Hysteroscopic metroplasty in diethylstilboestrol-exposed and hypoplastic uterus: a report on 24 cases

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The aim of this study was to determine the feasibility and safety of hysteroscopic metroplasty in cases of diethylstilboestrol-exposed and hypoplastic malformed uterus. Twenty-four patients were referred for primary infertility \((n = 9)\), secondary infertility \((n = 1)\) or infecundity \((n = 14)\). Fifteen had been exposed to diethylstilboestrol in utero. All patients had a hypoplastic uterus and/or uterine deformity as seen by hysterosalpingography and each served as their own control. All patients underwent hysteroscopic metroplasty. Outcome measures included postoperative hysterosalpingography and the ability to conceive and to carry pregnancy to live birth. Postoperative hysterosalpingograms revealed improvement in 23 cases; the final result was considered excellent in 15 cases and 11 pregnancies occurred. The abortion rate decreased from 88% in previous pregnancies to 12.5%, and the rate of term deliveries increased from 3% to 87.5%. Ten patients were delivered after 30 weeks’ gestation of healthy infants and one delivered more prematurely. Six deliveries were normal and four required a Caesarean section. We conclude that hysteroscopic metroplasty gives good results. This technique can be used in women with diethylstilboestrol-exposed or hypoplastic malformed uterus, suffering from severe infertility, recurrent pregnancy loss or implantation failures in an in-vitro fertilization programme.

Key words: diethylstilboestrol/hypoplastic uterus/hysteroscopic metroplasty/uterine deformity

Introduction

Hypoplasia and dysmorphism of the uterine cavity are unfavourable factors for fertility and fecundity (Palmer and Meylan, 1964). The aetiology of uterine hypoplasia is generally unclear, apart from in-utero exposure to diethylstilboestrol (DES).

In the past, if these abnormalities were recognized, no treatment could be proposed except for septate or bicornuate uterus. Although under such circumstances conventional metroplasty procedures gave good results (Ayan et al., 1992), hysteroscopic management has more recently become the ‘gold standard’ for resection of the septate uterus. With operative hysteroscopy it is now also possible to achieve an enlargement of the uterine cavity in many uterine hypoplasias. Our first trial was performed in 1992 (Garbin and Dellenbach, 1996), since when 24 women have been treated. Nagel and Malo (1993) reported their experience with a similar technique.

Materials and methods

Patients

Twenty-four women of mean age 30.4 (range 24–41) years had a hypoplastic malformed uterus as seen by hysterosalpingogram (HSG). Of these women, 15 were known to have been exposed to DES in utero.

Among nine patients who suffered from primary infertility, four had unsuccessful assisted-medical procreation which included 14 failures of intrauterine insemination and 10 implantation failures after in-vitro fertilization (IVF).

Fifteen patients had previous pregnancies (Table I). One presented with long-term secondary sterility (despite seven intrauterine inseminations) and 14 had previous pregnancy losses or a history of ectopic pregnancy. This last group totalled 32 pregnancies without any live births. One patient with a previous history of ectopic pregnancy had three implantation failures after IVF. The mean duration of the infertility or infecundity was about five years.

All patients were informed of the nature of the procedure and had given their consent for the operation to be performed.

Surgical procedure: hysteroscopic metroplasty

The principle of the surgical technique, shown schematically in Figure 1, is simple. The pathological shortening of the uterine cavity seems to be linked to an excess of myometrium, particularly in the mid cavity, where it takes the form of subcornual spurs or of a constriction.

Table I. Pregnancies before and after hysteroscopic metroplasty

<table>
<thead>
<tr>
<th>Patients with previous pregnancies</th>
<th>Patients with previous primary infertility</th>
</tr>
</thead>
<tbody>
<tr>
<td>Pregancies before resection</td>
<td>Outcome after resection</td>
</tr>
<tr>
<td></td>
<td></td>
</tr>
<tr>
<td>No. of patients</td>
<td>15</td>
</tr>
<tr>
<td>No. of pregnancies</td>
<td>33</td>
</tr>
<tr>
<td>Spontaneous abortion</td>
<td></td>
</tr>
<tr>
<td>Before 12 weeks</td>
<td>27 (81.8)</td>
</tr>
<tr>
<td>Between 12–26 weeks</td>
<td>2 (6)</td>
</tr>
<tr>
<td>Premature deliveries</td>
<td>0</td>
</tr>
<tr>
<td>Term deliveries</td>
<td>1 (3)</td>
</tr>
<tr>
<td>Death in utero</td>
<td>0</td>
</tr>
<tr>
<td>Ectopic pregnancies</td>
<td>3 (9)</td>
</tr>
</tbody>
</table>

Values in parentheses are percentages.
Figure 1. Principle of the surgical technique. The hysteroscopic metroplasty consists of incising the lateral spurs and an arcuate fundus to obtain an enlargement of uterine size and an improvement of uterine shape.

An arcuate configuration of the fundus frequently increases the uterine dysmorphism and hypoplasia. The surgical procedure consists of restoring a more normal morphology to the uterine cavity with myometrial incisions. The difficulty of the technique lies in the balance between the depth of the incisions and the risk of uterine damage.

As in all operative hysteroscopies, the surgery must be carried out early in the follicular phase, immediately after the menses. We used an operative hysteroscope (nominal diameter 9 mm; actual diameter 7 mm), fitted with a monopolar hook (Resectoscope 7 mm, optical lens 2.9 mm, Ref. 26020FA; Iglesias’s jacket, Ref. 26055 LD; monopolar hook, Ref. 26055 L, Karl Stortz, Tüttlingen, Germany).

To perform the metroplasty, we used a glycocole solution, the flow of which was controlled electronically.

The point of the hook was introduced into the uterine horn and rested on the lateral spur. The incision was performed from the fundus to the isthmus, always under visual control, perpendicularly to the lateral wall of the uterus. Its depth decreased as the section was advanced. Under the effect of the distension, the myometrium was drawn aside progressively. Two or three incisions in the same groove were generally necessary, the same incisions being repeated on the other side. In case of an arcuate configuration of the fundus, we made horizontal incisions, using the blunt edge of the hook. Incisions were made slowly and carefully, especially in the fundus, where they were made with gentle pressure. To ensure security, the depth of incision did not exceed 7 mm.

Although the technique appeared simple, in reality its achievement was difficult. The cavity was confined and renewal of the glycocole was not easy because the suction system was acting against the lateral wall. Thus, the field of view was sometimes poor. However, these problems disappeared when the uterine volume was restored. Straightforward visualization of the ostium uterinum, which often was hidden, provided good anatomic results.

On completion of the procedure, a silastic sheet (Perthése, Laboratoire Perouse Implant, France) was placed to a depth of 1 mm into the uterine cavity. A single dose of second-generation cephalosporin was given at the time of the surgery. Discharge of the patients was authorized on the same day with sequential oestroprogestative medication for 2 months (seven tablets each of 50 µg of ethinyloestradiol and 15 tablets with combined 50 µg ethinyloestradiol and 2.5 mg lynestrol; Ovanon®; Organon SA, Saint-Denis, France).

Figure 2. Preoperative (upper panel) and postoperative (lower panel) hysterosalpingograms. After section of the lateral spurs, the morphology and the size of the uterine cavity are normal.

Results

Anatomical results

In 23 cases (96%), postoperative HSG were well improved, with real enlargement and improvement of the uterine shapes (Figure 2). Two patients required a second procedure.

The final results were considered excellent in 15 cases (63%), with a normal uterine cavity. In the other cases, the result was considered as good with one or two small residual lateral spurs or a residual arcuate configuration of the fundus. Failure occurred in only case (4%) due to severe hypoplasia. No enlargement was observed, though important adhesions
were apparent. The results seemed to be especially good when the preoperative HSG had shown severe dysmorphism. Radiological cervical incompetency was observed in seven cases (29%).

**Functional results**

After surgery, 11 intrauterine pregnancies occurred rapidly. Among these patients, one presented an ectopic pregnancy and two no longer wished to become pregnant. The mean follow-up was 20.3 (range 10–38) months.

Ten patients had 11 intrauterine pregnancies; eight of these were in cases of previous infertility and three in cases of fertility. These pregnancies were spontaneous except in three cases, two after intrauterine insemination and one after induction of ovulation.

Compared with previous pregnancies, the abortion rate fell from 88% to 12.5% and the rate of term deliveries increased from 3% to 87.5% (Table 1).

The outcome of pregnancy was known for all patients. One patient required a cervical cerclage at 24 weeks and two were threatened with premature labour. One patient aborted at 10 weeks and another had a premature delivery at 30 weeks. Six deliveries were normal, and four required a Caesarean section (one for an unexplained acute fetal distress, one for a chorioamnionitis and two were prophylactic for a breech presentation). The uterine cavity was considered as normal in all cases during the Caesarean section. The neonatal courses were good, except in one case with an initial low APGAR score.

**Operative complications**

No immediate complications such as perforation, bleeding or infection were observed. Three adhesions occurred, possibly due to movement of the silastic sheet which no longer fitted the uterine cavity and thus permitted adhesions between the two uterine faces. The first synechiae, at the isthmic level, was easily lysed at the time of diagnostic hysteroscopy. In the second case, adhesions lay on the two lateral walls, where the myometrium had been incised and treatment required a second surgical procedure. The final outcome was good in both cases. In the third patient, who had a severe non-malformed hypoplasia with previous synechias, important adhesions occurred. During her treatment, this patient had undergone three curettages and a trial metroplasty, and declined to undergo the new procedure.

**Discussion**

On the advice of gynaecological specialists, diethylstilboestrol (DES) was prescribed between 1950 and 1977 to treat the threat of abortion. DES was given to some 4 million women in the USA, while in France, approximately 80,000 women were exposed to DES in utero (Pons et al., 1988). Following reports by Herbst and colleagues (Herbst and Scully, 1970; Herbst et al., 1971), attention has been focused initially on the risk of vaginal adenocarcinoma. In fact, this risk is low, at about 1 in 1000. In contrast, abnormalities in the genital tract are frequent and include cervical adenosis and adnexial and uterine abnormalities. Kaufman et al. (1977), after studying 277 HSG, reported uterine abnormalities in 70% of the DES-exposed women, while the incidences of uterine anomalies (Cabeau, 1982) were reported as T configuration and hypoplasia (31%), T configuration (19%) and hypoplastic uterus (13%). In addition, strictures (in particular a constriction ring in the mid uterus), irregular uterine contours, dilatations, adhesions, rudimentary horn and diverticulae can coexist. Many studies, and in particular that of Senekjian et al. (1988), have reported an increase of infertility for women exposed in utero to DES (33% versus 14% for controls) that may be explained in a variety of ways, including cervical sterility, dysovulation, endometriosis and anomalies of the tubes. In the case of uterine dysmorphism, the risk of infertility changes with the type of dysmorphism. For Kaufman et al. (1986), this risk is multiplied by 1.49 in the case of T configuration, by 2.26 in case of mid striction, and by 2.63 if both anomalies are present. The pregnancy outcome in women with a history of DES exposure in utero often appears compromised, with more ectopic pregnancies, abortions and premature labour (Herbst et al., 1981; Pons et al., 1988). Infertility and obstetric complications can be explained by hypoplasia and dysmorphism. It can also be explained by histological or functional alterations of the uterine arteries, as described in velocimetric studies (Salle et al., 1996).

The aetiology of uterine hypoplasia, except in utero exposure to DES, is unclear. Palmer and Meylan (1964) described three types of uterine hypoplasia: simple hypoplasia, where the form of the uterus is normal but the size is small-scaled; elongated hypoplasia, with a narrow fundus but with a normal or elongated length; and malformative hypoplasia, with arcuate fundus, or T- or Y-configurations of the uterus. The latter type appears to be the most unfavourable (Palmer and Meylan, 1964).

Our corrective technique differs from that of Nagel and Malo (1993) on some points. These authors used hysteroscopic scissors, which were considered to be less destructive to the endometrium. We prefer to use a monopolar hook, which is easy to handle, and has a good angle that allows particularly clean incisions. Moreover, the hook tip slips into the uterine horn without difficulty. The laser may certainly be used, as was seen in the resection of the septate uterus (Donnez and Nisolle, 1997). On completion of surgery, Nagel and Malo (1993) used an intracavitary balloon to prevent the occurrence of adhesions. For the same reason, we placed a silastic sheet into the uterine cavity—a technique which has provided good results for many years in ultrasound-guided septum resection (Ohl and Bettahar-Lebugle, 1996). This sheet, being well tolerated, controls endometrial healing in association with oestrogens. Indeed, the three adhesions seen in the present study only occurred when the strip had moved.

Hysteroscopic metroplasty gives good anatomical results in the majority of cases. In the study by Nagel and Malo (1993), the anatomical result was good in 75% of cases (6/8).

In our data, the final result was good, especially when preoperative HSG showed severe dysmorphism and hypoplasia. Symmetrical hypoplasia does not appear to be a good indication.

With regard to functional results, hysteroscopic manage-
ment has already been shown as efficient, in particular for resection of the septate uterus (March and Israel, 1987). In uterine dysmorphism, Nagel and Malo (1993) obtained four pregnancies in women with a history of recurrent pregnancy loss, but none in cases of primary infertility. In our data, pregnancies have occurred in cases of long-unexplained infertility, after implantation failures in IVF and after recurrent pregnancy loss. These results are encouraging, although our sample size is limited and confirmation is required from further studies.

In considering these results, and the fact that the patients exposed in utero to DES have more failures of implantation in an IVF programme (Karabande et al., 1990), and in particular in T-shaped uterus (Noyes et al., 1996), we consider that hysteroscopic metroplasty may be offered to many women, including those with malformative hypoplasia or DES-exposed uterus with severe primary sterility. This would be especially applicable for implantation failures in an IVF programme or in those women with an inexplicable history of recurrent pregnancy loss.

The risk of operative complication still exists but, like Nagel and Malo (1993), we have not observed any. The most important risk is certainly uterine perforation. Preoperative measurement of myometrial thickness using three-dimensional ultrasound scanning provides a good evaluation of the operative possibilities (Figure 3). By restricting the depth of the incisions to 7 mm, using a careful technique, and with two-dimension ultrasound guidance to ascertain myometrial residual thickness, perforation—in particular in the fundus—should be avoided.

It is possible that, following hysteroscopic metroplasty, some complications can occur if the patient becomes pregnant. These include cervical incompetence, uterine rupture and pathology of delivery. Pregnancy in such a uterus must be considered as high risk. Postoperative HSG revealed seven cervical incompetences; one was diagnosed preoperatively. So the hysteroscopic procedure creates a cervical fragilization, even if the responsibility of the technique is not exclusive. In fact, the cervical incompetence of the DES-exposed uterus has been estimated in various ways to range from 4.2% to 17% (Berger and Golstein, 1980; Herbst et al., 1981; Sandberg, 1981). Three patients with postoperative cervical incompetence became pregnant and one required a cervical cerclage at 24 weeks; thus, careful prevention of premature labour is necessary. In the second phase of our study, while using a smaller operative hysteroscope (7 mm), we no longer observed radiological incompetence. Obstetric uterine ruptures after operative hysteroscopy have been described (Tannous et al., 1996), though no uterine rupture arose in the study by Nagel and Malo (1993) or in the present study. Uterine fragility induced by this type of surgery is surely a clear possibility and thus these uteri must be considered as cicatricial. Finally, careful supervision of the delivery and rechecking of the uterine cavity must be carried out.

In conclusion, hysteroscopic metroplasty can correct uterine deformity in the diethylstilboestrol-exposed uterus and hypoplastic malformed uterus and, in general, provides good anatomical results. Pregnancies occurring after the use of this technique must however be considered as high risk, in particular for cervical incompetence. This surgical procedure can be used for women with DES-exposed or hypoplastic malformed uterus suffering from severe infertility, recurrent pregnancy loss or implantation failures after IVF.

References

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