Carcinoma of the cervical stump. A case report and literature review

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Abstract

Subtotal hysterectomy for benign uterine disease was a common surgical procedure in the past decades. Keeping in place the uterine cervix predisposes to late complications such as cervico-vaginal prolapse, chronic cervicitis or even malignant transformations of the cervical stump. We present the case of a 56 year old patient who was submitted to subtotal hysterectomy for a large uterine fibroma 4 years before and who presented vaginal bleeding and pelvic pain. She was diagnosed with cervical stump carcinoma and a radical resection including trachelectomy with pelvic and para-aortic lymph node dissection was performed. The histopathological studies revealed a well differentiated squamous cell cervical cancer. **Keywords:** subtotal hysterectomy, cervical stump malignancy, radical resection.

Introduction

Subtotal hysterectomy for benign conditions had been largely performed since the early 1950's with decreased risks of intra-operative complications, good results in terms of early postoperative evolution⁽¹⁾. The main indications to perform a subtotal hysterectomy are uterine fibromas not suitable for conservative surgery (myomectomy) or postpartum hemorrhage⁽²⁾. Keeping the uterine cervix in place requires an adequate follow up for the cervix as part of screening for cervical cancer⁽²⁾. The reported incidence of cervical stump cancer widely varies between 2-6% in Jeffcoate's study conducted in USA⁽¹⁾ and up to 46% in Christ's study conducted in Germany⁽³⁾.

Case report

A 56 year old patient diagnosed with a uterine fibroma four years before who was submitted at that moment to subtotal hysterectomy presented for diffuse pelvic pain and massive vaginal bleeding. The local examination revealed a cervical tumor while the biopsy confirmed the presence of a well differentiated squamous cell carcinoma. Pelvic magnetic resonance imaging confirmed the presence of a cervical tumor with left parametrium invasion. She was submitted to neo-adjuvant radiation therapy and six weeks after completion of radiotherapy surgery was performed. A radical resection including trachelectomy, pelvic and para-aortic lymph node dissection was performed (Figures 1-8). The postoperative course was uneventful. The histopatological studies of the specimen confirmed the results of the preoperative biopsy.

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Discussion

Most studies concluded that malignancies of the cervical stump are defined to be all the malignancies which are diagnosed after an interval of at least three years after performing a supra-cervical hysterectomy. However, although most authors approve this 3 years interval, the largest series reported by Gibbons and Keys consider that a 'true' carcinoma of the cervical stump is that which arises more than two years after subtotal hysterectomy⁽⁴⁾.

The main symptoms associated with cervical stump malignant degeneration are vaginal bleedings. However, this might not be always associated with the presence of cervical cancer. In cases presenting retained endometrium and ovaries preservation, vaginal bleeding has no pathological significations. Christ et al. reported that 36% of patients included in their study presented for vaginal bleeding after subtotal hysterectomy and only 56% of them were diagnosed with malignant changes⁽³⁾.

More recent studies reported that about 4.5%-9.4% of patients diagnosed with cervical malignancies had been previously submitted to subtotal or supra-fascial hysterectomy. The same authors showed that although patients with carcinomas of the cervical stump have been on average a decade older when compared to those who hadn't been submitted to other uterine surgical procedures, the distribution according to the histopathological type was similar to those with no previous surgery (up to 90% of both cathegories developing a squamous cell carcinoma)⁽⁵⁻¹⁰⁾.

Hellstrom and contributors⁽¹¹⁾ retrospectively reviewed data of patients diagnosed with cervical cancer upon fifty years in Karoloinska Institute, Stockholm. From 8028 women diagnosed with cervical cancer, 161 patients (2%) were diagnosed with cervical stump cancer. For each patient with cervical stump cancer three control cases were selected and matched according to age, stage and histopathology. Mean time between subtotal hysterectomy and cervical stump cancer diagnosis was 17 years while mean age of patients at the moment of diagnosis of malignancy was 60 years.



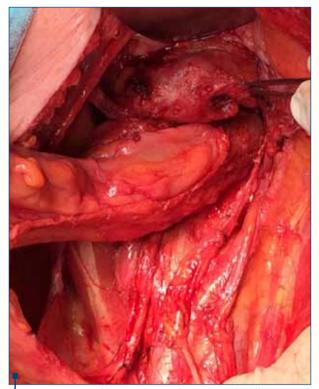


Figure 1. Dissection of the cervical stump after previous subtotal hysterectomy

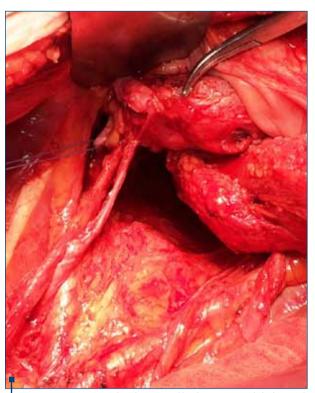


Figure 2. Uterine artery enlaced; ureter and lombo-ovarian pedicle dissected



Figure 3. Dissection of the two ureters

Thirty-one patients were diagnosed in advanced stages of the disease (23 women were diagnosed in FIGO stage III while 8 patients were diagnosed in FIGO stage IV). All patients were submitted to neo-adjuvant irradiation consisting in 2 brachytherapy sequences at a three week interval followed by external radiation therapy. Surgery consisted in cervicectomy and pelvic lymph node dissection. This study concluded that a



Figure 4. Dissection of the vagina and cervical tumor

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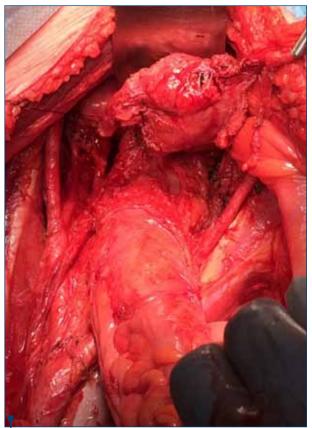


Figure 5. The final aspect after mobilization and pelvic lymph node dissection

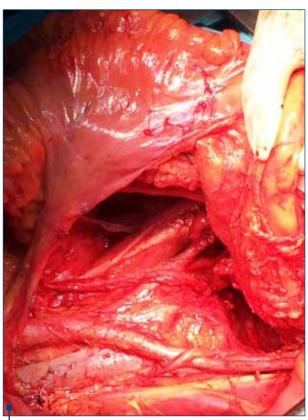


Figure 6. The para-aortic lymph node dissection was performed up to the level of the inferior mesenteric artery

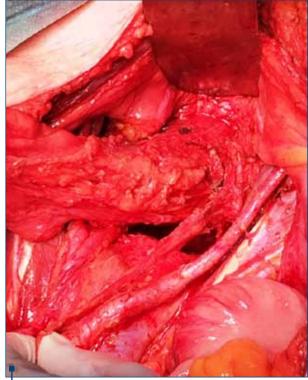


Figure 7. The final aspect after resection



Figure 8. The specimen presenting a hemorrhagic cervical tumor

correlation between subtotal hysterectomy and cervical stump cancer cannot be established due to the long period of latency between the two events. The same study also concluded that patients with stump cancer are more likely to be diagnosed in an advanced stage of the disease. This fact was explained by the fact that many patients didn't receive complete information regarding the initial performed surgery (i.e. the exact extent of the hysterectomy) and it was erroneously considered that a screening Pap smear test is no longer needed. However, even in cases who were submitted to Pap test the disease was diagnosed in more advanced stages when compared to those with no prior uterine surgery, concluding that Pap test might be less efficient in these cases. Diagnosis in a more advanced stage of the disease was also responsible for the poorer outcome of patients with stump cancer⁽¹¹⁾.

When it comes to the most appropriate therapeutic option, most studies showed that neo-adjuvant radiation therapy plays an important role. However, a lower volume of irradiation should be applied due to the absence of the uterine fundus and secondarily, a higher impact on the small bowel. Miller and contributors reported that patients with previous resections of the uterine fundus should be submitted to less than 50 Gy of external beam therapy associated with a transvaginal cone dose of 15 Gy⁽⁵⁾. Kovalic et al. also showed that prolonged external beam therapy is usually associated with a poorer evolution and prognostic (9). Severe side effects of radiation therapy in cases presenting previous uterine resections are related not only to the absence of the uterine fundus. Other factors which seem to interfere are the presence of previous episodes of pelvic inflammatory disease which increase the risk of small bowel pelvic adhesions or the modifications of the cervical stump which might appear shortened or deformed^(4,5). In all these cases both intra-cavitary and external beam radiation therapy should be careful performed. In Miller's study, which was conducted on 263 patients with cervical stump malignancies up to 30% of cases submitted to radiation therapy developed gastro-intestinal side effects such as gastro-intestinal toxicities (i.e. nausea, vomiting, diarrhea) or even more aggressive reactions such as bowel obstruction, hematuria, rectal bleeding or fistulas (reported in up to 9% of cases)⁽⁵⁾. Barillot et al. reported a 13% incidence of grade 3-4 complications in patients submitted to neo-adjuvant radiation therapy for both cervical stump cancer and intact cervix cancer. The same study concluded that the incidence of lethal complications was ten times higher (6% for those with cervical stump cancer and only 0.6% for those with intact cervix) and recommended that in cases with cervical stump malignancies a combined therapeutic protocol should be performed⁽⁷⁾.

After ending neo-adjuvant irradiation, surgery must be performed. However in all these cases surgery might be a demanding procedure due to the adhesion formation, fibrotic changes related to radiation therapy or other anatomic modifications. Other factors associated with a poorer oncological outcome are large volume tumors and high grade tumors^(2,7,9).

Conclusions

Although subtotal hysterectomy was considered long time as an effective procedure associated with shorter time of surgery and less intraoperative blood loss, preserving the uterine cervix necessitates screening for malignant degeneration. Even in cases submitted to regular cervical cancer screening test, neoplasia can be detected in a more advanced stage. Whenever cervical cancer stump is diagnosed, neo-adjuvant radiation therapy including both external beam therapy and brachytherapy should be performed. However irradiation doses should be adapted to the subsequent pelvic modifications. Radical surgery after irradiation should also be taken in consideration especially in cases presenting severe side effects of irradiation. In conclusion, these cases represent challenging cases for surgeons too, due to the local anatomic modifications related to the prior performed surgeries, adhesions or fibrotic changes associated with irradiation.

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