

Congenital Anomalies of Female Genital Tract

Functional Classification Based on Review of 56 Personal Cases and 500 Reported Cases

JAMES P. SEMMENS, M.D., CAPTAIN (MC), U.S.N.

THE SUBJECT of congenital anomalies of the female genital tract has captured the interest of many writers on and teachers of obstetrics and gynecology during the past 300 years. In 1699 Mauriceau reported a case of uterus bicornis with a pregnancy in a rudimentary horn. Many contributions in the form of case reports, proposed systems of classification, discussions of complications

From the Departments of Obstetrics and Gynecology, U. S. Naval Hospital, Portsmouth, Va., and U. S. Naval Hospital, Naval Base, Charleston, S. C.

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of pregnancy, and even controversy concerning superfetation and suprafecundation have appeared in the literature.

Kermauner (1912) and Jarcho (1946) offered methods of classification based on anatomic findings and embryologic development. This brought some semblance of order to the reporting of subsequent cases. However, a cumbersome classification in terms of nine groups is of little benefit to the clinician, who needs a simple yardstick to evaluate the occasional patient, usually seen in the gravid state. In 1957 Jones offered a functional classification based on the capacity of the uterus, the cervix, and the vagina. Two of his groups pertained to "pure" vaginal anomalies, and four were combinations taking account of the functional capacities of the uterus, cervix, and vagina. Again, use of these categories requires more information about the patient than the clinician usually has at hand.

The primary purpose of this report is to offer a simplified classification based upon the anatomic and physiologic capacity of the uterus, employing the variations of uterine architecture, the potential space for the developing fetus, and those physiologic abnormalities which hinder normal growth and

GENITAL-TRACT ANOMALIES

development of the fetus or contribute to premature or desultory labor, or abnormal presentation.

CLASSIFICATION

The clinical (functional) classification proposed is based principally on the potential capacity of the uterine cavity and its musculature. The space available for the developing fetus, the inherent variations in the muscular physiology, the variations in uterine circulation, and possible differences in innervation have a direct effect on continuation of gestation, presentation of the fetus, onset and behavior of labor, method of delivery, and over-all fetal wastage.

If the entire functional component has been derived from a single müllerian duct and its vaginal outlet is a cervical canal of similar origin, its capacity is smaller than that of the uterus resulting from fusion of bilateral ducts and subsequent reabsorption regardless of degree. Thus, as shown in Fig. 1, the functional hemiuterus becomes Group I and includes uterus bicornis bicollis, uterus unicornis, and uterus bicornis unicollis, in all of which one horn is rudimentary. If the uterus is derived from two müllerian ducts and empties into the vagina through a cervix of bilateral müllerian origin, its capacity is as large as or larger than that of the uterus so

formed and reabsorbing normally. Clinically, the problems associated are identical, whether there is a septum or a fundal indentation. Thus, Group II is made up of the variations of uterus bicornis unicollis, namely bicornuate, septate, subseptate, and arcuate uteri (Fig. 1).

The finding in a patient of vaginal duplication should serve little more purpose than to arouse suspicion of the presence of uterine anomaly. The septum may cause dyspareunia, sterility, obstruction of menstrual flow and hematocele, or dystocia in labor and delivery. It is difficult to obtain satisfactory hystero-graphic studies with the septum intact. Therefore resection should be accomplished in the nongravid state and hystero-graphs taken thereafter. The presence of the septum at delivery represents an acceptance of complications which are avoidable.

MATERIAL

Cases

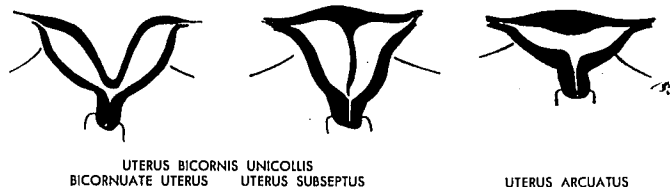
A personal series of 56 patients with congenital anomalies of the female genital tract is presented. Thirty-three patients were seen at the U. S. Naval Hospital, Portsmouth, Va. (from July 1, 1954, to June 30, 1957) and 23 at the U. S. Naval Hospital, Charleston, S. C. (from July 1, 1957 to Feb. 1, 1961).

Fig. 1. Functional classification of uterine anomalies.

GROUP I. FUNCTIONAL UTERI OF SINGLE MULLERIAN ORIGIN



GROUP II. FUNCTIONAL UTERI OF DUAL MULLERIAN ORIGIN WITH VARYING DEGREES OF FAILURE OF FUSION OR ABSORPTION OF THE MEDIAL SEPTA



Twenty-three patients had uterus bicornis unicollis, 25, uterus bicornis bicollis, 4, uterus bicornis unicollis with one rudimentary horn, 2, uterus arcuatus, 1, uterus unicornis, and 1, agenesis of the vagina and uterus.

An additional 500 complete obstetric and gynecologic histories culled from the literature were statistically analyzed in terms of the proposed functional classification. These included 202 cases of uterus bicornis unicollis, 200 of uterus bicornis bicollis, 59 of bicornis unicollis with one rudimentary horn, 25 of uterus arcuatus, and 14 of uterus unicornis.

Pregnancies

Of the 146 pregnancies in the personal series, 52 occurred in bicornis bicollis uteri, 78 in bicornis unicollis uteri, and 11 in uteri in which one horn was rudimentary, while 5 were in the only married patient with an arcuate uterus.

The literature also showed an increased number of pregnancies (576) among women with uterus bicornis unicollis, while those with complete duplication (bicornis bicollis) had 417. Sixty-three pregnancies occurred in the group with uterus arcuatus, 19 in those with uterus unicornis, and an additional 159 in patients with uterus bicornis unicollis in which one horn was rudimentary, making an over-all total of 1234 pregnancies in patients reported in the literature.

INCIDENCE

The incidence of uterine anomalies seen in the two naval hospitals was 33 births in 20,636 deliveries, or 1:625; and 33 of 59,170 outpatients had gynecologic complaints related to uterine anomalies, or 1:1800. Ninety-six per cent of all these patients had major uterine anomalies.

DIAGNOSIS

Of paramount importance in the diagnosis of uterine anomalies is clinical awareness of

their existence and of the problems most frequently associated with them. Falls,⁷⁹ Way, and others stressed notching and broadening of the uterine fundus, floating head at term in a primigravida in the absence of accepted causes of disproportion, as well as abnormal lie, recurring breech presentations, trapped or retained placenta, and prolonged third stage of labor. Unfortunately these occur too infrequently, and many uncomplicated deliveries delay clinical investigation of the patient with uterine anomaly until she is seen in the climacteric years with menorrhagia. Even the habitual aborter experiences considerable fetal wastage before hystero-grams are taken and the diagnosis of anomaly is made.

In 1958 Hay, in discussing criteria for the diagnosis of minor congenital uterine anomalies, brought attention to physical signs observed at routine prenatal examination that should arouse suspicion of their presence. One or more of the following findings (illustrated in Fig. 2) is an indication for hystero-graphic evaluation post partum and leads to confirmation of anomaly in almost 100 per cent of cases: fetal limb flanking with overlie; breech flanking with contralateral limb flanking; fetal limb flanking with fundal notching; broad fundus with fundal notching; cystic formation of uterus; positive Piscacheck's Sign early in pregnancy; persistent breech presentation; axial deviation of the uterus; and floating vertex at term in a primigravida.

If manual removal of the placenta becomes necessary, the obstetrician should be aware of the phenomenon of triangular spasm and cornual pocketing, as described by Hay. He was able to demonstrate this only in the abnormal uterus. He found that oxytocics failed to produce it in the normal uterus, whereas they increased the degree of pocketing in the abnormal uterus.

Investigation of the genital tracts of all patients found to have congenital absence

GENITAL-TRACT ANOMALIES

of the kidney, fused kidneys, or ectopic kidneys is indicated because of the close kinship of the two tracts developmentally. In the 56 personal cases, 24 intravenous pyelograms were done, and 6 (25 per cent) showed agenesis of the kidney unilaterally, and 2 (8 per cent) showed kidney anomalies. Five of 12 intravenous pyelograms (41.6 per cent) in patients with uterus bicornis bicollis showed complete absence of one kidney. Two of 8 pyelograms (25 per cent) of pa-

tients with uterus bicornis unicollis showed ectopic or fused kidneys, but not agenesis. Woolf and Allen reported major renal involvement in 100 per cent of patients with imperfect development of one müllerian duct and a major urinary-tract anomaly in 35 per cent of patients with bilateral müllerian-duct imperfection. Collins (1932) presented 231 females with unilateral renal anomalies and found an associated genital anomaly in 90 per cent.

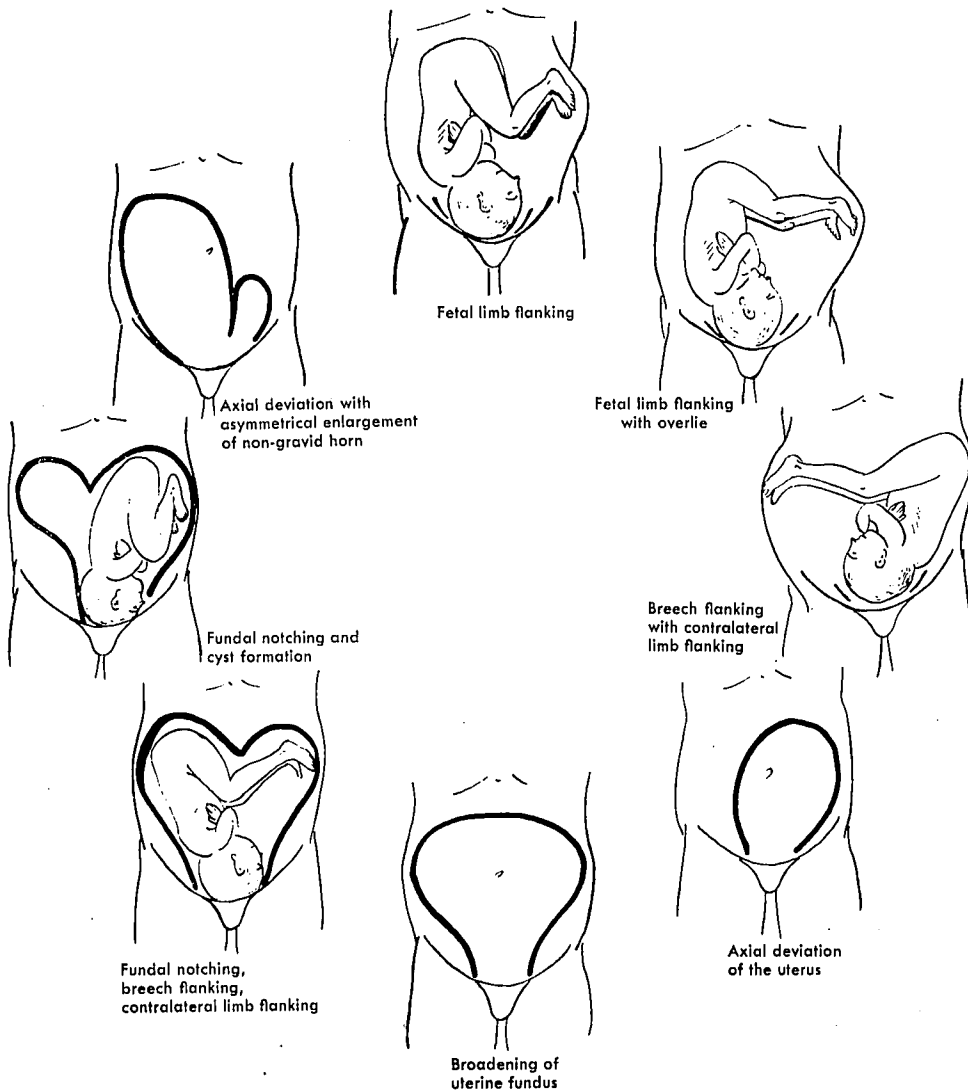


Fig. 2. Diagnostic signs of uterine anomalies. (Modified from Hay.)

CLINICAL BEHAVIOR

Personal Series

GROUP I. The distribution of the 30 cases in this group was as follows: uterus bicornis bicollis, 25; uterus unicornis, 1; and uterus bicornis unicollis with one rudimentary horn, 4. Of 23 of the patients with uterus bicornis bicollis who were capable of conceiving, 21 did so 52 times, an average of 2.47 pregnancies per patient. The 52 pregnancies (one undelivered) terminated in the delivery of 34 living infants. The fetal wastage was 33 $\frac{1}{3}$ per cent: there were 17 spontaneous abortions, 11 occurring before the third month and 4 occurring at 20 weeks in 1 patient whose hystero-grams showed 50 per cent of one uterine cavity to be displaced by a fibroid tumor.

The patient with uterus unicornis, 21 years of age, was operated upon for endometrioma of the right ovary and found to have occlusion of her solitary right tube. She was treated by salpingolysis and ovarian cystectomy. The left tube, uterus, ovary, and upper half of the left broad ligament were absent. Postoperative hystero-grams confirmed tubal patency, and urologic studies showed a normal upper urinary tract.

In 2 of the 4 cases of uterus bicornis unicollis with one rudimentary horn, the condition was diagnosed and the rudimentary horn removed surgically. A third patient underwent uterine suspension operation elsewhere, but hemihysterectomy was deferred for some unknown reason. In the fourth patient, a hematometra of the rudimentary horn developed, and drainage was accomplished through a stab wound in the vaginal fornix that entered the lateral wall of the rudimentary uterine horn. This wound became a fistulous tract, and 12 years later a conception in the rudimentary horn was aborted at 3 months. These patients had a total of 11 pregnancies, all in the left normal hemiuterus, with the exception of the pregnancy that occurred via the vaginal fistula.

Seven of the 11 pregnancies terminated in delivery of viable infants. The fetal wastage was 36.4 per cent, with all abortions occurring before the twentieth week. Excluding the rudimentary horn pregnancy, the fetal wastage was 30 per cent.

There was thus a total of 63 pregnancies among 25 patients in Group I, an average of 2.5 pregnancies per patient. The average age of the patients at the time of the first conception was 20 years, indicating little, if any, fertility problem.

Complete menstrual histories for all 30 patients of Group I were available. The average age at menarche was 13.6 years, with 94 per cent of the periods regular (28–30 days), and only 8 per cent reporting occasional menorrhagia. Menstruation was observed to occur simultaneously from both uteri in all 25 patients with uterus bicornis bicollis. Sixty per cent complained of dysmenorrhea, but only 15 per cent required medication or were incapacitated. Two patients with rudimentary horns had hemihysterectomies for dysmenorrhea, and one of the removed uteri contained a hemocast. The dysmenorrhea of a patient with uterus bicornis bicollis was relieved by dilation of the cervix of an almost infantile hemiuterus. One patient with vaginal atresia and associated hematoceles experienced no dysmenorrhea or pain until 12 hours prior to admission, though she presented with one uterus distended with blood and comparable in size to a 3-month gestation.²²⁴

Of 40 presentations in Group I patients, 67.5 per cent were vertex and 32.5 per cent, breech, and there were no transverse lies. Of 41 deliveries, 46.3 per cent were spontaneous, 12.1 per cent by forceps, 14.6 per cent by breech assist, 9.6 per cent by breech extractions, and 14.6 per cent by cesarean section. Two sections were for cephalopelvic disproportion after adequate trials of labor. One elective section was performed elsewhere for the second pregnancy of a patient who had previously aborted spontaneously

GENITAL-TRACT ANOMALIES

at 3 months. The remaining three sections were on the same patient. Her first two pregnancies were both obstructed during labor by incarceration of the non gravid horn, which, incidentally, occurred in opposite uteri in each pregnancy. Her third pregnancy was terminated by elective repeat cesarean section.

Of the infants delivered in Group I patients, 22.5 per cent were premature by weight and gestational age—30 per cent weighed less than 6 lb., 67.5 per cent, less than 7 lb., 25 per cent, between 7 and 8 lb., and only 7.5 per cent, over 8 lb.

Intravenous pyelograms were done on 16 of the 30 patients in Group I, and 6 had agenesis of one-half of the upper urinary tract (37.5 per cent). Five of the 6 patients with unilateral renal agenesis had uterus bicornis bicollis.

Notable complications of pregnancy associated with Group I anomalies of the personal series were as follows.

<i>Complication</i>	<i>No. cases</i>	<i>%</i>
ANTE PARTUM		
Pyelitis (50 per cent associated with urologic anomaly)	6	14.6
Passed decidual cast	5	12.5
Bleeding	5	12.5
INTRA PARTUM		
Septal dystocia (requiring resection)	5	12.5
Incarcerated non gravid horn	2	5.0
Retained placenta	2	5.0
POST PARTUM		
None	0	0

GROUP II. Of the 25 patients in this group, 23 had uterus bicornis unicollis (bicornuate, septate, and subseptate), and 2 had uterus arcuatus. Twenty-three of these patients were able to conceive, and there were 84 pregnancies (3 undelivered), an average of 3.65 per patient. The 81 pregnancies terminated in the delivery of 41 living infants and 40 spontaneous abortions, a fetal wastage of 49.3 per cent. One abortion occurred before the twelfth week. There was

one set of twins among the 7 abortions that occurred between the thirteenth and twentieth weeks.

Complete menstrual histories were available for all patients in this group. The average age at menarche was 13.1 years; 92 per cent of the cycles were regular (23–30 days); 8 per cent were noted to have menses as infrequently as every 90 days; and 20 per cent were incapacitated by disabling dysmenorrhea. In later years, 36 per cent of the patients in this group presented themselves to the clinic with moderate to severe menorrhagia refractory to treatment, necessitating hysterectomy. This incidence of menorrhagia and dysmenorrhea is high, and it should be noted that these symptoms persist despite increasing multiparity.

One patient who had five consecutive spontaneous abortions and disabling dysmenorrhea was treated by a presacral neurectomy, and a diagnosis of bicornuate uterus was made. Although no corrective surgery of the uterine anomaly was attempted, this patient was relieved of her dysmenorrhea and carried her sixth and eighth pregnancies to term. Delivery was accomplished by cesarean section after a trial of labor in her sixth pregnancy, and by repeat section in her eighth pregnancy. Is the high rate of fetal wastage in these patients due entirely to faulty implantation of the placenta and inadequate uterine circulation of the medial septa, as suggested by Falls, or is there possibly a neurogenic factor as well?

Of 41 presentations in Group II patients, 78 per cent were vertex, 19.5 per cent, breech, and 2.5 per cent (1 case), transverse lie. Ninety-four per cent of the pregnancies terminated in active labor, and only 7 per cent had labor prolonged beyond 24 hours, with 88 per cent delivering in less than 12 hours and 66 per cent in less than 6 hours. There was only 1 case of uterine inertia, a complication frequently reported to be associated with this group of uterine anomalies.

SEMMENS

Delivery was spontaneous in 56.1 per cent, by breech assist in 15 per cent, by forceps in 20 per cent (this includes 2 forceps rotations), and by cesarean section in 7.5 per cent. Two sections were described above, and the third was for vaginal bleeding and transverse lie.

Of the infants delivered in Group II, 22.9 per cent were premature by weight and gestational age; 34 per cent weighed less than 6 lb., 53.6 per cent, less than 7 lb., 22 per cent, between 7 and 8 lb., and 25 per cent, more than 8 lb. Four infants weighed more than 8 lb., indicating that although the prematurity rate is about equal in the two groups, the infants that tend to be larger at birth are delivered from the uterine cavity originating from more than one müllerian duct.

Eight intravenous pyelograms were done on patients in Group II, and no evidence of agenesis of the upper urinary tract was noted. One patient with recurrent pyelitis in each of three pregnancies was found to have a horseshoe kidney. Despite the fact that she was known to have a uterine anomaly, the urinary tract was never investigated until she was evaluated for hysterectomy because of recurrent menorrhagia.

A Strassmann unification procedure was done for a patient having one premature delivery followed by three successive abortions. Unfortunately, she was lost to follow-up because of military transfer. Group II patients beyond the twentieth week of gestation have relatively few complications until the postpartum period. Postpartum hemorrhage, the most severe complication, resulted from uterine atony in the immediate postpartum period in 4 cases. One hemorrhage was delayed until the fourth day, when a fragment was found adherent and removed; the uterus required packing to control hemorrhage. Manual removal of the placenta became necessary in 4 cases because of pocketing and trapping of the placenta in the gravid horn as a result of more rapid involution of

the nongravid horn. Complications experienced in the delivery of the 41 viable infants in Group II were as follows.

<i>Complication</i>	<i>No. cases</i>	<i>%</i>
ANTE PARTUM		
Bleeding	10	24
Threatened abortion	4	
Pyelitis	3	
Horseshoe kidney	1	
Passed decidua cast	2	
Premature rupture of BOW	5	12
INTRA PARTUM		
Uterine inertia	1	
POST PARTUM		
Hemorrhage	6	14.6
500 cc.	2	
1200 cc.	2	
1500 cc.	2	
Retained, trapped placenta	4	10

Literature

Since the clinical material of the personal series was insufficient to permit conclusions, the literature was thoroughly screened, and the 500 additional complete case reports of patients with pregnancy associated with uterine anomalies that were found were analyzed statistically according to the clinical classification proposed.

GROUP I. The distribution of the 273 cases in this group was as follows: uterus bicornis bicollis, 200; uterus unicornis, 14; and uterus bicornis unicollis with one rudimentary horn, 59. Of these 273 patients, 262 were capable of conceiving, and only 2.2 per cent failed to do so. There were 699 pregnancies, an average of 2.66 pregnancies per patient, with 81 per cent delivered at term and 19 per cent premature by weight and gestational age. The fetal wastage was 23.3 per cent for uterus bicornis bicollis, 21 per cent for uterus unicornis, and 34.3 per cent for uterus bicornis unicollis with one rudimentary horn. In the latter group, elimination of the pregnancies in the rudimentary horn resulted in a fetal wastage of 14.9 per cent from the functional hemiuterus. Of the abortions among the patients with uterus

GENITAL-TRACT ANOMALIES

bicornis bicollis, 82 per cent occurred before the twelfth week, and 100 per cent of the abortions (excluding those in the rudimentary horn) from the other types of hemiuteri in the group also occurred before the twelfth week. Four infants survived abdominal delivery from the rudimentary horn.

Of 572 pregnancies occurring in the normal hemiuterus, 118 terminated in abortion (7 therapeutic) and 27 in the delivery of stillborn infants. Correcting for therapeutic abortion, the fetal wastage was 24.4 per cent.

There were 150 complete menstrual histories available for analysis. The average age at menarche was 13.1 years, with 88.4 per cent of the patients having regular cycles (28–30 days), and 85 per cent having a moderate flow of 5 days or less. Thirty per cent complained of dysmenorrhea requiring treatment. Dysmenorrhea was a primary complaint among 50 per cent of the patients in whom one uterine horn was rudimentary, though no correlation was made as to whether there was communication with the functioning hemiuterus. Kehrer (according to Eastman) found no communication in 84 of 108 patients.

Seventy per cent of the presentations were vertex, 25 per cent, breech, and 5 per cent, transverse lies. Three hundred-eleven labors were described, as were 32 laparotomies for rupture of the rudimentary horn. Of the labors, 69 per cent were normal, 2.8 per cent, precipitous, and 5.1 per cent prolonged beyond 24 hours. Primary cesarean sections were done after a trial of labor in 2.2 per cent, 18.2 per cent were sectioned electively without a trial of labor, and an additional 2.5 per cent were delivered electively by repeat sections. Delivery was spontaneous in 54 per cent, by breech assist in 5 per cent, by breech extraction in 2.6 per cent, by forceps in 7.9 per cent, and by section in 30.5 per cent.

Major complications experienced in the delivery of the 454 pregnancies that reached

viability in the normal hemiuterus were as follows:

<i>Complication</i>	<i>No. cases</i>
ANTE PARTUM	
Bleeding	20
Placenta previa	5
Premature rupture of membranes	30
Passed decidua cast	10
Fetal dystocia	14
Unengaged vertex	4
Twin pregnancy	10
Septal dystocia	5
INTRA PARTUM	
Septal dystocia	18
Incarceration of non gravid horn	16
POST PARTUM	
Hemorrhage	16
Less than 500 cc.	9
More than 1000 cc.	7
Adherent placenta	10
Passed decidua cast	8
Maternal deaths (rudimentary horn rupture)	2

GROUP II. In this group, there were 202 cases of uterus bicornis unicollis (bicornuate, septate, and subseptate) and 25 of uterus arcuatus. All patients except 1 conceived, and the total number of pregnancies was 639. The average number of pregnancies was 2.94 per patient among those with uterus bicornis unicollis and 2.4 per patient among those with uterus arcuatus. There were 360 live-born infants delivered of patients with uterus bicornis unicollis, with 15.7 per cent premature by weight and gestational age. There were 51 viable infants delivered of patients with uterus arcuatus, all at term. The over-all fetal wastage was 37.1 per cent in patients with uterus bicornis unicollis as compared to 19 per cent in uterus arcuatus. Of particular interest is the fact that the incidence of late abortion in Group II was twice that in Group I (34.7 per cent as compared to 17.3 per cent).

Complete menstrual histories were available for only 70 of the patients in this

group. The average age at menarche was 14.7 years; the cycles of 78 per cent of the patients were regular (28–30 days), with moderate flow, 22 per cent had profuse periods, and 30 per cent complained of dysmenorrhea which required treatment.

There were 310 presentations described in Group II patients: 68 per cent were vertex, 22.2 per cent, breech, and 9.8 per cent, transverse lies. Of 278 labors recorded, 75 per cent were normal, 1.5 per cent, precipitous, and 6.8 per cent, prolonged beyond 24 hours. Primary sections were accomplished in 3 per cent after a trial of labor, 12.6 per cent were delivered by elective section without a trial of labor, and 1 per cent were repeat sections. Of 250 deliveries, 50 per cent were spontaneous, 10 per cent by low forceps, 4.8 per cent, by breech assist, 10.4 per cent, by breech extraction, 2.5 per cent, by internal podalic version and extraction, and 22.3 per cent, by cesarean section.

Only 17 of the patients had the benefit of upper urinary-tract evaluation, of these, 6 had unilateral renal agenesis and 1 had an ectopic kidney, indicating a 40 per cent incidence of anomalies among this sparse sampling.

In the 402 pregnancies resulting in viable fetuses in Group II, the following major complications were noted.

<i>Complication</i>	<i>No. cases</i>
ANTE PARTUM	
Fetal dystocia	34
Transverse lie	32
Bleeding	20
Placenta previa	9
Premature rupture of membranes	20
Preeclampsia	13
Septal dystocia	8
Incarceration nongravid horn	6
Passed decidual cast	4
INTRA PARTUM	
Inertia	10
Prolapsed cord	6

POST PARTUM	
Adherent placenta	28
Hemorrhage	26
500 cc.	9
1000 cc.	5
1500 cc.	8
2000 cc.	4
Uterine atony	14
Failure of uterine involution	11
Maternal deaths (peritonitis, 1, ruptured uterus, 2, convulsions, 1, and shock, hemorrhage, and sepsis, 1)	7

DISCUSSION

Two factors are of paramount importance: First, there must be a clinical awareness of the existence of the anomaly, and second, if the exact nature of the anomaly is known, most of the complications of pregnancy can be anticipated.

Suspicion of the presence of these anomalies must be aroused in the mind of the clinician on the basis of the patient's obstetric and gynecologic history. These patients have a similar behavior pattern in past pregnancies, and they are prone to present with dyspareunia, infertility, habitual abortion, disabling dysmenorrhea, and menorrhagia which persists despite curettage and hormone therapy.

Aids in diagnosis have been advanced recently by such writers as Hunter and Hay, who pointed out the difficulty in arriving at the true anatomic diagnosis in either the gravid or the nongravid state. It has been increasingly evident that there is a real clinical need for a functional classification that can be employed in the management of these patients. The classification described above may meet this need.

The cervix is an aid to diagnosis rather than a deterrent to delivery, whether it is of single or double müllerian origin. The presence of an asymmetrically located cervix in the vaginal fornix, an excessively large cervix with or without a septum in a nullipara, or a duplicated cervix may indicate the presence of uterine anomaly and warrants investiga-

GENITAL-TRACT ANOMALIES

tion. Funneling of the cervix in the hemi-uterus and failure of the presenting part to engage were mentioned by Hunter¹¹⁹ but were not observed in this series in 63 pregnancies occurring in uteri of single müllerian origin. The vaginal septum is a clue to the presence of uterine anomaly. It may cause dyspareunia, sterility, hematocele, or dystocia. Often a deterrent to adequate investigation of the uterine cavity, the vaginal septum should be removed at the earliest opportunity, preferably in the nongravid state.

Hay suggested specific clinical findings as opposed to clinical behavior which enabled him to diagnose minor congenital anomalies in 10 per cent of 2088 patients. These findings, discussed above under *Diagnosis*, are especially noteworthy at routine prenatal examination. If two or more of these signs are present, especially late in pregnancy, the probability of the existence of an anomaly is increased. Hay also discussed pocketing of the cornu and triangular spasm of the fundus, a phenomenon found only in the anomalous uterus and accentuated by the use of oxytocics.

The diagnosis can be made positively only by hysterosalpingography or at laparotomy. Any findings at vaginal examination, uterine palpation, or exploration of the uterine cavity with sounds must be confirmed by X-ray. A normal urinary tract should never be assumed in the presence of a genital-tract anomaly. In the present survey of case re-

ports, only 19.4 per cent of the 273 patients in Group I and 7.5 per cent of the 225 patients in Group II had the benefit of urologic investigation. The over-all percentage of upper urinary-tract studies reported among the 500 cases surveyed in the literature was a mere 13.4 per cent (Table 1). A positive hystero-gram warrants investigation of the upper urinary tract by intravenous pyelography. The importance of urinary-tract evaluation was stressed by Wharton, Collins, and Schumacker, and Woolf and Allen stated that failure to do so was tantamount to negligence. They found that when one müllerian duct was imperfectly developed, there was major renal involvement in 15 of 15 cases, and that when both müllerian ducts were imperfect, there was a major urinary anomaly in 35 per cent. Urinary-tract examination is especially important if one is contemplating a major pelvic operation.

Conversely, an investigation of the genital tract of every patient with renal agenesis or ectopia is equally important. Collins found associated genital anomalies in 90 per cent of 231 females with unilateral renal anomalies.

In most of the personal cases, the diagnosis was based on the results of hystero-graphy, but the presence of uterine anomaly was suspected because of one or more of the following: (1) The presence of a vaginal or cervical septum, asymmetric location of the cervix in the lateral vaginal fornix, or an irregular uterine contour; (2) abnormal configurations of the uterine fundus in the third

TABLE 1. RESULTS OF UROLOGIC STUDY IN PATIENTS WITH GENITAL-TRACT ANOMALIES

	<i>No. patients</i>	<i>Intravenous pyelograms</i>	<i>Laparotomy autopsy*</i>	<i>Agenesis of kidney</i>	<i>Abnormal or ectopic kidney</i>	<i>Percentage patients studied</i>
<i>Group I</i>						
Personal series	30	16	0	6—37.5%	0	53%
Literature	273	53	0	27—50.8%	5—9%	19.4%
<i>Group II</i>						
Personal series	25	8	0	0	1—12.5%	30.0%
Literature	227	14	2	6—43%	1—7%	7.5%

* On patients who died with uterine rupture or, shortly after birth, with uterine anomaly and renal agenesis.

trimester; (3) abnormal presentations or failure of the presenting part to engage at term or in early labor in a pelvis free of the usual causes of dystocia; (4) partial or complete separation of the placenta in the third stage, followed by its trapping in the uterus as the result of an abnormal contraction pattern; and (5) past obstetric history of habitual or successive abortions occurring early in gestation, recurrent breech presentations, or recurrent pyelitis in pregnancy in the absence of chronic renal disease.

Recently, I have employed hysterographic studies on all postpartum patients with axial deviation or overlie and flanking of the breech or extremities during the prenatal period. Thus far the incidence of anomalies discovered in such patients has been less than 2 per cent.

In the personal series, only 3 patients with uterine anomalies were found as a result of infertility study, whereas Hunter¹¹⁹ reported 73.7 per cent fertility, a smaller total number of pregnancies, and fewer viable pregnancies among 38 married women with double uteri. Table 2 shows the fertility of the patients in Groups I and II, both in the personal series and in the literature, according to type of anomaly. Patients with uterus unicornis were the least fertile (85.7 per cent), averaging 1.34 pregnancies. The average number of pregnancies was 2.1 and 3.65 in Groups I and II of the personal series and 2.2 and 2.84 in Groups I and II of the reported cases.

Table 3 summarizes the menstrual histories. The hemiuterus of a single müllerian origin appears to be functionally superior to the uterus derived from an imperfection of dual origin. Menarche was normal, periods were regular in 88–94 per cent, and menorrhagia was noted in only 8–12 per cent. Conversely, menarche was delayed, and periods were irregular and more profuse in the uteri of larger capacity. Dysmenorrhea appeared to be prominent in both groups and was persistent despite increasing age and parity. The

50 per cent incidence of dysmenorrhea associated with rudimentary uteri reflects a mechanical problem. The endometria of rudimentary horns removed surgically showed a lack of end-organ response in some, adenomyosis in others, and pressure atrophy in one hemiuterus with a hemocast.

Fetal wastage offers the greatest challenge. Patients should no longer experience three or more successive abortions before being evaluated for the possibility of uterine anomaly. Table 4 shows the hemiuterus to be functionally superior to the anomalies of Group II in terms of viable infants delivered. That any infants survived delivery from a rudimentary horn is commendable. When diagnosed, it is a surgical emergency, and only by early intervention, whole-blood transfusion, antibiotics, and newer anesthetic technics has the maternal mortality been kept down. It is noteworthy that the fetal loss from the functional hemiuterus associated with the rudimentary horn was less than 15 per cent.

The 49.3 per cent fetal wastage in Group II of the personal series is reflected in the histories of 4 patients having a total of 17 abortions before uterine anomalies were diagnosed. Unsuspected, the diagnoses were made at laparotomy, dilatation and curettage, and cesarean section for transverse lie. Early consideration of anomaly and adequate study with surgical correction might have reduced this unnecessary fetal loss. Strassmann²³⁹ showed improved fetal salvage (from 4 to 84 per cent) in a review of 225 patients with similar obstetric histories.

Jones and Jones stressed endocrine and metabolic study prior to surgical correction since the anomaly may be only a secondary factor in repeated abortion. Reboul and Maës did pregnandiol studies and employed supplemental hormones with successful termination of pregnancies treated. We have used increasing doses of diethylstilbestrol and 300 mg. of ascorbic acid daily for this purpose.

TABLE 2. FERTILITY OF PATIENTS WITH GENITAL-TRACT ANOMALIES*

	Group I						Group II			
	Personal series			Literature			Personal series		Literature	
	BB	UU	BURH	BB	UU	BURH	BU	UA	BU	UA
No. patients	25	1	4	200	14	59	23	2	202	25
Fertility (%)	91.3	0	100	98.4	85.7	94.9	92	84	100	100
No. pregnancies	52	0	11	417	19	163	84	84	576	63
Average No. pregnancies per patient	2.47	0	2.75	2.21	1.4	2.9	3.65	3.65	2.94	2.4
OVER-ALL AV.	2.1		2.2		2.84					

* "BB" indicates "uterus bicornis bicollis"; "UU," "uterus unicornis"; "BURH," "uterus bicornis unicollis with one rudimentary horn"; "BU," "bicornuate uterus"; "UA," "uterus arcuatus."

TABLE 3. CHARACTERISTICS OF MENSTRUAL HISTORIES OF PATIENTS WITH GENITAL-TRACT ANOMALIES

	Group I		Group II	
	Personal series	Literature	Personal series	Literature
Av. age at menarche (yr.)	13.6	13.1	13.1	14.7
Regular cycle (%)	94.0	88.4	92.0	78.5
Moderate flow (%)	92.0	85.0	64.0	78.0
Menorrhagia (%)	8.0	12.0	36.0	22.0
Severe dysmenorrhea (%)*	15.0	30.0	20.0	30.0

* In patients with uterus bicornis unicollis with one rudimentary horn, incidence of severe dysmenorrhea was 50 per cent in both personal series and reported cases.

TABLE 4. PERCENTAGE OF FETAL WASTAGE IN PATIENTS WITH UTERINE ANOMALIES ACCORDING TO FUNCTIONAL TYPE

	Group I				Group II			
	Personal series		Literature		Personal series		Literature	
	UU	BURH	BB	UU	BURH	BU and UA	BU	UA
Percentage series	33.3	0	29.3	21.0	34.3	49.3	37.1	19.0
Rudimentary horn wastage		100.0			90.0			
Normal hemiterus wastage		30.0			14.9			
Over-all fetal wastage		35		30.7		49.3		35.3
Loss prior to 12 weeks gestation		64		82.7		82.5		67.6



TABLE 5. FETAL PRESENTATIONS IN PATIENTS WITH GENITAL-TRACT ANOMALIES

	Vertex		Breech		Transverse lie	
	Personal series (%)	Literature (%)	Personal series (%)	Literature (%)	Precipitous	No labor, Trial operative
Group I						
Personal series (%)	67.5	32.5	0			
Literature (%)	70.0	25.0	5.0			
Group II						
Personal series (%)	78.0	19.5	2.5			
Literature (%)	68.0	22.2	9.8			

TABLE 6. LABOR IN PATIENTS WITH GENITAL-TRACT ANOMALIES

	Precipitous		Normal		Prolonged only		del.	
	Personal series (%)	Literature (%)	Personal series (%)	Literature (%)	Personal series (%)	Literature (%)	Personal series (%)	Literature (%)
Group I								
Personal series (%)	15	70	0.3	0.3	3.4			
Literature (%)	1	70	0	0	19.0			
Group II								
Personal series (%)	0	88	7	2.5	2.5			
Literature (%)	1.4	75	6.8	3.0	12.6			

GENITAL-TRACT ANOMALIES

The high percentage of abnormal presentations is illustrated by the 32.5 per cent incidence of breech presentations occurring in Group I of the personal series and the 20–25 per cent incidence in all other groups of patients (Table 5). The transverse lies occurred in the larger uterine cavity associated with Group II anomalies. The fetus soon assumes a hammock position as the fundal notch or septum is drawn up by the rising fundus. It is of particular interest that the incidence of breech presentations closely paralleled the findings in a recent X-ray study of fetal lie in deformed uteri in the second trimester. Hunter¹¹⁹ called attention to funneling of the lower uterine segment and fundal moulding of the fetal head in the hemiuterus as a deterrent to natural version of the fetus late in the second or early in the third trimester.

Labor is influenced by the abnormal contraction patterns caused by the variations in architecture of the uterine fundus as well as by the occasional presence of a weakened mesial uterine wall. A failure to permit even a trial of labor has eliminated as many as 30 per cent of the patients in some groups reviewed in the literature. It should be noted in Table 6 that 70 per cent of the labors in Group I were normal and that an additional 15 per cent were precipitous. Fenton and Singh reported anomalies associated with precipitous delivery three times more frequently than in normal uteri. Prolonged labor was evident only in Group II and was associated with abnormal contractions of dual cornua contracting individually against a single cervix in an oblique pattern.

The methods of delivery used are indicated in Table 7. It is obvious that in the personal series vaginal delivery of breech presentations and the use of low forceps were preferred to abdominal delivery which was preferred in the literature. These patients are normal obstetrically until proved otherwise. If labor progresses normally, vaginal delivery is the rule. One cannot appre-

ciate the functional capacity of these uteri if 12–19 per cent are sectioned electively without a trial. Any increase in incidence of cesarean section should reflect only those complications associated with uterine anomaly which are indications for abdominal delivery. Inertia should be managed by abdominal delivery rather than Pitocin stimulation, which would produce pocketing of the cornu, abnormal fundal contraction patterns, and placental separation.

Study of the birth weights (Table 8) indicates that the incidence of prematurity was identical in the two Groups; however, the causes of premature delivery were entirely different. In Group I the limited uterine capacity of the hemiuterus promoted premature delivery, whereas in Group II, the large, soft cervix inherently weakened by abnormal muscular development permitted premature rupture of the membranes and subsequent premature delivery. If the cervix is free of defect, the increased uterine cavity fosters the development of larger infants (8 lb. plus) three times more often than in the functional hemiuterus.

Only the more frequent complications of labor and delivery are shown in Table 9. It is evident that bleeding was the most common problem. The bleeding of threatened abortion deserves special note, in view of the decidual cast and because misinterpretation of the gross tissue specimen could lead to curettage of a gravid uterus and inadvertent interruption of pregnancy. This is especially important when the two uteri are capable of functioning independently, i.e., in uterus bicornis bicollis and in uterus bicornis uncollis of the bicornuate variety if the fundal indentation approaches the cervical segment. All aborted tissue must be examined before initiating further treatment. The differential diagnosis between decidual cast and placenta can be accomplished by means of frozen section. Grossly, the tissue is a hollow cast, but microscopically, there is an absence of placental villi. Many more casts than are re-

SEMMENS

TABLE 7. METHODS OF DELIVERY OF PATIENTS WITH GENITAL-TRACT ANOMALIES

	<i>Spontaneous</i>	<i>Forceps</i>	<i>Breech assist</i>	<i>Breech extraction</i>	<i>Version & extraction</i>	<i>Cesarean section</i>
<i>Group I</i>						
Personal series (%)	46.3	12.1	14.6	9.6	0	14.6
Literature (%)	54.0	7.9	5.0	2.6	0	30.5
<i>Group II</i>						
Personal series (%)	56.1	20.0	15.0	0	0	7.5
Literature (%)	50.0	10.0	4.8	10.4	2.5	22.3

TABLE 8. FETAL BIRTH WEIGHT IN PATIENTS WITH GENITAL-TRACT ANOMALIES

	<i>Premature</i>	<i>Less than 6 lb.</i>	<i>Less than 7 lb.</i>	<i>7-8 lb.</i>	<i>8 lb. or more</i>
<i>Group I</i>					
Personal series (%)	22.5	30	65.5	25	7.5
Literature (%)	18.5				
<i>Group II</i>					
Personal series (%)	22.9	34	53.6	22	25.0
Literature (%)	18.0				

TABLE 9. COMPLICATIONS OF PREGNANCY IN PATIENTS WITH GENITAL-TRACT ANOMALIES

	<i>Group I</i>		<i>Group II</i>	
	<i>Personal series</i>	<i>Literature</i>	<i>Personal series</i>	<i>Literature</i>
<i>Antepartum complications</i>				
Pyelitis of pregnancy (%)	14.6	0	6.6	0
Passed decidua cast (%)	12.5	2.2	4.4	1.0
Bleeding, all causes (%)	18.5	4.4	22.0	9.0
Premature rupture of membranes (%)	0	6.6	12.5	5.0
Toxemia of pregnancy (%)	0	2.2	0	3.2
Fetal dystocia (%)	0	0	0	8.5
<i>Intrapartum complications</i>				
Incarcerated nongravid horn (%)	5	4	0	1.5
Septal dystocia (require-resection) (%)	12.5	4.6	0	2.5
Inertia (%)	0	1.6	2.2	2.5
Fetal distress (%)	0	0	0	4
<i>Postpartum complications</i>				
Retained placenta (%)	5	2.2	10.0	7.0
Hemorrhage (%)	0	3.2	14.6	6.5
Passed decidua cast (%)	0	1.6	0	0.75
Maternal deaths (%)	0	0.8	0	1.75

GENITAL-TRACT ANOMALIES

ported were passed by these patients; thus the true incidence remains unknown. Placenta previa also appears increased, and trauma to the septa of the uterine cervix or vagina with descent of the fetus may cause considerable bleeding.

Knowledge of the patient's anomaly is of decided advantage in management. Hunter¹¹⁹ described breech presentations which perforated the septa and prevented the descent of the presenting part, thereby requiring abdominal delivery.

Premature rupture of the membranes was more common in patients in Group II of the personal series, but in the over-all study it occurred equally often in both groups. Since the incidence of prematurity is equal, I feel that while the decreased uterine capacity may account for those in Group I, the large, patulous cervix with poor muscular tone predisposes to early rupture of the membranes and subsequent premature labor in Group II, thus presenting two entirely different mechanisms with the same ultimate result.

Pyelitis was frequently associated with congenital anomalies of the upper urinary tract in the personal series, although this complication was not mentioned in the literature. Fetal dystocia was a mechanical problem, owing to the increased number of transverse lies, axial deviation, asynclitic engagement, and failure to engage until late in pregnancy.

There is a calculated risk of dystocia associated with a persistent vaginal septum. It is recommended that resection be done as soon as this condition is recognized—early in pregnancy or even early in labor, if it cannot be accomplished in the nongravid state.

Incarceration of the nongravid uterine horn is a relatively infrequent complication peculiar to patients with uteri bicornis bicollis and bicornis unicollis and is an indication for immediate abdominal delivery. Salmon reported a case of 10 repeated abortions prior to delivery of a term infant. The uteri were described as two saddlebags which

straddled the rectum. He resected the rectovesicle septum, denuded the mesial serosal surfaces of the uteri, and approximated the uterus mesially to effect suspension. This procedure has possible merit.

Bleeding is the principal postpartum complication and is associated almost entirely with abnormal uteri of dual müllerian origin. Adherent placenta, trapped placenta, uterine atony, and subinvolution occur in 10 per cent or more of the cases. Blair reported subinvolution of the uterus in 25 of 65 patients with uterine anomalies. The frequency of these problems among the patients with Group II anomalies warrants adequate blood typing in labor, liberal use of oxytocics, and careful observation immediately following delivery, as well as thorough examination prior to discharge. Prolonged hospitalization is advisable.

SUMMARY

1. The management of 556 patients with congenital uterine anomalies is presented—56 with 146 pregnancies from the author's personal series, and 500 with 1234 pregnancies reported in the literature.

2. A simplified clinical classification, offered as a practical approach to the management of these patients, is based entirely on the functional capacity of the uterine cavity and divides female genital tract anomalies into two groups: Group I, hemiuterus of single müllerian origin, paired or otherwise, and Group II, uterus of dual müllerian origin associated with varying degrees of absorption.

3. It is suggested that vaginal septa and minor cervical abnormalities be treated by excision, preferable in the nongravid state.

4. Aids in the diagnosis of congenital anomalies of the female genital tract are discussed with the aim of increasing clinical awareness of their existence.

5. Attention is directed to the need to evaluate the upper urinary tract in the study of these patients.

6. The clinical behavior of Groups I and II is described, on the basis of experiences with the 56 personal cases, as well as case reports, with emphasis on fertility, fetal wastage, menstrual behavior, fetal size, development, and presentation, behavior in labor and methods of delivery, and complications associated with prenatal, antepartum, intrapartum, and postpartum management.

7. The need for clinical awareness is stressed in order that these patients are properly treated before excessive fetal wastage occurs or extensive surgical procedures are accomplished that often terminate fertility.

8. Conservative management is advocated; plastic procedures, such as the Strassmann uterine unification, should be reserved for patients whose habitual abortion and infertility problems are positively related to their anomalies rather than to endocrinologic or metabolic disorders.

*Obstetrical and Gynecological Service
U. S. Naval Hospital
Naval Aviation Medical Center-54
Pensacola, Fla.*

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