

Didelphys Uterus and Cervical Cancer: A Case Report and Review of Literature

Dr. Victor E. Valdespino¹

¹ Social Security Mexican Institute

Received: 16 December 2017 Accepted: 1 January 2018 Published: 15 January 2018

Abstract

Congenital malformations of the female genital tract are defined as deviations from normal anatomy resulting from embryological maldevelopment of the Müllerian or paramesonephric ducts. This condition represents a rather common benign condition with a prevalence of 4-7

Index terms—

1 Didelphys Uterus and Cervical Cancer: A Case

Report and Review of Literature Abstract—Congenital malformations of the female genital tract are defined as deviations from normal anatomy resulting from embryological maldevelopment of the Müllerian or paramesonephric ducts. This condition represents a rather common benign condition with a prevalence of 4-7%. Cervical cancer and didelphys uterus is an infrequent condition in clinical practice. Association between cervical cancer and Müllerian malformation is limited to medical references. We present a surgical treatment with a result IB1, with systematic pelvic and paraaortic nodal dissection, with poor prognostic factors, she is chemoradiotherapy treatment. She is a patient 55 years old, with no symptoms in young adulthood or teenager in relation to didelphys uterus.

Always it is possible we encourage the primary surgical treatment, we can get prognostic factors and is possible scan other congenital malformation, also the point A is not constant for planned a radiotherapy treatment finally lymphatic channels in anatomical distortion could be evaluated and measure the nodal affection, and improve and personalize radiotherapy treatment. This case is an absolutely infrequent in the clinical practice.

2 I. Introduction

cervical cancer is typically preventable if precancerous lesions are detected and treated early. Cervical cancer screening by means of cytology, or the Papanicolaou smear, seeks to detect precancerous or cancerous cervical lesions prior to symptom on set. Research has consistently observed that cervical cytology screening is highly efficacious against invasive cervical cancer incidence and death among women of reproductive age 1. Therefore, regular cervical cancer screening and follow-up are critical.

Cytological screening will most likely decline in favor of HPV-based screening because of its superiority over cytology in the 2 characteristics that influence test efficacy; HPV DNA testing can detect invasive cervical cancer risk for a longer period than cytology (2,3), and its sensitivity is an absolute 40% higher than that of cytology (4,5). Thus, the relationship between these screening modalities efficacies is knowable-the efficacy of HPV-based screening is expected to exceed that of cytology, all things being equal. Analysis of extant data on cytology screening, therefore, may offer a minimum estimate of HPV-based screening efficacy among older women. However, screening by cytology alone remains acceptable under all current guidelines, and Papanicolaou smears continue to be widely used. Further, a study to evaluate the efficacy of HPV DNA testing among women will not be possible for years after an HPV DNA-based screening program is implemented until a sufficient number of deaths have occurred to make meaningful comparisons on the basis of prior HPV DNA screening history.6, 7.

Müllerian duct anomalies are congenital defects of the female genital system that arise from abnormal embryological development of the Müllerian ducts. These abnormalities can include failure of development,

fusion, canalization, or reabsorption, which normally occurs between 6 and 22 weeks in utero. Most sources estimate an incidence of these abnormalities to be from 0.5 to 5.0% in the general population 8,9

Septate uterus is the commonest uterine anomaly with a mean incidence of 35% followed by bicornuate uterus (25%) and arcuate uterus (20%) 9

Unicornuate and didelphys uterus have term delivery rates of 45%, and the pregnancy outcome of patients with untreated bicornuate and septate uterus is also poor with term delivery rates of only 40%. 9

Most women with a didelphys uterus are asymptomatic, but some present with dyspareunia or dysmenorrhea in the presence of a varying degree of longitudinal vaginal septum. Rarely, genital neoplasms, hematocolpos, hematometocolpos, and renal anomalies are reported in association with didelphys uterus. Despite some of these complications, there are many cases of women with a didelphys uterus that did not exhibit any reproductive or gestational challenges.

The VCUAM classification (Vagina, Cervix, Uterus Adnex Associated Malformation) is anatomical. Organs are classified as separated similar to TNM classification, (tumor, nodal, metastases). This manner allows a categorization, is precise, detail, and very representative. Different anatomical anomalies could be described and the practitioner has a good idea of each organ is affected in a single manner.10.

Lee reports a case of a congenital abnormality of uterus didelphys in a patient who developed invasive carcinoma of the cervix. The patient received radical radiotherapy by a combination of external beam pelvic radiotherapy and high dose rate brachytherapy by insertion of afterloading catheters into both uterine canals. A newly defined prescription point was used midway between the two catheters and 2 cm above the mean cervical position. The classical point A was regarded as inappropriate in this patient with a rare condition. Acute toxicity was minor and the patient is tumor free with no significant normal tissue late effects after follow-up of nearly 3 years.

Depends on main cervical tumor is localized, the classical point A, could change, in position, in consequence, the radiotherapy treatment should be personalized and very precise for a better response on the tumor. 11

In addition, we can consider cervical cancer in a patient with Mullerian anomalies, we must offer the best treatment option, it is possible to get the nodal status, by lymphadenectomy or radical surgery by laparoscopic surgery or traditional surgery, when the stage allow it, or chemoradiotherapy.

When the cervical cancer is treated with surgery, we choose a specific surgery with a Querlow -Morrow hysterectomy, the patient does not need more morbidity with the greatest surgery, in our clinical practice when we performed a hysterectomy control, we always practice standing nodal affection pelvic and paraaortic lymphadenectomy, and we can get specific information about the nodal tumoral invasion, it is necessary specific adjuvant treatment.

3 II. Case Report

The present case is a women 55 years old, with hypertension 12 years of history, cholecystectomy at 32 years old, no more familiar background, gynecological antecedent menarche 12 years old, 28 x 5 days, 4 pregnancies, 1 labour, 3 caesarean, menopause 50 year old. In a yearly control cervicovaginal cytology reported an epithelial neoplasia grade II, in the medical first level unit, the patient was sent to colposcopy in a third level medical unit, in this evaluation (colposcopy) they notice two cervixes, one of them with cervical cancer (right) and left cervix without tumoral damage. A curettage endocervical was performed in both cervix, squamous invasive cancer was reported on the right cervix, endocervical glands without alterations on the left cervix. Colposcopy service, operate a conization on right cervix with definitive report squamous cell carcinoma measure 0.8 x 0.5 cm margin was positive an invasive tumor. An ultrasound was made, cervix reported 32 x 26 x 30 mm no tumor was obvious, uterine corpus 46 x 48 x 20 mm and we performed a hysterectomy Querlow -Morrow B2 on right side and Querlow -Morrow A on left side, we carry on a systematic lymphadenectomy pelvic and paraaortic with 17 nodes without tumor in pelvis and 24 nodes without metastases in retroperitoneal area. 12. The final tumoral measure was 27 mm, tumoral get involvement all right cervix, with lymph-vascular infiltration, and tumor comprises lower uterine segment. Surgical stage final was IB1 epidermoid cervical right cancer. The left cervix does not expose a tumoral injury, including no cervical dysplasia. At the moment of transoperative, we found a double uterine body, in a relationship with double cervix we achievement, a didelphys uterus and cervical cancer. The patient suffers a ureteral leak, it was resolved with a catheter JJ, she was sent to radiotherapy and chemotherapy, she is on concomitant treatment right now with good tolerance.

In the current clinical practice, this association between uterus didelphys and cancer are very rare, we performed a surgery a Querlow-Morroe B2 in right side and a Querlow-Morroe A in the left side also pelvic and paraaortic lymphadenectomy. 12 As Chiappa and coworkers, we improve our clinical point of view with a cervical ultrasound this value measure, is extraordinarily helpful because improve our clinical diagnosis, and we performed this as a routine in our service when a patient will be programmed for a surgery or chemoradiotherapy by cervical cancer. 13 In addition cervical cancer in a didelphys uterus is absolutely infrequent even in historical technical literature do not is mentioned technical change performing a hysterectomy, just is refer briefly to get free neoplastic margin.14 Rarely, cervical cancer and endometrial carcinoma are reported in association with cases of didelphys uterus 15. 16 Most women with a didelphys uterus are asymptomatic but may present with dyspareunia or dysmenorrhea in the presence of a thick, sometimes obstructing vaginal septum. This obstructing vaginal septum can lead to

hematocolpos/hematometocolpos and thus present as chronic abdominal pain as well. Or some problems if the patient desire a pregnant.

In the present report, the patient has no knowledge about dydelphys uterus because she has no problems at reproductive age and develops 4 pregnancies with successful evolution. Previously at his childhood and teenager, she does not refer chronic pelvic pain or sexual discomfort in early adulthood. This does not agree with medical reports.

It is generally accepted that having a uterine anomaly is associated with poorer pregnancy outcomes such as increased chances of spontaneous abortion, premature labor, cesarean delivery due to breech presentation, and decreased live births, compared to a normal uterus. However in the present report could get 4 pregnant, with 1 labours delivery and 3 cesarian. 8 The modalities for correct diagnosis frequently used include highly invasive methods such as hysteroscopy, hysterosalpingography, and laparoscopy/ laparotomy, also ultrasound. 3D ultrasound is becoming more commonly used for diagnosis as it is not only noninvasive, this analytic tool gives all the information needed for morphological classification 10,17. Magnetic resonance imaging is also just as accurate and valuable in diagnosing müllerian abnormalities, as hysterosalpingograms, hysteroscopy, and laparoscopy are, even more so as it is noninvasive and can diagnose associated urinary tract abnormalities at the same time 13. Nonetheless, it is still difficult to distinguish between these different anomalies on imaging modalities due to subjectivity; differences in morphology are often subtle and changing classification systems. 17

In opposition to the medical reports, this patient was diagnosed until medical assistance on cancer standing; colposcopy and ultrasonography evaluation. 18 Other malignant tumors have been reported in Muellerian anomalies, as Iavazzo, reported a case on didelphys uterus an uterine carcinosarcoma. 19 Present case report presents an IB1 cervical cancer with nodal evaluation pelvic and retroperitoneal negative, why a cervix develops cervical cancer and others do not develop any malignant or premalignant injury we can not answer this question, maybe by epigenetic changes because the viral exposition was positive on both cervix.

Sugimori, reported two cases of cervical cancer in uterus didelphys. One was extensive adenocarcinoma and one was squamous cell carcinoma in situ. 20

4 IV. Conclusion

If a patient has a Müllerian duct anomalies and cervical cancer, clinical staging can be ambiguous, the natural history may be altered, also common association with renal agenesis, or other anatomical variation. Some treatments which could influence the use of potentially nephrotoxic agents, like cisplatin, then are a part of standard chemoradiotherapy, must be considered at moment on select a therapy.

Treatment decision making needs to be precise and personalized, in view of the minimal amount of prior literature on the topic.

Applicator placement for intracavitary brachytherapy may be fraught with this patients. Because inability to define a point A in patients with anomalies featuring double cervix and uterus is a challenge. Is very useful the surgical approach because we can get prognostic factors, and real pathology stage and another abnormal anatomical variation could be evident and to be evaluated. 21 That's why always it is possible the patients must be treated with surgery the local (pelvic) disease and lymphatic nodes and retroperitoneal, because no available literature to describe the lymphatics of the various Müllerian ducts anomalies. In fact, we recommend performing a lymphatic node dissection pelvic and retroperitoneal in stage IIB or advanced, and know the specific node pathological of the disease and improve radiotherapy field treatment. 22 Among patient with cervical cancer who have Mullerian anomalies, radical surgery should be selected over radiotherapy in the early operable stages. Surgery provides a real stage for nodal metastases pelvic and retroperitoneal, and personalities treatment could be given with more success and less morbidity.

When the surgery is not indicated concurrent chemoradiotherapy must be used.



Figure 1: Fig. 1 :

- [Khan et al.] , M J Khan , Castle , A T Lorincz , S Wacholder , M Sherman , D R Scott , B B Rush , A G Glass .
- [Gynecol Oncol ()] , doi: 10.1016/j.ygyno.2017. 01.011. *Gynecol Oncol* 2017 Apr. 2017 Jan 13. 145. 145 (1) p. .
- [NCCN Clinical Practice Guidelines in Oncology Cervical Cancer Version ()] , *NCCN Clinical Practice Guidelines in Oncology Cervical Cancer Version* 2018-October 25, 2017. 1.
- [Chiappa et al. (2015)] ‘Agreement of two-dimensional and three-dimensional transvaginal ultrasound with magnetic resonance imaging in assessment of parametrial infiltration in cervical cancer’. V Chiappa , Di Legge , A Valentini , AL , Gui B Miccò , M Ludovisi , M Giansiracusa , C Testa , A C Valentin , L . doi: 10.1002/uog.14637. *Ultrasound Obstet Gynecol* 2015 Apr. 2015 Mar 1. 45 (4) p. .
- [Schiffman et al. ()] ‘Atypical Squamous Cells of Undetermined Significance/Low-Grade Squamous Intraepithelial Lesion Triage Study Group. Human papillomavirus DNA remains detectable longer than related cervical cytologic abnormalities’. M Schiffman , Wheeler , P E Castle . *J Infect Dis* 2002. 186 p. .
- [Lee et al. ()] ‘Bilateral radical radiotherapy in a patient with uterus didelphys’. C D Lee , M Churn , N Haddad , J Davies-Humphries , R K Kingston , B Jones . *Br J Radiol* 2000. 73 p. .
- [Mayrand et al. ()] ‘Canadian Cervical Cancer Screening Trial Study Group. Human papillomavirus DNA versus Papanicolaou screening tests for cervical cancer’. M H Mayrand , E Duarte-Franco , I Rodrigues , S D Walter , J Hanley , A Ferenczy , S Ratnam , F Coutlée , E L Franco . *N Engl J Med* 2007. 357 p. .
- [Sugimori et al. ()] ‘Cervical cancers in uterus didelphys’. H Sugimori , T Hachisuga , S Nakamura , N Matsuo , G Nakamura . *Gynecol Oncol* 1990. 36 p. .
- [Rustagi et al. ()] ‘Cervical Screening and Cervical Cancer Death Among Older Women: A Population-Based, Case-Control Study’. A S Rustagi , A Kamineni , Sheila Weinmann , S Reed , S D Newcomb , P Weiss , SD . *Am J Epidemiol* 2014. 179 p. .
- [Querleu and Morrow ()] ‘Classification of radical hysterectomy. Classification of radical hysterectomy’. Denis Querleu , Paul Morrow . *Lancet Oncol* 2008. 9 p. .
- [Pk and Heinonen ()] ‘Clinical implications of the didelphic uterus: long-term follow-up of 49 cases’. Pk , Heinonen . *Eur J of Obstetrics Gynecol Reprod Biol* 2000. 2 p. .
- [Saint M Gildengorin and Sawaya ()] ‘Current cervical neoplasia screening practices of obstetrician/ gynecologists in the US’. Saint M Gildengorin , G F Sawaya . *Am J Obstet Gynecol* 2005. 192 p. .
- [Rezai et al. ()] *Didelphys Uterus: A Case Report and Review of the Literature. Case Reports in Obstetrics and Gynecology*, S Rezai , P Bisram , Lora Alcantara , I Upadhyay , R Lara , C Elmadjian , M Case Report . 10.1155/2015/865821. ID 865821. <http://dx.doi.org/10.1155/2015/865821> 2015. 2015.
- [Wu et al. ()] ‘Herlyn-Werner-Wunderlich syndrome consisting of uterine didelphys, obstructed hemivagina and ipsilateral renal agenesis in a newborn’. H Wu , T Wu , T Ng , S Ng , P Su , J Chen , S Chen . *Pediatr Neonatol* 2012. 1 p. .
- [Rastogi et al. ()] ‘Müllerian duct anomalies and ther effect on the radiotherapeutic managment of cervical cancer’. M Rastogi , S Revannasiddaiah , P Thakur , P Thakur , M Gupta , M Gupta , R K Seam . *Chin J Cancer* 2013. 32 p. .
- [Ronco et al. ()] ‘New Technologies for Cervical Cancer screening (NTCC) Working Group Efficacy of human papillomavirus testing for the detection of invasive cervical cancers and cervical intraepithelial neoplasia: a randomised controlled trial’. G Ronco , P Giorgi-Rossi , F Carozzi , Dalla Confortini , P Palma , Del Mistro , A Ghiringhello , B Girlando , S Gillio-Tos , A , De Marco , L Naldoni , C Pierotti , P Rizzolo , R Schincaglia , P Zorzi , M Zappa , M Segnan , N Cuzick , J . *Lancet Oncol* 2010. 11 p. .
- [Moyer ()] ‘Screening for cervical cancer: U.S. Preventive Services Task Force recommendation statement’. V A Moyer . *Ann Intern Med* 2012. 156 p. .
- [Schiffman ()] ‘The elevated 10-year risk of cervical precancer and cancer in women with human papillomavirus (HPV) type 16 or 18 and the possible utility of type-specific HPV testing in clinical practice’. M Schiffman . *J Natl Cancer Inst* 2005. 97 p. .
- [Grimbizis et al. (2013)] *The ESHRE/ ESGE consensus on the classification of female genital tract congenital anomalies. Hum Reprod*, G F Grimbizis , S Gordts , Di Spiezio Sardo , A Brucker , S , De Angelis , C Gergolet , M Li , T C Tanos , V Brölmann , H Gianaroli , L Campo , R . doi: 10.1093/humrep/det098. 2013 Aug. 2013 Jun 14. 28 p. .
- [Oppelt et al. ()] ‘The VCUAM (Vagina Cervix Uterus Adnex Associated Malformation) Clas-sification: a new classification for genital malformations’. P Oppelt , S P Renner , S Brucker , P L Strissel . *Fertil Steril* 2005. 84 p. .
- [Swales et al.] *The Wertheim hysterectomy: Development, modifications, and impact in the present day*, A Swales , A Gockley , R Phaëton , J P Kesterson .

4 IV. CONCLUSION

- 205 [Iavazzo et al. ()] *Uterine carcinosarcoma in a patient with didelphys uterus. Case Rep Obstet Gynecol*, C Iavazzo
206 , F Kokka , A Sahdev , N Singh , K Reynolds . 10.1155/2013/401962. 2013. 2013. p. 401962.
- 207 [Martínez et al. ()] ‘Uterus didelphys with septate cervix and unilateral endometrial carcinoma: a case report’.
208 M Martínez , J Giménez , P Acién . *Journal Genital Syst Disor* 2012. Issue1. 1.
- 209 [Pk Heinonen ()] ‘Uterus didelphys: a report of 26 cases’. Pk Heinonen . *Eur Journal of Obstet Gynecol and*
210 *Reprod Biol* 1984. 5 p. .