CASE REPORT

Mucocele formation 20 years after an appendiceal uterine transplantation for infertility mistaken for hydrops tubae profluens

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A 56 year old woman was admitted to our hospital with a 9-year history of recurrent, lower abdominal pain and mucoid vaginal discharge 20 years after an appendiceal uterine transplantation. The removal of the uterus and the attached appendix resulted in the disappearance of the symptoms. A mechanism linking the appendiceal mucoid discharge with abdominal pain in this menopausal patient is suggested.

Key words: appendiceal uterine transplantation/late complications/mucocele

Introduction

Pathological alterations of the Fallopian tube are frequent factors that influence infertility. The era prior to in-vitro fertilization (IVF) was characterized by multiple surgical procedures to establish tubal patency. Although postoperative patency rates have been reported in the range of 70-90%, the term ‘pregnancy rate’ has been disappointingly low—between 0 and 48% (Rock, 1985). These procedures offered no solution to totally destroyed or missing tubes for reconstruction of a lumen to re-establish tubal patency. Among the tissues used as a substitute for the oviduct were the ileum and the appendix (Charles et al., 1962; O’Neill, 1966; Halelamira et al., 1968a; Halelamira et al., 1968b). The appendix was thought by some to be a reasonable substitute because of its blood supply, wall muscularity, motility, columnar epithelium and slight alkalinity that resembled the oviduct (O’Neill, 1966; Halelamira et al., 1968a). Even though we found two case reports of appendiceal uterine transplantation, only one anecdotal case of successful pregnancy was reported (Halelamira et al., 1968b). Review of the case disclosed that during the procedure one oviduct remained and there was no proof that fertilization took place on the appendiceal side. The procedure was abandoned.

We present a case of late sequelae of such a procedure, with recurrent abdominal pain and cervical mucoid discharge 21 years after appendiceal uterine transplantation

Case report

A 56 year old woman presented at our clinic because of recurrent episodes of abdominal cramp, followed by vaginal mucoid discharge for the previous 9 years. Thirty-five years earlier she had had a successful pregnancy and delivery. During the next 5 years she had three ectopic pregnancies that ended with bilateral salpingectomy. Twenty-one years ago she was offered the procedure of appendiceal uterine transplantation. The procedure was performed using the technique reported by O’Neill (1966). The post-operative course was uneventful. The patient did not conceive again and did not recall the operation.

The patient had been menopausal for the previous 9 years. Over the same period she started to experience episodes of lower abdominal cramps every few weeks. These episodes of abdominal pain and cervical mucoid discharge were recurrent and continued for the previous 9 years.

Figure 1. The junction between the uterus (left) and the appendix (right). Note inflammatory process of the endometrium and the intestinal mucosa (H&E, X 10).
lasted for 2–3 days, and ended with the appearance of a mucoid vaginal discharge. Ultrasonography showed a small uterus, with fluid in the cavity. An attempt at endometrial biopsy and fractionated dilation and curettage (D&C) yielded only mucoid fluid without pathological tissue. After the cervical dilatation there were no more episodes of abdominal pain, but she complained of a continuous vaginal discharge of large volumes of mucoid fluid. The mucoid watery vaginal discharge was suggestive of tubal pathology and surgery was indicated. On laparotomy, a thickened appendix was noted to be connected to the right horn of the uterus, with its distal end adhering to the pelvic wall. The Fallopian tubes were absent. A total hysterectomy was performed, including removal of the appendix. The post-operative course was unremarkable, and the patient is now free of abdominal pain or vaginal discharge.

Pathological examination revealed a 7 × 5 × 2.5 cm uterus with the appendix attached to its right horn. Histological examination showed a uterus with an inactive endometrium, and areas of chronic and subacute endometritis. The appendix exhibited inflammatory infiltrate in the lamina propria (Figure 1). Injection of contrast material through the cervix showed the patency between the uterine cavity and the appendiceal lumen.

Discussion

Since the first report of Schroder in 1884 of a unilateral ampullary cuff salpingostomy, there have been multiple surgical procedures to obtain oviduct potency (Rock, 1985). The main purpose of these procedures was to achieve a patent lumen between the ovary and the uterine cavity. The poor results using tuboplasty led to a search for alternatives to the damaged or missing Fallopian tubes. Use of ileum for oviducts was reported in dogs (Charles et al., 1962). O'Neill (1966) and Halelamira et al. (1968a) reported on transplants of the appendix as a tube, and showed patency of the appendiceal lumen by hysterosalpingogram. Later, Halelamira et al. (1968b) described a patient who conceived and delivered successfully, although there was no proof that the fertilization took place in the transplanted appendix. It was later observed that the oviduct is not a simple, inert tube, and that the lymphatic nodules, the non-ciliated epithelium or the mucoid secretions of the appendix could be harmful to the ova or spermatozoa. All these procedures were abandoned with the development of microsurgical techniques in tuboplasty. Today, IVF has revolutionized our approach to the treatment of the infertile couple, and most of the reconstruction procedures of the Fallopian tube have become merely of historical interest.

Although rare, reconstructive procedures may become clinically important when considering complications. Possible sequelæ may appear many years after the initial operation, which, in our patient, had even been forgotten. Moreover, today's physicians are not familiar with procedures such as appendiceal uterine transplantation.

In the present case, the lumen patency from the appendix to the uterus caused the symptoms. The appendix secreted the usual mucoid fluid, which was probably intensified by the inflammatory process. After menopause, the cervix became stenotic and the fluid accumulated in the uterine cavity, forming a mucocele as was demonstrated by ultrasonography. The distended uterus caused by the fluid probably evoked the episodes of abdominal pain that ended when the intrauterine pressure rose to cause drainage of the fluid. Dilatation of the cervix released the cervical obstruction, with continuous fluid drainage. The final treatment was the removal of both the uterus and the attached appendix.

The importance of the reported case is to note the additional differential diagnosis to the complex of hydrops tubae profuens. This condition is almost pathogenomic for the rare malignancy of the Fallopian tube (3.6/1 000 000 women/year) (Disaia and Creasman, 1993). In this age group, we may find some patients today who had had an appendiceal uterine transplantation many years previously, and now have a better prognosis than the former cases. To our knowledge, this is the first report of a case of this type.

References


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