Cervical Shirodkar cerclage may be the treatment modality of choice for cervical pregnancy

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BACKGROUND: Our objective was to evaluate the use of cervical suture in cervical pregnancy. METHODS AND RESULTS: All cases of cervical pregnancy diagnosed and treated in the gynaecological department at the Sheba Medical Center between 1994–2000 were included in the study. Eight such cases were diagnosed. The first four cases were treated medically. The last four cases (the study group) of cervical pregnancy, including one case of heterotopic pregnancy, were treated successfully with placement of Shirodkar cerclage. CONCLUSION: Cervical cerclage may be considered as the treatment of choice in cases of cervical pregnancies. It may be the only therapy in cases of heterotopic pregnancies (intrauterine and cervical pregnancy).

Key words: cervical pregnancy/heterotopic pregnancy/Shirodkar suture

Introduction
Whereas great advances have been made in the diagnosis and treatment of most extrauterine pregnancies, cervical pregnancy remains a challenge. Reportedly occurring in between one per 1000 and one per 18 000 pregnancies (Ratten, 1983; Dicker et al., 1985; Ushakov et al., 1996), cervical pregnancy, even in the new millennium, presents a life-threatening situation, in which delay of diagnosis and treatment can lead to an inevitable hysterectomy.

The risk of maternal mortality due to cervical pregnancy was previously reported to be as high as 40–50%, and in recent years has dropped to 0–6% (Wolcott et al., 1988; Ushakov et al., 1996). However, massive haemorrhage from cervical pregnancy remains a serious cause of maternal mortality. Ushakov et al. have reported an incidence of 29.1% of massive vaginal bleeding at the time of admission to hospital (Ushakov et al., 1996). In only 20.2% of patients was the bleeding described as mild or spotting. Thus, many patients require emergency methods to control bleeding.

The various treatment modalities used for cervical pregnancy include hysterectomy, as well as more conservative approaches such as: curettage and intracervical balloon tamponade (Heller, 1968), curettage and local prostaglandin injection (Spitzer et al., 1997), methotrexate (MTX) administration (Oyer et al., 1988), circumsuture (Scott et al., 1978; Bernstein et al., 1981; Davis et al., 1990), hysteroscopic resection (Ash and Farrell, 1996), bilateral internal iliac ligation (Nelson, 1979), descending uterine artery ligation (Ratten, 1983), selective uterine artery embolization (Simon et al., 1991; Su et al., 1999) and intra-arterial MTX followed by selective prophylactic hypogastric artery embolization (Yitzhak et al., 1999).

We present our experience with a minimally invasive conservative approach, with preservation of the intact uterus, by placement of a Shirodkar cerclage. The purpose of the cerclage was for haemostasis, and while reported in cases in the past (Scott et al., 1978; Bernstein et al., 1981; Davis et al., 1990), this modality has not been recommended as the preferred treatment for cervical pregnancy.

Materials and methods
Between 1994 and 2000, we diagnosed and treated eight cases of cervical pregnancy at the Sheba Medical Center. The study group consisted of the last four patients. The patients’ details are described in Table I. The mean maternal age was 36.7 years, with a range of 25–46 years, and the gravidity ranged from 1–6 pregnancies. Cervical pregnancy was diagnosed by ultrasound. The diagnostic criteria were as follows: a gestational sac embedded in the cervical region, with or without demonstration of a viable fetal pole. Cervical pregnancy was diagnosed by ultrasound. The diagnostic criteria were as follows: a gestational sac embedded in the cervical region, with or without demonstration of a viable fetal pole. Diagnosis was further confirmed by the demonstration of active trophoblastic tissue, with the aid of colour Doppler flow (Su et al., 1999). Sonographic evaluation was performed using HDI 3000 (Advanced Technology Laboratories, Bothell, WA, USA or Synergy, Elscint, Haifa, Israel) with a transvaginal multifrequency transducer (4–8 Mhz).

Prior to this study, cases of cervical pregnancy were treated medically, with MTX, locally or systemically (1 mg/kg on alternate days with folinic acid in between). In the first two cases in the study, massive bleeding developed after MTX administration, and an emergency Shirodkar cerclage (Shirodkar, 1955) was placed with concomitant aspiration of the intracervical tissue. The cerclage was...
Table I. Cervical suture – patients details

<table>
<thead>
<tr>
<th>Case</th>
<th>Age (years)</th>
<th>Obstetric and past history</th>
<th>Present pregnancy</th>
<th>Gestational age (weeks)</th>
<th>Ultrasound findings</th>
</tr>
</thead>
<tbody>
<tr>
<td>1</td>
<td>25</td>
<td>G1</td>
<td>Spontaneous</td>
<td>6 + 2</td>
<td>Alive cervical pregnancy</td>
</tr>
<tr>
<td>2</td>
<td>42</td>
<td>G3P2A1</td>
<td>IVF (ZIFT)</td>
<td>6 + 2</td>
<td>Alive cervical pregnancy</td>
</tr>
<tr>
<td>3</td>
<td>46</td>
<td>G0</td>
<td>IVF</td>
<td>6 + 3</td>
<td>Alive cervical pregnancy</td>
</tr>
<tr>
<td>4</td>
<td>34</td>
<td>G6P1A4 – s/p salpingectomy s/p cervical suture</td>
<td>IVF</td>
<td>8 + 3</td>
<td>Heterotopic pregnancy – alive intrauterine + cervical pregnancy</td>
</tr>
</tbody>
</table>

Table II. Course and outcome of pregnancies

<table>
<thead>
<tr>
<th>Case</th>
<th>Initial HCG (mIU/ml)</th>
<th>Day of cerclage</th>
<th>HCG on day of cerclage (mIU/ml)</th>
<th>HCG on day of discharge (mIU/ml)</th>
<th>Course and outcome</th>
</tr>
</thead>
<tbody>
<tr>
<td>1</td>
<td>2351</td>
<td>8</td>
<td>8041</td>
<td>566</td>
<td>Failure of MTX Tx bleeding; cerclage – normal outcome</td>
</tr>
<tr>
<td>2</td>
<td>9200</td>
<td>6</td>
<td>32 350</td>
<td>800</td>
<td>Same as case 1</td>
</tr>
<tr>
<td>3</td>
<td>9574</td>
<td>3</td>
<td>19 200</td>
<td>1606</td>
<td>Normal outcome</td>
</tr>
<tr>
<td>4</td>
<td></td>
<td></td>
<td></td>
<td></td>
<td>Heterotopic pregnancy; NVD (39 weeks gestation)</td>
</tr>
</tbody>
</table>

Tx = therapy; NVD = normal vaginal delivery; MTX = methotrexate.
Day of cerclage = day of hospitalization; normal outcome = resolution and disappearance of the cervical pregnancy.

Figure 1. Sagittal view of a heterotopic pregnancy at the age of 8 weeks 3 days—note the intracervical gestational sac along with an intrauterine sac containing a viable embryo and a yolk sac.

performed electively in the two subsequent cases. Shirodkar cerclage was chosen over the McDonald suture due to its placement in a higher location on the cervix in order to suture the gestational sac as much as possible and thus achieving better haemostasis.

Results

Detailed results are presented in Tables I and II. Of the four patients treated by Shirodkar cerclage placement, only one patient conceived spontaneously while the rest conceived by assisted reproduction treatment. The fourth patient presented with heterotopic pregnancy (Figure 1) and a diverse previous history of laparoscopic salpingectomy for tubal pregnancy, four habitual abortions and one term vaginal delivery with cervical suture. The Shirodkar cerclage was performed as an emergency salvage procedure in two cases due to massive bleeding after MTX administration. The procedure was subsequently electively performed in cases 3 and 4. All four cases were successfully treated by this modality, and no further medical or surgical intervention was necessary.

Mean HCG levels at the time of cervical placement were 19 864 mIU/ml (range 8041–32 350) and at the time of discharge dropped to a mean level of 990 (range 566–1606 mIU/ml). (These values do not include the fourth, heterotopic, case.) The mean length of hospitalization following cerclage was 6 days (range 5–9 days).

Case number 4, the heterotopic pregnancy, ended in normal vaginal delivery at 39 weeks gestation of a male infant, weighing 3010 g, with an Apgar score of 9 and 10 in 1 and 5 min post-partum respectively. Figure 2 demonstrates the intrauterine pregnancy and the cervical suture, 1 month after performance of the Shirodkar cerclage.

Discussion

The reported incidence of cervical pregnancy varies between one in 1000 to one in 18 000 pregnancies and it accounts for <1% of all ectopic gestations (Ratten, 1983; Dicker et al., 1985; Ushakov et al., 1996). However, it is thought to be on the rise due to the introduction of assisted reproductive techniques and improved diagnostic modalities (Ushakov et al., 1996), and indeed, in our series of eight cervical pregnancies, performed electively in the two subsequent cases. Shirodkar cerclage was chosen over the McDonald suture due to its placement in a higher location on the cervix in order to suture the gestational sac as much as possible and thus achieving better haemostasis.

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Various treatment modalities have been applied for the treatment of cervical pregnancies. For many years, hysterectomy was the only option available to alleviate the massive bleeding and save the woman’s life. As modern developments in diagnostic techniques have made diagnosis possible at an earlier gestational age, conservative techniques have been introduced, with MTX administration gaining more and more popularity due to the high rate of success and the avoidance of invasive procedures.

Scott *et al.* first reported the successful application of Shirodkar cervical cerclage in cervical pregnancy (*Scott et al.*, 1978). However, in the reported case, the diagnosis of cervical pregnancy was based upon the manual digital examination, and was not supported sonographically. The procedure was performed on an emergency basis after the failure of curettage, and was concomitant to the placement of intra-cervical obturator. Bernstein also reported this procedure (*Bernstein et al.*, 1981). Again, the diagnosis was done clinically and the McDonald cerclage was placed as an emergency method before planned hysterectomy. The same procedure was described in a heterotopic pregnancy (*Davis et al.*, 1990), again as a method to control bleeding. To the best of our knowledge, this procedure, especially with the Shirodkar suture, has not gained acceptance as an elective modality of treatment of cervical pregnancy.

MTX treatment of cervical pregnancy was first reported in 1988 (*Oyer et al.*, 1988) and since then it has gained widespread acceptance. Indeed, in the four cases that presented to our department between 1994 and 1999, MTX administration (either intracervical or systemic) was the treatment of choice, and all four cases resolved successfully. However, the next two cases (first patients in this study) failed to respond to MTX therapy, beta levels did not recede and massive cervical bleeding ensued. Prior to offering the patients a hysterectomy, as a life saving procedure, the option of placing a cervical suture as a hemostatic procedure, together with aspiration of the cervical pregnancy, was suggested. The patients agreed to undergo this new procedure, and their recovery was uneventful. In the next case (number 3), which presented at 6 + 3 weeks gestation with vaginal bleeding with diagnosis of viable cervical pregnancy, the placement of Shirodkar suture (along with aspiration) was offered as the first option and performed successfully. Patient number 4 presented with a heterotopic pregnancy, a normal intrauterine 8 week pregnancy, as well as a cervical pregnancy (Figure 1). Naturally, MTX therapy could not be offered and she too was treated by cervical suture, successfully.

Heterotopic pregnancies, i.e. combined intra- and extrauterine pregnancy, occur rarely among spontaneous pregnancies, with an estimated rate of 1 in 30 000 pregnancies (*Bello et al.*, 1986; *Bearman et al.*, 1986). However, among patients who have undergone IVF therapy, the incidence is much higher, approaching 1% (*Bearman et al.*, 1986; *Dor et al.*, 1991; *Goldman et al.*, 1992). Combined intrauterine and cervical pregnancy is even rarer. As more women with a past history of recurrent abortions, as well as cervical interventions such as dilatation and curettage and cerclage, utilize assisted reproductive techniques, the rate of heterotopic cervical pregnancy will undoubtedly rise (*Ushakov et al.*, 1996). *Carreno et al.* have described treatment of heterotopic cervical pregnancy with reduction of the cervical pregnancy with the aid of saline injection into the gestational sac in the cervix (*Carreno et al.*, 2000). Although the outcome was favourable, we think this method carries a great risk of haemorrhage and therefore suggest our method should be considered the treatment of choice in these cases. *Peleg et al.* have also described conservative treatment of combined intrauterine and cervical pregnancy.
(Peleg et al., 1994). However, they have used intra-arterial MTX, which did not preserve the intrauterine pregnancy.

In our opinion, this new modality, i.e. placing a cervical Shirodkar cerclage as treatment of cervical pregnancy, has many advantages: (i) control of the massive bleeding created by the invasion of the cervical tissue by trophoblast, thus alleviating the need for hysterectomy and/or the need of transfusion of blood products. The Shirodkar suture, placed higher in the cervix, can contribute to better haemostasis; (ii) avoidance of the systemic side-effects of parenteral MTX administration such as nausea, vomiting, hepatic disorders, stomatitis and decreased white blood cell counts (Kojima et al., 1990); (iii) safety and applicability in cases of heterotopic pregnancy with an intact intrauterine pregnancy—systemic therapy cannot be applied, and the cerclage allows continuation of the intrauterine pregnancy; (iv) another possible advantage of this treatment modality is a quicker response – rapid decline of HCG levels. Whereas in MTX therapy, it can take between 5 and 60 days (Kojima et al., 1990), in our small series of three cases (the heterotopic pregnancy was not included), HCG levels dropped substantially within 5–6 days and returned to normal over a mean period of 21 days. The mean hospitalization time in the study group (6 days, with a range of 5–9 days) is also much shorter than mean time in the patients treated with MTX only in our department (14 days). One must, of course, bear in mind the possible disadvantages of Shirodkar cervical cerclage. The procedure takes place under general anaesthesia and it can be associated with complications (such as haemorrhage and bladder injury), especially in the hands of inexperienced physicians. Our patients did not suffer from any of those complications.

In summary, we have shown here our experience of treatment of cervical pregnancy with the use of Shirodkar cervical suture. By serving as a useful haemostatic technique, this therapy should be offered, in our opinion, as an alternative treatment in cervical pregnancy. It should not serve only as an emergency option when massive bleeding occurs after failure of other treatment modalities, but as a primary option, thus reducing the rate of inevitable hysterectomy, the need for blood transfusions and preservation of fertility. This approach will also avoid the systemic side effects of MTX, shortens the hospitalization period and should be the only treatment option in heterotopic pregnancy. More experience is needed in order to validate our preliminary results, although the incidence of cervical pregnancy makes this difficult.

References

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