Unaudited Clinical Experience

To the Editor (Letter 1 of 3):

I enjoyed reading the case report by McBean and Brumstead (1) in which the authors reported a complete septal uterine anomaly associated with a double cervical opening. The authors were apparently unable to find this anomaly in a standard classification scheme (2), nor could they find previous reports of similar cases.

This anomaly was reported by Daly et al. in 1983 (3) and included in the classification of uterine anomalies in two standard texts during the 1980s (4, 5). The three forms of a septate uterus are shown in Figure 1 from *Progress in Infertility,* 3rd ed. (5). The challenge associated with this anomaly was how to approach it surgically. Daly et al. (3) described a hysteroscopic approach that permitted removal of the septum with conservation of the double cervical opening during an outpatient surgical procedure. I found this extremely interesting and included a discussion and drawing of this procedure in the 3rd edition of *Progress in Infertility* (5).

I appreciate the opportunity to discuss this subject and hope this information will be helpful.

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August 9, 1994

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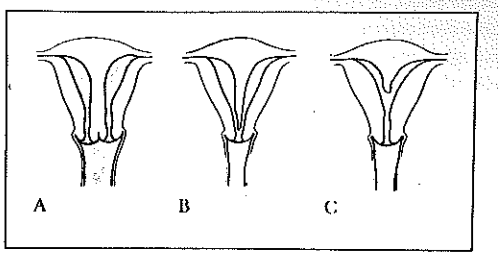

Figure 1 Reproduced by permission from the author of the chapter, and co-editor of the text (5).

To the Editor (Letter 2 of 3):

I was reading the August issue of *Fertility & Sterility* and was interested in the brief communication from Drs. McBean and Brumsted in which they report a "previously unreported case of complete uterine septum, cervical duplication, and a longitudinal vaginal septum" (1).

You might direct these authors' attention to one of the standard textbooks in our field, *Danforth's Obstetrics and Gynecology,* in which this anomaly has been clearly described in both the current (7th) as well as the previous edition. In the 7th edition, the anomaly is pictured in Figure 2-13E (p. 21) and described on pages 21–22.

Fortunately, however, there is no apparent disagreement among authors concerning the probable pathogenesis and recommendations for management.

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To the Editor (Letter 3 of 3):

Concerning the paper "Septate Uterus with Cervical Duplication: A Rare Malformation" by McBean and Brumsted (1), allow us to make some

comments. In our department, where we treat many patients for infertility, we do occasionally encounter this type of malformation because hysterosalpingography is routinely carried out. Indeed, between 1986 and 1993, 118 cases of uterine septum have been observed: 103 patients presented a partial uterine septum (87%) and 15 patients presented a septate uterus with cervical duplication (13%). A vaginal septum was noted in 8 cases (7%). The diagnosis of this type of malformation may sometimes be delayed, as in the article, particularly if a vaginal septum is associated. Indeed, the vaginal septum can be easily misdiagnosed by gynecological examination and at hysterosalpingography, the uterus appears to be unicornuate except if there is a fistula between the two uterine cavities. However, in the absence of a vaginal septum, the diagnosis is simple because two distinct external cervical orifices are clearly visible. The opacification through these two orifices allows the diagnosis of a septate uterus with cervical duplication.

In 1987, Rock et al. (2) described resectoscopic techniques used to treat a complete uterine septum, although there was no vaginal septum. Nisolle and Donnez (3, 4) have also described the laser technique applied in the treatment of all types of uterine septum (partial and complete with or without a vaginal septum). In cases of complete uterine septum, the main difference is that Rock et al. (2) leave the cervical septum intact whereas we remove it completely in one or two steps. Indeed, during the first step, the vaginal and cervical septum were resected using a CO^2 laser. The second step, performed 2 months later, consisted on the Nd:YAG laser resection of the uterine septum. In the last 5 cases, the resection of both uterine and cervical septum was performed in one step. Among the 5 patients, 3 had a complete vaginal septum, which was also resected. In our series, 10 of 15 patients became pregnant and no sign of cervical incompetence was observed. Because in the case described, there were no particular consequences of this malformation, no treatment was discussed. However, this type of malformation can often cause complications such as recurrent pregnancy loss; thus the description of the treatment is essential.

Considering personal series of 15 cases, we think that the diagnosis of a complete septum with a duplication of the cervix (associated sometimes with a longitudinal vaginal septum) is not such a rare malformation as described in the paper of McBean and Brumsted (1). Probably, this pathology is often underdiagnosed. Moreover, we proposed a complete resection of uterine, cervical, and vaginal septum as the optimal therapy.

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Reply of the Authors:

The purpose of our article was twofold; first, to report a müllerian anomaly that is not formally described in the literature. Second and most importantly, we wanted to review the embryology of this finding as it appears to contradict standard teaching of the development of the müllerian duct. It was our hope that the discussion of the embryology would not get lost in a debate about whether or not this is the first such case report in the literature. A septate uterus with complete cervical and vaginal duplication shouldn't exist if one believes that müllerian duct fusion occurs in a unidirectional, caudad to cephlad fashion. Despite its exclusion from a commonly used classification scheme (1) and its absence from several major gynecology texts (2, 3), we suspected that this was not an infrequent clinical finding. Several people have written to us with similar reports and we have also identified an additional patient in our own practice. The incidence of 7% reported by Nisolle and Donnez may be unique to their population. Our infertility service performed approximately 350 hysterosalpingograms in 1993 and only identified two women with this exact anomly (<1%). Before the advent of endoscopic techniques and common use of transvaginal

ultrasound, this anomaly may have been classified as either a bicornuate or didelphic uterus with a vaginal septum.

Our search of the literature through *Medline* was extensive, including over 5,000 titles dating back as far as 1960. We also included some major texts, but regrettably did not *Danforth's Obstetrics and Gynecology,* 7th ed. The anomaly is pictured in this textbook; however, the accompanying discussion does not include a consideration of the embryology of the anomaly. The septate uterus pictured in *Progress in Infertility* (4) does not appear to have an associated vaginal septum as was found in our patient. A septate uterus with cervical duplication does, however, raise the same questions regarding its embryologic development as the anomaly reported in our paper. Previous reports including that of Daly (5) have described the existence of a septate uterus associated with both a cervical and vaginal septum. The patient reported in our paper has a different anomaly because of the presence of two completely separate cervices, not a cervical septum. Hysteroscopic treatment is appropriate for both of these anomolies when associated with pregnancy loss.

Symmetric defects of the uterus are the most common uterovaginal anomalies. Therefore, we are not surprised to find that the anomaly reported in our paper has been documented by others. In the future we hope to find this anomaly included in major classification schemes.

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Editorial Comment
Unaudited Clinical Experience

This deluge of letters was precipitated by a seemingly simple, straightforward case report by Doctors McBean and Brumstead describing "a septate uterus with cervical duplication." No one, especially the authors, expected the publication to be such a bell ringer. In a few years when we are all electronically united the dialogue that you read here between the authors, correspondents, and editor will be on your computer network, and you can inject your own unaudited clinical experience.

It is apparent from this discussion that authors reporting unique cases should include details on the methodology and scope of their literature review. The provision of this information in the author's response letter is very helpful for the reader. The issue of true cervical duplication may depend on whether the medial surface of each cervix, when viewed from below, is discrete and distinct. From the cartoon provided by the authors, the medial aspect of both cervices appear to be fused and continuous with the uterine septum. No medial fornices are visible. The discussion makes one wonder if the anomaly is infrequently reported, infrequently well-described or just infrequently recognized. Doctor Musich's contribution is interesting, because of the apparent rarity of reports of vaginal delivery, or alternatively a selection bias in the literature against this outcome. One suspects that the septum during pregnancy, parturition, and immediately post partum would be displaced in the direction of the contralateral uterine wall. A quick inspection might conclude that the septum is gone.

The editor has seen a case that presented in the immediate post-partum period with unilateral genital tract obstruction due to inability of blood, lochia, or decidua to escape through the partially obliterated non-parturient cervix. In matters of anomalies, observations need to be precise and agreed upon by independent observers or raters. Otherwise authors, correspondents, experts, non-experts, and editors alike have overactive and underactive imaginations which tend to introduce pet distortions and opposing interpretations of these anomalies.

The appropriate treatment in the symptomatic patient will never be tested by an appropriate clinical trial unless a registry is developed for this purpose. The clinical management at this time as seen from the correspondence must depend on "unaudited clinical experience." The danger of unaudited clinical experience, when the anomaly or problem is infrequent, is that one can repeat the same mistake but with increasing confidence.

In addition to the post-partum obstructed genital tract the editor has seen two additional cases that turn out to be a case-control study of one. Each of the cases was associated with viable, third trimester deliveries and abnormal presentations. In the first case the physicians who saw the patient initially, elected to remove the septum in its entirety. The upper third was removed by a standard transuterine Tompkins procedure and the lower third was resected from below with two Kocher clamps. The patient never carried a subsequent pregnancy in spite of multiple procedures attempting to correct an incompetent cervix that resembled a "giant Krispy Kreme doughnut". The second patient, who was ascertained as a footling breech presentation at 35 weeks and delivered by caesarean section, never had a surgical correction. She had a similar event in a subsequent pregnancy, which was managed again by abdominal delivery. It would appear from the unaudited clinical experience of one uneducated and uninformed editor that the lower third of the septum in these cases should be treated with reverence and perhaps left intact. Doctor Patten alluded to this in his correspondence and the authors in the Daly-Riddick publication (1) attempted to conserve the lower third of the septum. Yet just as I had begun to feel increasingly confident about my unaudited clinical experience or unproven notions, a final letter came from two well-respected authorities in Bruxelles. They contributed their larger, but still unaudited clinical experience to the discussion. Without trepidation they removed the entire septum in two stages in a large series of 15 patients and did not experience the dreaded complication of cervical incompetency.

The final epiphany to this series of letters will not be written here. A higher celestial voice might give evidence for the "entire septum" or only the "upper third." An alternative approach is to have a forum called "Curbstone Consultations" at the annual meeting where cases such as this can be discussed by some type of group judgment method. Perhaps at that time some "unaudited clinical experience" can be vindicated, and treated with more respect. For cases that cannot wait for the annual meeting, the use of the new National Science Foundation organized internet or some other form of electronic dialogue should help the physician who is struggling with the appropriate management for these infrequent clinical problems. These forms of electronic communication and digital libraries should help the clinician to gather multiple opinions, and share experiences. This might help to avoid the distinctive distortions of these problems that arise from the limited experience of a single expert, or the pejorative influence of an authority. Last but not least there is no better ink on paper publication covering all of these malformations than was published in the second volume of Obstetrics and Gynecology *in 1953 by Ralph Woolf and Willard Allen (2). This paper, notable for its comprehensive treatment of the subject of uterine anomalies, was published almost 25 years after Dr. Allen, with his colleague and mentor George Corner (3, 4), isolated progesterone from sow corpus luteum—how's that for continued productivity?*

Paul G. McDonough, M.D., Editor, Letters

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Cumulus Cell Co-Culture—Efficacy?

To the Editor:

The paper by Saito et al. (1) concerning the use of cumulus cell co-culture in human IVF requires comment on the statistical significance assigned to part of the results by the authors and the methodology used to achieve cumulus cell co-culture for embryos.

First, the authors claim that there is a statistically significant improvement in the proportion of good quality embryos after 72 hours in the co-culture group (10 embryos out of a total of 34 cultured = 29%) compared with the proportion of good quality embryos in the control group at 72 hours (eight