the child.

In the less extensive type of defect only one or two vertebrae may be involved, and the vertebral halves may show no tendency to diverge.

The alimentary tract usually lies in close relationship to the vertebral defect. Great variation however occurs in its disposition. In some cases part of it passes through the vertebral cleft and then either protrudes upon or opens on to the back. In others it is connected with the spinal portion of
the central nervous system either by a strand or a diverticulum, which likewise traverse the vertebral cleft. Such a strand usually consists of nerve tissue at its "neural" end, and smooth muscle at its "visceral" end, while a mucosal lined diverticulum not infrequently tapers off into a strand which then makes its way toward the cleft. The designation of such a strand or diverticulum as a "neurenteric" connection has much to commend it, provided it be employed only in a literal or descriptive sense, and not in the sense of its being a persistence of the neurenteric canal of the early ovum.

The vertebral canal or gutter and its contents are necessarily, in the region of the cleft, more or less closely related to the alimentary tract. When it is appreciated that the canal or gutter may be occupied by either a medullary plate or definite spinal cord, in either a single or cleft form, it will be understood why the alimentary tract in some cases connects with the central nervous system, and in others passes through it on to the back.
The oesophagus, stomach and small intestine appear to be the portions of the alimentary tract most commonly connected with such vertebral clefts, although involvement of the large intestine is not unknown. See Fig. 12. Certain alimentary adnexa sometimes become involved. The spleen, for example, may be carried into the cleft along with the stomach, while the position of the bile duct and pancreas not unnaturally depend on the situation of the duodenum.

In many cases there is a diaphragmatic defect, particularly on the left side, and the result is that the stomach, small intestine and even large intestine, may be found displaced to a greater or lesser degree above the diaphragm. These structures then lie either in a mediastinal extension of the peritoneal cavity or actually within one of the pleural cavities.

Such diaphragmatic defects and visceral displacements appear to be more commonly associated with a defect in the upper part of the vertebral column, and doubtless depend in certain cases on a
"neurenteric" connection established at an early date in ontogenetic history. Such a connection serving as an anchor and impeding the growth and elongation of the primitive alimentary tract, results in the retention of the derivatives at an abnormal level, and in consequence interferes with the normal development of the diaphragm.

In a series of ingenious sketches Budde (1912) attempted to indicate the relationship existing between such a connection and the descent of the diaphragm. After remarking that the diaphragmatic anlage initially lies at the level of the fourth cervical vertebra, and later descends to its final position, he indicated that a "neurenteric" connection occurring above the initial level of the diaphragm cannot interfere with its descent, while one occurring within the range of its descent must necessarily interfere with its normal developmental process. In addition he tried to depict in these some of the secondary changes occurring during the later stages of foetal growth. For example he indicates how an
oesophageal connection with the nervous system may be elongated as a result of growth, thereby explaining the elongated "neurenteric" strands and diverticula sometimes met with in these cases.

In the light of this, it is interesting that Bell (1923) in his review of a series of cases showing both an anterior and posterior spinal defect, saw fit to classify both them and the associated abnormalities, according to whether they were supra- or infra-diaphragmatic in position.

Although various attempts have been made to classify such cases, the scheme suggested by Feller and Sternberg in 1928 appears to be the most satisfactory. Their grouping of cases showing a "vertebral cleft" - anterior and posterior spina bifida - is based on the relationship of the axial structures of the body, i.e. central nervous system, vertebral column and alimentary tract, to one another (Fig.13).

In their first group they include those cases in which the central nervous system has remained open, and the alimentary tract passes back through the
vertebral cleft to open on a field lying in the centre of the modified medullary plate or area medullo-vasculosa. The cases of Gruber (ii), Lehmann-Pacius, Lucksch (iii), Morel-Gross, Rembe, Schlippe and Stoltzenberg belong to this group.

In the second group of their classification the intestinal tract passes back through the cleft vertebral column. The neural tube has closed to form a spinal cord, but this divides into two in the region of the cleft, thereby surrounding, as it were, the intestinal field visible on the back of the subject.

Adelmann in 1920 described a calf in which there was a cleft involving the entire lumbar region. The large intestine had herniated dorsally, forming an entodermal field on either side of which

*In certain instances a writer has described several cases. The small numerals indicate which of his cases is being referred to.
the divided spinal cord passed caudally. His case, as well as the case forming the subject of this paper, belong to this group. The only other case which could possibly be included within this group was that described by Lucksch (i) in 1903.

The third group consists of those cases in which the central nervous system has remained open, as in group I, but in which the alimentary tract has closed. This group can be divided into two. In the first subdivision, we have those cases in which the central nervous system and alimentary tract are connected either by a strand or diverticulum, which traverses the vertebral cleft and contains tissue of ecto- and ento-dermal origin. The cases of Gruber (i), Risel (i) and (iii), and possibly the first of those described by Veraguth, belong to this subdivision. Those cases in which no such connection exists, or at least has not been specifically mentioned, constitute the second subdivision of this group. It includes the cases of Lucksch (ii), Gruber (iii), Spanner (i), Stüizer and Gaddi.
In the fourth group, both the central nervous system and alimentary tract are closed. Like the third group it is divisible into two. Here the first subdivision consists of cases in which a strand or diverticulum connects the central nervous system and alimentary tract through the medium of the vertebral cleft. It includes the cases described by Risel (ii), Bell, Budde, Feller and Sternberg (i), and perhaps those of Rindfleisch and Muscatello.

The second subdivision of group four corresponds to that of group III, in that it encompasses those cases in which no connection between the central nervous system and alimentary tract was evident, although the two are closely related in the region of the cleft. Within it fall the cases of Dammann, Spanner (ii), and Feller and Sternberg (ii).

Korff in 1937 described an interesting case in which there was a vertebral cleft extending from the fifth thoracic vertebra down to the first lumbar. The medullary plate had remained open and was cleft in two; within this fissure lay an S shaped loop of
intestine, which was blind at either end, and closely akin to the ileum in structure. A diverticulum arising from the ileum, close to the ileo-caecal valve, was directed toward the vertebral cleft, although no strand connected the two.

At first sight this unique case would appear to require a separate and additional group, but it does, as he points out, fall within the scope of group III, and at the same time represents a link in the development of groups I and III. Its interest lies in the fact that Korff regarded it as proving Feller and Sternberg's theory concerning the development of this abnormality, and it will be referred to again later.

Figure 13 has been reproduced from Feller and Sternberg's work. It is an interesting diagram in that it serves not only to illustrate the groups mentioned above, but also to indicate the manner in which they are supposed to have arisen from one fundamental type or form. It will be noticed that it includes Korff's case, who added it to the original
schema in order to show the relationship existing between his groups I and III. The disposition and relationship of the axial structures of the body to one another at the site of the defect is immediately apparent.

The first diagram represents group I, and shows the alimentary tract opening on the centre of the area medullo-vasculosa or modified medullary plate. The closure of the two halves of the medullary plate on either side of the intestinal opening results in group II, as depicted in diagram two, and accounts for the intestinal field in such cases being surrounded by a cleft spinal cord.

Group III, as represented by the diagram of that number, is seen to arise from group I as a result of the closure of the dorsal opening of the alimentary tract. The original connection with the medullary plate persists either in the form of a strand or diverticulum, as suggested by the dotted line, while the medullary plate continues to remain open, later becoming the area medullo-vasculosa.
Normal closure of the medullary plate is seen to account for group IV, in which only a vertebral cleft remains, the original connection between alimentary tract and medullary plate again persisting either as a strand or diverticulum, as indicated by the dotted line.

From this alone it will be apparent that Feller and Sternberg believe that the condition of the axial structures in hitherto recorded cases is explainable on the assumption that initially an open connection, embraced on either side by the halves of the cleft notochord and primitive vertebral column, unites the nervous system and alimentary tract. They further believe that the various forms encountered are due to the fact that although this peculiar connection persists in group I, in the other groups it undergoes some modification, during the course of development, with a greater or lesser degree of repair of the structures involved, so that eventually only a cleft notochord, and consequently cleft vertebral column may remain.
On the basis of these premises, they have sought to explain certain other malformations of the vertebral column and spinal cord - diagrams Va, Vb, and Vc - which appear to be more or less closely related to the groups under discussion.

In essence although they appear to be cases of posterior spina bifida with a greater or lesser degree of disturbance of the spinal cord, they show signs of a former cleft, as indicated by the imperfect fusion of the halves of the vertebral column. It will be appreciated that they regard their origin as due to a continuance of the reparative processes mentioned above, with but little of the original state of affairs persisting.

Apart from affording a satisfactory classification of such cases of anterior and posterior spina bifida as have been described, and a plausible explanation of the factors associated with their ultimate form, their schema introduces a new concept - the appreciation that a complete examination of a case is only achieved when a histological examination of the
vertebral column has been carried out. For they point out that the discovery of a cleft notochord in any of the malformations considered, must be regarded, apart from other factors, as a continuum of the original relationships of the axial structures, and consequent proof of their hypothesis.

(ii) **Embryology**

Owing to the greater frequency of posterior spina bifida, more attention has not unnaturally been paid to the etiology of that condition. Numerous theories have been advanced as to the causative influences involved in its production, but the majority, if not now of mere historical interest, appear to have little or no applicability to the anterior and combined forms of spina bifida. These forms have, as regards this aspect, received but scant attention in our own language. That they have not altogether been free of controversy may be gained from reading Gruber's discussion (1926) of their formal genesis, for therein he outlines the views of the earlier German workers.

Hertwig's classical essay on "Urmund und Spina..."
Bifida" which appeared in 1892, profoundly affected views pertaining to the origin of abnormalities of this type, for the chemical agents which he employed appeared to delay the closure of the blastopore in the frog's eggs on which he experimented, and gave him a series of "ring" embryos which bore a certain similarity to these cases. In them the primitive alimentary tract opened upon the medullary plate, and was accordingly surrounded on either side by the halves of the medullary plate and notochord.

He ascribed these malformations to a persistence of the blastopore and concluded that the closure of the blastopore in the frog was occasioned by a caudally directed fusion of its lateral lips. He furthermore saw therein a proof of the concrescence theory promulgated by His, and regarded a disturbance of this process as the factor underlying the formal genesis of a vertebral cleft.

This work attracted a great deal of attention, and many accordingly subscribed to Herwig's view that a vertebral cleft was due to an incomplete fusion of the
lateral blastoporic lips whereby the alimentary tract remains connected with the open medullary groove. For example Korff states that Grosser regarded such cases as would fall within Feller and Sternberg's groups I and II as proof of this fusion or concrescence theory, and Ebel mentions that Budde employed the case of Morel-Gross, in which the stomach communicated with the exterior through a complete vertebral cleft (group I), as proof that Hertwig's observations on amphibian embryos were applicable to the human embryo.

It is of interest that Adelmann, whose case occurred in a calf, and which, as already pointed out, closely resembles the case now under consideration, felt that the defect in his case had arisen early in the development of the individual, and was essentially the same as the spina bifida or "ring" embryos produced by Hertwig.

He pointed out that although there is no evidence that gastrulation is accompanied by blastoporic lips in the calf, that the primitive streak and blastoporic lips might be regarded as homologous,
in that both give rise to spinal cord, notochord, and mesoderm. And further, that the failure of the blastoporic lips (primitive streak) to approximate, would, differentiation not being retarded, result in an opening bounded by material which becomes spinal cord, notochord, mesoderm, and perhaps a small amount of entoderm. The latter, he deemed, would in any case adhere to the edges of the opening thereby forming a passageway into the primitive digestive cavity which might or might not coincide with the potential neurenteric canal. He interpreted the dorsal herniation of the mucous membrane in his case as due to the ventral wall of the intestine pushing up through the gap in the dorsal wall of the intestine at the primitive streak.

Both Feller and Sternberg, as well as Korff, whose papers constitute the most recent work on the subject, regarded the fusion theory as inapplicable to these abnormalities in the human. Their reason for this stand is embraced in Korff's sentence - "Doch kann diese Ansicht kaum eine Erklärung für die Entstehung dieser menschlichen Fehlbildungen geben, da bei
Amnioten auf keiner Entwicklungsstufe ein offener Urmund vorhanden ist, und auch im Bereich des Primitivstreifens keine Verwachungsvorgänge, sondern Massenverschiebungen stattfinden (Gräper, Kopsch, Wetzel)

In contrast with his earlier views, Dudde later (1926) suggested that the appearance of a vertebral cleft, instead of being regarded in terms of the blastopore or canalis neurentericus, was to be considered as due to an unusually long persistence of the primitive streak. The primitive streak serving then as an abnormal connection between entoderm, and ectoderm, which later undergoes secondary penetration, and so forms a dorsal opening in the alimentary tract.

This concept was likewise rejected by Beller and Sternberg, on the grounds that in the region of the primitive streak there is only a connection between ectoderm and mesoderm. Seeking then to explain the "neurenteric" connections found in many of these cases, and apparently realising that any theory which is to satisfactorily explain these abnormalities
must take cognisance of all three germinal layers, they developed a theory in connection with the primitive knot, on the basis that the three germinal layers are only connected at that point.

They suggest that, while the primitive knot migrates caudally, and gives rise to the notochord and portions of the medullary plate, for some reason or other, a portion of the undifferentiated cell mass of the primitive knot might persist in the midline. Thus it would form a cell rest, or primary connection between the three germinal layers, on either side of which the notochord would gradually develop, thereby taking up a cleft form.

It was supposed that after this as the cleft notochord interferes with the normal fusion of the sclerotomes about it, the vertebral centra in that region are laid down on either side of it in two separate halves, so determining the vertebral cleft. While the subsequent behaviour of the primitive knot cell rest itself was deemed responsible for the different types that are met with.

For instance they suggested that the formation
of an opening in the cell rest resulted in the alimentary tract opening dorsally, and subsequently led to a protrusion of the alimentary tract through the cleft spine, as in cases belonging to groups I and II. This however on the basis that the only point of difference in the development of these, was that the medullary plate either had or had not closed on either side of the opening, so that the alimentary tract either opened upon the back or upon a modified medullary plate or area medullo-vasculosa.

Then again, the failure of such an opening to appear in the cell rest, or the degeneration of the cell rest in the later stages of development, was regarded as the explanation of groups III and IV and their subdivisions. In that in those cases there either is or is not a strand, composed of cells of ectodermal and entodermal origin, passing through the vertebral cleft between the alimentary tract and the spinal cord or area medullo-vasculosa.

In support of their theory they pointed out that Jablonowski had observed abnormally situated portions of the primitive knot in hen embryos, and remarked on Kolmer's discovery (1926) of a cat embryo of
fourteen somites in which the notochord was double in the region of its middle third. Korff felt that his case confirmed this theory, in that he was of the opinion that it could only be explained by the differentiation of a cell rest or cell complex, composed of all three germinal layers, which connected the neural and intestinal tubes. He also appeared to think that it indicated that the cell rest or connection between the central nervous system and alimentary tract might, instead of degenerating, or persisting in the form of a strand or diverticulum, continue to differentiate and form an organ!

Suggested Theory

Their whole theory appears as yet to rest upon somewhat slender evidence. It does however not only account for all cases that have been described, but also takes cognisance of their associated abnormalities, and they must be credited with focussing attention upon the cleft condition of the notochord. Until more evidence is forthcoming, it would seem safer to regard this peculiar state of the notochord as the starting
point of these abnormalities. That it cannot be lightly discredited is indicated by the fact that Budde discovered a forked notochord in the region of the cleft in his case, and that Feller and Sternberg discovered notochordal rests in either half of the vertebral column in both of their cases. It has already been seen that a similar state of affairs existed in this case.

Consideration of Kolmer's illustrations (Fig. 14) will suggest that adhesion of entoderm and ectoderm between the two portions of the notochord might reasonably account for groups I and II, provided it underwent penetration, and at the same time account, according to the degree of its persistence, for groups III and IV and their subdivisions; the ultimate form of all being dependent on the behaviour of the medullary plate.

That such an adhesion should contain cells of both these germinal layers, and produce either a "neurenteric" strand, or traction diverticula at one or both points of attachment, seems but a logical
conclusion. As for the fistulous openings observed in some of these cases, it is possible that they owe their presence to a foetal necrosis which brings about the penetration of such an adhesion.

And further, if the notochord does adopt the form seen in Kolmer's illustration, it is not difficult to conceive of the vertebrae in this region being laid down in two halves. Whereupon the possibility of their developing side by side and even partially fusing (groups Va, Vb, and Vc), or being prevented from so doing by reason of an adhesion, or penetration, with or without herniation of some part of the alimentary tract, seems both a logical sequence and explanation of the varying regional vertebral disturbance which characterises these cases.

As to the more remote visceral displacements, they must of necessity be due to interference with the normal growth processes, being qualified both by the gravity and site of the vertebral cleft, as well as by the degree of involvement of the alimentary tract with the cleft. It is not difficult to appreciate that
these factors might induce innumerable types of visceral disturbance, either by reason of their ability to impede the growth and elongation of the primitive alimentary tract, with the retention of its derivatives at an abnormal level, or to interfere with its normal processes of rotation. And it may well be that the circulatory disturbances which accompany these account for many of the peculiarities met with, such as the varying degrees of intestinal atresia, which incidentally is a common associate of errors of rotation.

With regard to the above mentioned possibility of the vertebral halves, in the presence of such a notochordal defect, subsequently undergoing a degree of fusion, it is not unlikely that we have therein, apart from an adequate explanation of such cases as fall within the limits of groups Va, Vb, and Vc, an explanation of the osseous and cartilaginous processes sometimes arising from the posterior surface of the vertebral bodies in cases of "posterior spina bifida". These processes project into the spinal canal, and dividing it more or less completely into two
compartments, are attended by a varying degree of division of the spinal cord. The literature concerning them has been well surveyed by Ballantyne (1904), Greig (1929) and Hamby (1936).

Humphry (1886) was of the opinion that the processes had a morphological significance, and Clelland (1889) felt that their shapeliness, and in certain instances their sequence and separate centres of ossification, supported that view. The latter came to the interesting conclusion that they were associated with a partial fission of the embryo.

The Clinical Society of London (1885) in their report on spina bifida apparently recognised an association of the processes with anterior spina bifida, for they say of such a projection that connected the arch of the 10th dorsal vertebra to the posterior surface of the lower dorsal vertebrae - "It is interesting to observe that there is a duplication of the centra of the bodies of the last two dorsal vertebrae with which the intercalated element (projection) is connected. This appears to be related to the condition
in which a portion of the vertebral column is completely cleft".

Greig in a case, which as will be seen later, appears to have been one of combined spina bifida with reparative changes of the anterior defect, also encountered one of these processes. Of it he said - "To the writer it appears that the bifocal ossification of the vertebral centra induces a tendency which is seldom given effect to; but a step in this direction is the formation of a dorsal prolongation from one or both of the contiguous hemi-centra at their mesial extremities. Two more or less complete tunnels are produced in each of which lies a segment of the locally duplicated spinal cord".

The feasibility of these processes being related to such cases as fall within Feller and Sternbergs five groups, appears to be strengthened both by these remarks, and by the fact that the appearance of a dorsal prolongation or process from the medial margin of one or both of the contiguous hemi-vertebrae in cases such as are illustrated by diagrams Va, and
Vb, would explain why in some cases there is a portion of the spinal cord on either side of the process, and why in other cases there is but a mere groove in the spinal cord.

The more frequent association of these processes with a bifid state of the spinal cord may be due to the mesoderm, prior to the eventual fusion of the vertebral halves, seeking not only to grow about the portion of notochord related to its side, but also striving to provide a protective covering for the spinal cord of its side. The presence or absence of a "neur-enteric" connection or adhesion, and the degree of differentiation attained by the vertebral halves prior to fusion, might well be additional factors influencing their appearance.

An interesting illustration of the relationship between these processes and the above mentioned groups is to be had by comparing the present case with those of Cooperstock and Greig. In the last of these, there was, in the thoracic region, a bony process invading the spinal canal, and as Greig points out, probably a bifi-
dity of the spinal cord. The vertebrae immediately above the process were cleft, but showed no sign of separation. In the case described by Cooperstock and Elzinga there was a marked cleft in the thoraco-lumbar region, and an attempt to form a double neural arch. The vertebral halves were separated somewhat from one another. There was no mention of any visceral displacement, but in view of what was said of the X-ray appearances of the neural arches, it is probable that there was a duplicated spinal cord. As regards the present case it has already been seen that there was a duplicated cord, hernial loop occupying the cleft, and a marked separation of the vertebral halves. And further, that there were a series of accessory half arches arising from the medial side of the left hemi-vertebrae.

Of these it will be seen that while the last falls into group II, Greig's corresponds to group Va, if a bony process be introduced between the two cords, and that Cooperstock's case must occupy an intermediate position between these two groups. This might be expressed differently, by pointing out that had
been no hernial loop, the accessory half arches of the present case would have given a picture not unlike that presented by Cooperstock's case, and if fusion of the hemi-vertebrae had taken place the accessory half arches would have then taken up a position corresponding to the bony process found in Greig's case.

Application to Present Case

Turning then to the present case, the vertebral cleft and its attendant anomalies must for want of really convincing evidence as to the manner in which the notochordal disturbance is induced, be regarded as having been initiated from a cleft condition of that structure. Thereafter, it would appear that this defect of the notochord, and perhaps the early existence of an axial ecto-entodermal connection or adhesion, interfered with the fusion of the sclerotomes about it, with the result that a series of hemi-vertebrae were laid down on either side of it.

It must not however be supposed that the hemi-vertebrae were first laid down and then followed by a herniation of the hindgut, for how then is one to
reconcile the fact that each division of the spinal
cord possessed a central canal, and was embraced by
the hemi-vertebrae of its side? Rather it would seem
that the laying down of the hemi-vertebrae, division of
the spinal cord, and dorsal penetration of the gut
had proceeded within a short space of one another, if
not together.

Normally the rising up of the medullary folds is
accompanied by the appearance of an important develop-
mental process - the cleavage or segmentation of the
mesoderm on each side of the medullary folds into
segments or somites, and the closing off of the neural
tube towards the end of the 4th week, when some thirty
somites have been laid down. From this it will be seen
that the processes of mesodermal segmentation and
closure of the medullary plate march together. Then
this, and the fact that the skeletal elements of the
somites, or sclerotomes, usually surround the notochord
and neural tube simultaneously, suggest, since each
division of the spinal cord possessed a central canal
and was embraced by its hemi-vertebrae, that at the
same time as the notochordal cleft was established, or shortly after, there was formed an axial ento-
ectodermal connection which underwent penetration, with the resultant accompaniment of the division of the medullary plate, and the growth of the sclerotomes about those divisions as they underwent closure. All of which, together with the knowledge that the medullary folds rise up towards the end of the 3rd week, enables one to suggest that the essential facts of this case were being laid down during this phase of development.

With regard to the vertebral column and its cleft, it has already been pointed out that the general conclusion is that there has been a suppression of half a segment, with the addition of a half lower down. That irregular segmentation should have accompanied such apparently conjoint disturbances of the notochord, primitive alimentary tract, and neural tube, is not surprising, especially in the face of the extraordinary disposition of the nerves in this region.

As for the arrangement of the nerves, it was unfortunate that the bifurcated cord was not well enough preserved for histological study. It will be
recalled that while the right and left divisions of the spinal cord both gave off nerves from their lateral aspects, that the latter division also gave off a medial set of nerves, whose roots embraced it as they made their way medially toward the cleft, although certain of them eventually passed to join its lateral set. To account for the arrangement of the nerves in this region is difficult, and all that can be said is that there appears to have been an unequal division of the medullary plate, rotation of the left division of the spinal cord, and possibly a displacement of developing nerve roots by the growing hernial loop. Other developmental aspects, such as the abnormal length of the spinal cord, have already been dealt with, but it may be mentioned here that the form of the accessory half arches on the medial aspect of the left hemi-vertebrae is possibly associated with the medial set of nerves arising from the left division of the cord.

When the disposition of the abdominal viscera in this case is considered, it is apparent that there has been a derangement of intestinal rotation which must
be ascribed to the establishment of an axial ento-ectodermal adhesion or connection. Since this connection, now represented by the fistula or accessory anus at the apex of the hernial loop, involves a colonic loop, it is obvious that at an earlier date it may have involved either the mid or hind-gut, in that both of these contribute to the formation of the colon. The suppression of the inferior mesenteric artery suggests that the connection involved the hind-gut. The fact that the superior mesenteric artery supplied both limbs of the hernial loop need raise no objection to this view, for the presence of the caecum upon the entering limb stamps it as a mid-gut derivative which should so receive its blood supply, while the abrupt termination of the returning limb in the rectum, suggests that the latter could not be other than a hind-gut derivative, which is receiving a compensatory blood-supply from the superior mesenteric artery in the absence of its accepted source of supply.

Prior to rotation, the distal portion of the fore-gut, represented by the duodenum at the level of the biliary papilla, becomes a fixed and constant point, a little to the right of the midline, from which the
mid-gut depends. And the hind-gut, occupying the midline, is "hitched up" by a retention band, which induces a flexure - colic angle - between it and the midgut. Here it would seem that the ento-ectodermal connection may have served as a retention band, replacing that which normally produces the colic angle, but differing from it in function. Instead of imparting a relatively higher position to the angle, it must have eventually been forced by the growth of the structures about it, to migrate caudally, thus accounting for the present position of the colic angle in this case, which is now represented by the limbs of the hernial loop.

Resorting to Dott's practice (1923) of regarding the rotation of the gut as occurring in three stages, it would appear that this connection could have in no way interfered with the first stage of rotation, in which the ends of the mid-gut loop undergo an anti-clockwise rotation and come to lie side by side.

Dott states that the chief factor which determines the second and essential stage of rotation is the sequence in which the intestine is returned from the
umbilical cord to the abdomen. He regards the factors causing a disorderly sequence of return as such as would render the small intestine more difficult of reduction or the caecum easier, or the umbilical orifice so large that they could be reduced with equal facility.

Here the restraining effect of the connection must have brought about an unduly early return of the caecum and caused it to take up its position alongside the hind-gut (returning limb of the hernial loop), with the result that the rest of the mid-gut loop (small intestine) disposed itself, owing to its inability to displace the anchored hind-gut to the left, both in front of and to the right of the colic angle (limbs of the hernial loop).

Some support for the view that the caecum returned unduly early, is provided by the fact that the small intestine was somewhat short, which suggests that the mid-gut loop was unable to attain its full growth in the presence of the rapidly enlarging liver. The paucity of arterial arcades in the mesentery is possibly yet another factor responsible for its
shortness. It measured 137.5 cms. (55 ins) from the pyloro-duodenal junction to the ileo-caecal junction. In the new-born the small intestine varies from 300 to 350 cms. in length (Scammon quoted by Scott 1933) and averages 380.9 cms. between the 3rd and 6th months of post-natal life. However it must be appreciated that any argument based on post-mortem intestinal measurements is highly controversial.

It cannot be doubted that the ento-ectodermal connection was responsible for the non-rotation of the mid-gut loop. The disposition of the intestine shows many features associated with a disturbance of the second stage of rotation. The duodenum descends from its fixed upper part (foregut), down the right side of the superior mesenteric artery, and is free; the small intestine lies chiefly to the right of the midline; the terminal ileum crosses the midline to reach a left iliac caecum. The caecum is mobile, and is reversed, that is, the ileum enters it from the right. Instead however of the ascending colon passing upward behind the greater curvature of the stomach, and
then being connected by a narrow loop of transverse colon with a normally placed splenic flexure, from which the descending colon pursues its usual course, it has been seen that the ascending colon made its way into the midline fossa to communicate with the accessory anus.

Figure 15 shows how remarkably the disposition of the small and large intestine resembles the condition of the alimentary tract in about the 8th week. Owing to the ento-ectodermal connection having presumably involved the hindgut, and produced a colic angle, the angle which is there depicted need only be visualised as being at a lower level, as already explained, to more or less faithfully reproduce the conditions as found in this case. In short, the midgut loop had not rotated, and these relations have been maintained, including the embryonic mid-line position of the hind-gut.

The herniation associated with the colonic loop is to be explained as due to the dorsal wall of the intestine gradually growing and pushing its way through the gap in the dorsal wall provided by the establishment of an aperture in the ento-ectodermal connection or adhesion. To be more precise there
has been a prolapse of that part of the dorsal wall forming the cephalic (superior) wall of the gap. If the mucous membrane of the dorsal wall of the returning limb be traced to the accessory anus (Fig. 6), it will be seen to become continuous with the skin at the lower side of this anus, that is, at point B. On the other hand, the mucous membrane of the entering limb continues outwards and upwards, thereby forming the "mucous pad" on the back, and terminates at point A where it joins the skin of the back. Owing to the alternative designations which have previously been applied to the limbs of the hernial loop, it may not perhaps be out of place to state that the dorsal herniation visible on the back of the child possibly represents in part the absent transverse colon.

Other points may now be considered. With regard to the great omentum, it is clear that it must have continued to elongate, and in the absence of the transverse colon, descended and fused - on the right with the mesentery, on the left with the anterolateral aspect and lower pole of the left kidney, and
posteriorly with the anterior surface of the same
kidney, with the result that its lower border extends
between the mesentery and kidney.

As regards the low position of the left kidney, it
seems that as the kidney develops in the sacral
region and receives its blood supply from the lower
part of the aorta or iliac arteries, and then ascends
to receive a second and permanent blood supply from
the aorta higher up, that here the local disturbance,
and displacement of the aorta to the right, both inter-
fered with its ascent and robbed it of its chance to
obtain vessels from the aorta, with the result that it
has remained at a low level and retained its source of
supply from the left iliac artery.

(iii) **HOMENCLATURE**

Feller and Sternberg refer to these cases as
examples of "vordere Wirbelsäulenspalte (Rachischisis
anterior)", but more commonly employ the term
"Wirbelkörperspalte", and imply the presence of an
arch defect. Korff speaks of them as cases of
"vollstänigter Wirbelspalte (Rachischisis anterior"
and posterior)".

In the literature of these cases the term rhachischisis is used in many ways. Sometimes it is used as a synonym for spina bifida, sometimes as a term implying an extensive anterior or posterior defect, spine and again in the literal sense of a cleft in two. When used in the first two senses it may be qualified by terms such as anterior and posterior, the extent of the defect, as indicated in the text, alone serving to indicate the manner in which it is being employed. When used in the literal sense, it is often merely qualified by the region affected e.g. rhachischisis cervico-dorsalis. Occasionally cases are simply referred to as cases of spina bifida, again using the term in its literal sense.

Speaking of spina bifida in general, there has long been a tendency to employ the term rhachischisis either as a synonym for spina bifida, or as a term indicating an extensive defect, with the reservation of the term spina bifida for a localised imperfection. Obviously the arbitrary use of the term, as in the
latter case, is unsound, and therefore it is suggested that if it must be used at all, it should be employed as a synonym, carrying no special value. To utilise the terms rhachischisis and spina bifida in their literal sense in the description of these cases, is likewise unsatisfactory, for although etymologically sound, the practice is offset by the fact that both of these terms have been widely used in the description of other types of spina bifida. Indeed the greater frequency of posterior spina bifida has resulted, at any rate in clinical parlance, in the term "spina bifida" coming to imply a posterior spinal defect.

As Ballantyne pointed out, not a little difficulty was introduced into the subject of posterior spina bifida by want of care in the naming of varieties, and the hasty identification of a given case with one described by another writer in another language. It would seem then that if the condition of anterior spina bifida is to escape this, it must be appreciated that while some employ the term to denote a defect of the vertebral body alone, others include under it
even such cases as show a defect of both the vertebral body and arch. Now it will be seen that the term should only be employed when the vertebral body alone is affected, and that when there is a concomitant posterior defect attention should be drawn to it by speaking of the case as one of anterior and posterior spina bifida, or as it might well be termed, combined spina bifida.

In a paper entitled "Anterior Spina Bifida and its Relation to a Persistence of the Neuenteric Canal", Bell states that true anterior spina bifida is only found in the sacral region, and mentions that posterior spina bifida usually occurs in association with anterior spina bifida of the cervical and thoracic regions. In the face of this however, he unfortunately referred to a series of cases collected from the literature, which showed both an anterior and posterior spinal defect - not limited to the sacral region - as cases of anterior spina bifida.

That it is not desirable to employ the term anterior spina bifida for such cases is reflected in the fact that our textbooks more often than not, fail to
appreciate that when there is an anterior spina bifida in either the cervical, thoracic or lumbar regions, there is frequently a concomitant posterior defect, and that accordingly it is always necessary to mention the state of the vertebral arches. In addition they do not seem to have recognised that when these regions are affected, there is often not only a combined anterior and posterior defect, but also a series of associated abnormalities whose character is not only different, but generally more severe, than those found attending an anterior spina bifida of the sacral region.

In a number of clinical reports it is extremely difficult to assess the case in that no mention has been made of the state of the vertebral arches. Due regard should therefore be paid to the terms suggested above, in the hope that it will result in the vertebral body and arch being accorded equal attention, with consequent clarity and accuracy of description.

The only objection which may be raised to the adoption of these terms is that it may be said that the posterior defect accompanying an anterior spina
bifida is not always comparable to that which we normally associate with a posterior spina bifida, and that in fact the vertebral arches may be well formed, but unable to unite with their fellows owing to the position which their vertebral hemi-centra have taken up. In short, the posterior defect is sometimes merely a secondary factor dependent upon the position adopted by the vertebral hemi-centra. In this case for example, although there was a posterior defect, it was flanked on either side by a ridge composed of fused, but well formed half arches, showing none of the laminar insufficiency usually associated with a posterior spina bifida.

Be this as it may, regardless of whether the half arches are well formed or not, it seems more important to indicate the presence of both an anterior and posterior defect, by referring to such a case as one of anterior and posterior spina bifida, or combined spina bifida. Indeed it may be mentioned here that in these cases the anterior and posterior defect are seldom co-extensive, the latter being frequently not only
more extensive, but more of the nature of a true spina bifida.

The advisability of considering the condition of anterior spina bifida under two headings - (a) (True) Anterior spina bifida, and (b) Anterior and Posterior Spina Bifida or Combined Spina Bifida - becomes even more apparent when it is realised that such a subdivision tends to focus attention upon the vertebral arch, so warning the reader of the possibility of encountering a concomitant posterior defect, and at the same time allowing the clinician to convey to a colleague in a direct and unambiguous manner, the true state of affairs.

Such a subdivision is supported by the general features pertaining to these two types. True anterior spina bifida would appear to be a condition usually confined to the sacrum. It is characterised by anterior meningocele, and is, although rare, a well known clinical entity which often permits the subject to attain maturity. On the other hand, combined spina bifida may involve any region of the vertebral column, although it occurs more frequently in the cervical
and thoracic regions. It is often attended by marked abnormalities of the alimentary tract and nervous system, and these as a rule are of such a nature that they do not permit the subject to live. This type is thus not well known, and this fact together with the European origin of its literature possibly accounts for the inadequate attention which our textbooks usually accord to both the vertebral arch and the associated abnormalities when discussing anterior spina bifida in regions other than the sacrum.

With regard to the literature on cases showing a combined defect, there is a commendable tendency, at least among the more recent authors, to devote equal descriptive attention to both the vertebral body and arch. The nomenclature however, as has been indicated, is not very satisfactory, both by reason of the variety and the indiscriminate use of certain terms; and further, the fact that some of the terms have now come to have a somewhat different meaning in our own language, makes it seem inadvisable to adopt the nomenclature used by the foreign authors. It will
thus be apparent why it is deemed necessary to resort to simple terminology, and in the interests of accurate description and classification to discriminate between the anterior, and anterior and posterior, or combined form, of spina bifida.

(iv) **General Features**

**Sex**

It is of interest that regardless of the type, the majority of the cases have been in females. Of seventeen cases of (true) anterior spina bifida involving the sacral region, Bell discovered that all but one had occurred in adult females. He found no record of anterior spina bifida limited to the sacrum in males. In thirty-six cases of combined spina bifida, which number includes the present case, nineteen were females, six males, and in eleven instances the sex was not ascertained. Both Bell (1923) and Sharpe (1928) emphasised this sexual proclivity on the part of the female in the former type, but with regard to the latter, this aspect does not seem to have attracted any attention, although here again there appears to be a significant
Longevity

The fact that so many cases of the sacral type of anterior spina bifida attain adult life, is no doubt due to the low position of the defect, and the relatively simple character of the associated abnormalities. In comparison, the combined form, which is generally both higher in position (Fig.12) and attended by severer abnormalities, seldom permits its subjects to survive long after birth.

While the majority of cases showing a combined defect are but of teratological interest, quite a number of cases have survived for some time. De F. Willard's case (1904) was two months old when he brought it before the Philadelphia Academy of Surgery, and the present case, as has been seen, survived for four months. Bell mentions two cases, of which one lived for sixteen and the other eighteen months. Budde's case lived for a year and nine months, while another, that of Oehlicker (1909), had attained the age of six and a half years before she was first brought for
examination. In the latter case, it was a spinal curvature that led to an X-ray examination and the discovery of an "anterior spina bifida" in the region of the cervico-dorsal junction. It falls however into the category of a combined spina bifida in that there was an attendant posterior defect, apparently not unlike that found in this case.

The recently described case of Cooperstock and Elzinga, which as already pointed out, falls within this category, was that of a young girl who had attained her seventh year at the time of their report. Like the previous case, a spinal curvature indicated an X-ray, and of this it was said "there is no question that almost complete lack of fusion of the two halves of the entire vertebral mass exists in the thoracic eighth to the twelfth inclusive, and the entire group of lumbar vertebrae. In addition there is a spina bifida occulta of fairly marked degree in the entire sacrum". Although more extensive, the vertebral defect in their case appears to have a number of features in common with this case. Their mention of
a developmental attempt at the formation of a double neural arch recalls the state of affairs on the left side of the vertebral cleft in this specimen, and suggests that their case too has a duplication of the spinal cord.

Greig's case, also previously referred to, was a female congenital idiot aged eight. Of it, he said "there is a congenital scoliosis, an extensive posterior rachischisis and probably there was duplication or bifidity of the spinal cord". This case too was probably a combined spina bifida, but one in which the anterior defect had undergone reparative changes. Evidence of anterior spina bifida was provided by several malformed and cleft vertebral centra lying side by side, and a buttress of bone dividing the vertebral canal into two. It was the latter structure, in the absence of the spinal cord, that led him to suggest that a bifid state of the cord had originally existed.

The absence of any gross involvement of the alimentary tract and nervous system doubtless accounts for the greater age attained by cases such as those.
of Oshlicker and Cooperstock. It is possible of course that there may have been a duplicated spinal cord in both of these cases, but it is recorded that such a condition has been discovered in quite a number of apparently healthy subjects.

Regional Incidence of the Defect and Degree of Alimentary Involvement

Some reference has already been made to the regions of the vertebral column commonly affected in such cases, as well as to the portions of the alimentary tract usually involved in the cleft. In order however to obtain a clearer picture of these facts, and as no graphic representation had hitherto been compiled, the author deemed it necessary to construct the graph indicated in Fig.12.

The extent of the vertebral cleft in each case is shown as well as the portion of the alimentary tract related to the cleft, but owing to the fact that in certain instances the original work was not available, and that in others the summarised accounts provided by Feller and Sternberg, Gruber, and Bell, proved...
inadequate, these facts were, for want of accurate information, indicated by means of dotted lines. With regard to that portion of the chart which deals with the vertebral column, it should be noted that it only serves to indicate the number of cleft vertebral bodies, want of adequate data unfortunately rendering it impossible to plot the extent of the posterior defect in each case. These cases were first plotted in the order in which they were encountered in the literature, but since they appeared to fall within several groups, they were arranged in their present form.

It will be noticed that in the majority of the cases the cleft commences at the cephalic end of the vertebral column and that then it either extends caudally throughout the whole length of the vertebral column, or else involves the cervico-dorsal and cervical regions to a varying degree. As for the rest, it is interesting to note, apart from the fact that the cleft commences at some distance from the cephalic end of the column, that in remarkably few cases of this type has the cleft been found involving
the lumbar region of the column. In contrast it might be mentioned that posterior spine bifida is per se commonest in the lumbo-sacral region.

As will be seen from the chart, the majority of the cases were attended by a cranial defect, which generally took the form of an anencephaly (acrania) or cerebral hernia. Ballantyne has already pointed out that there is no clear line of division between acrania and the different varieties of cerebral hernia (encephalocele, meningocele etc.) and in his discussion of the somewhat complicated nomenclature of the condition of anencephaly he has incorporated therein much which throws light upon the terms, e.g. cranioschisis, crania bifida, used by certain of the writers to describe the nature of the cranial defect in these cases.

With regard to the association between the alimentary tract and the vertebral cleft, no attempt has been made to indicate whether it was the proximal or distal portion of an alimentary segment, e.g. stomach, small intestine, that had been involved, in
that it appeared more important to visualise such an association broadly so that it could be more readily interpreted in terms of the three portions of the embryonic gut. Where the region affected was a junctional one e.g. gastro-oesophageal, this has been suggested.

In conformity with Feller and Sternberg's classification, it will be appreciated that the portion of the alimentary tract indicated did not in every case open or present itself dorsally, but in a few cases simply lay within the cleft, no diverticulum or connecting strand having been found. In a number of cases again there was either a diverticulum (D), or diverticulum terminating in a strand (D+S), or merely a strand (S) passing toward the vertebral cleft. In most of the cases in which these existed, they were connected with the central nervous system and justified their description as "neurenteric connections", but in a few instances they lost themselves in the region of the cleft.

As regards those parts of the alimentary tract most commonly affected, it will be noticed that they are derivatives of the fore and proximal mid-gut.
This is not altogether surprising however, when it is considered that the vertebral cleft in the majority of the cases involves the cervical and cervico-dorsal regions.

Turning then to the vertebral cleft, the fact that in the majority of these cases the cleft commences at the cephalic end of the vertebral column, and extends caudally for a varying distance, seems to suggest that not only is the defect one which is initiated at an early stage of development, but also that it is one which is overcome with varying rapidity as growth proceeds. As for those cases in which the cleft commences at some distance from the cephalic end of the column, we seem to have an indication that the processes bringing about the defect may be initiated at a later date and yet be overcome. From what has previously been said, it will be appreciated that these statements should be considered in terms of the notochord etc., in that we are dealing with a condition which must necessarily manifest itself in the presomite stage of development.
In many respects, all would appear to be modified structural expressions of a common initiator, that, to follow Stockard (1921), merely selected different moments at which to bring about a developmental interruption. As Stockard has pointed out, the sensitive period or "critical moment" of an organ coincides with the high activity which attends its inception, so that if we may justifiably draw a close relationship between this condition and a disturbance of the notochord, its greater incidence in the cephalic half of the vertebral column is only to be expected, in that the first formed and consequently most easily affected portions of the notochord would naturally constitute the anlage for that region.
VI. SUMMARY

1. The subject, a female infant, was admitted to hospital when two days old, with a red mass projecting from the lumbar region of the back. Examination proved this to be a pad of mucous membrane overhanging a small fistula. Since a catheter introduced into the fistula emerged at the anus, and a radiograph revealed a vertebral cleft in the region of the mass, it was proven that the red mass and fistula were occasioned by the large bowel opening through the cleft on to the back. The child passed faeces both via the anus and "accessory anus" or fistula. She died when 4 months old.

2. A detailed dissection of the body was carried out. This revealed that the large intestine did not pursue its usual course. Instead, it passed from a left-sided ileo-caecal junction, into a midline peritoneal fossa, communicated with the dorsal fistula, and returned via the lower part of the fossa to become continuous with the rectum. The peritoneal fossa occupied the position normally taken up by the lumbar
and upper sacral vertebrae. It was bounded by bony margins - the right and left halves of the cleft vertebral column. The cleft extended from the 1st lumbar vertebra to the 2nd sacral inclusive. The left side of the cleft contained an additional piece intercalated between the 12th thoracic vertebra and the 1st left lumbar hemi-vertebra. A series of accessory half arches lay on the medial aspect of the left hemi-vertebrae. Each half of the cleft column housed a division of the spinal cord, for the cord bifurcated at the upper end of the cleft and reunited at its lower end. The right division of the spinal cord gave off one set of nerves, the left gave off two. The large intestine, flanked by peritoneal culs-de-sac, passed back through the cleft vertebral column and between the divisions of the spinal cord before it communicated with the fistula. The manner in which the identity of the vertebral pieces was determined by correlation with the spinal nerves is explained, and an explanation is given of the form of the vertebral pieces.
3. A survey of allied cases is given, together with a description of Feller and Sternberg's classification, and the present case placed within group II of that classification. Group II hitherto contained but two cases.

4. The embryological theories of causation are discussed, with special emphasis on that of Feller and Sternberg. Accepting their theory that a cleft condition of the notochord presages a vertebral cleft, the author proffers a modification favouring an ento-ectodermal adhesion rather than a primitive knot cell-rest as the causal factor underlying the different groups. The bony and cartilaginous processes sometimes found projecting into the spinal canal in cases of "posterior spina bifida" are discussed in the light of this modification. The anomalies of the present case are investigated from an embryological viewpoint.

5. The vagaries of the existing nomenclature are considered. While this and allied cases fall within the category of anterior spina bifida, it is pointed out that they should, in order to avoid confusion with
true anterior spina bifida, be referred to as cases of anterior and posterior spina bifida. An alternative term - combined spina bifida - is suggested.

6. The regional and sexual incidence, and longevity of cases of combined spina bifida is discussed. A graphic representation of the extent of the vertebral cleft, and degree of alimentary involvement, in cases hitherto recorded, is presented.
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