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Research Article

Verneuil's Disease or Hidradenitis Suppurativa Complicated by Verrucous Carcinoma: Description of a Case with Extremely Extended Pathology in Pregnancy

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ARTICLE INFO

Article history:

Received: 2 November, 2020

Accepted: 18 November, 2020

Published: 24 November, 2020

Keywords:

Verneuil's disease

hidradenitis suppurativa

verrucous carcinoma

infliximab therapy

RB turn off expression by HPV

ABSTRACT

Background: Verneuil's disease is a rare suppurative pathology of apocrine glands and pilosebaceous system of the inguinal-pelvis and perianal region with a multifactorial aetiology. The disease produces skin fistulas, draining a serum-purulent material on the skin, and finally with spontaneous healing. Verneuil's disease can be complicated by recurrence, and unfortunately, by squamous carcinoma. We have found in literature, just 2 cases of Hidradenitis Suppurativa, complicated by Verrucous Carcinoma.

Main Observation: we describe a patient F, 34-year-old, comes to our observation at 4th gestational month, with suppurative sinus pilonidal diagnosis and with the absolute indication of the gynaecologist to the abortion. We decided to continue the pregnancy. The patient gives birth with C. to 9 months; the new-born baby was perfectly healthy and after the maternal milk analysis, the baby also nursed for about 2 months. Finally, we decided to perform a wide excision biopsy and after a controversial histology (first Warty Carcinoma, then after a "second view" Suppurative Hidradenitis), we started a series of medical treatments, including biological therapy with Infliximab, but unfortunately with fleeting results. The patient did not answer substantially. Therefore, the last chance appears a combined demolitive and plastic surgical treatment. The histology described a Verrucous squamous cell carcinoma maximum diameter of 38 x 26 cm, with lateral deep margins free of neoplastic involvement.

Conclusion: This very advanced clinical behaviour of the Hidradenitis Suppurativa can present in pregnancy, therefore, motivating the interruption of the same one. Our experience contributes to underlining the possibility to bring to term the pregnancy. This case report shows how the hormonal activity can anyway influence the development of the hidradenitis and an interesting hypothesis could consider the hormonal activity of the pregnancy and the HPV promoting gene regulation (RB) from inflammatory pathology to neoplastic pathology in VC like a model to study.

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Background

The Verneuil's disease is a rare suppurative pathology of the apocrine glands and pilosebaceous system of inguinal-pelvis and perianal region

with a multifactorial aetiology. First time described in 1839 by Velpeau, but only in 1854 Aristide Stanislas Auguste Verneuil discovered the cause of the disease: it affects the apocrine glands due to the occlusion of a specific portion of the hair follicle (infundibulum); that is why it is

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<http://dx.doi.org/10.31487/j.JSO.2020.06.01>

also known as inverse acne or hidradenitis suppurativa [1]. It is still unclear what exactly triggers the problem. Sometimes it can be hereditary and affects about 4% of the women, usually with a longer menstrual phase duration and a shorter monthly cycle; evolution looks to be influenced by unknown hormonal activities like suggested by the behaviour during pregnancy and childbirth [2, 3]. Typical incidence is after puberty, beginning with an infundibulum occlusion, resulting in non-drainage of secretions on the skin that accumulates in the gland that swells and becomes inflamed, forming painful nodules [4]. The disease produces skin fistulas, draining a serum-purulent material on the skin, and finally to heal. Verneuil's disease can be complicated by recurrence, and unfortunately, by squamous carcinoma [5]. We have found in literature, just 2 cases of Hidradenitis Suppurativa, complicated by Verrucous Carcinoma, a specific disease entity independent of the other epidermoid carcinomas for morphological characteristics, clinical behaviour and favorable prognosis [6]. The Verrucous Carcinoma (VC), a variant of well-differentiated squamous cell carcinoma of low grade, was described for the first time by Lauren Ackerman in 1948 [7]. The incidence is unknown and infection with human papillomavirus (HPV) and the chemical carcinogenesis appear to be involved in the pathogenic mechanism. Few descriptions of skin lesion usually involve the plantar surface of feet [8].

Main Observation

The patient F, 34-year-old, comes to our observation at 4th gestational month, with suppurative sinus pilonidalis diagnosis and with the absolute indication of the gynaecologist to the abortion. It was immediately clear of an unusual case of a wide extension of Hidradenitis Suppurativa, Hurley III stage with highest points in the Sartorius Stage : wide anatomic regions involved, many types of lesions involved (abscesses, nodules, fistulas, scars, points for lesions of all regions involved), no distance between lesions, and no or few normal skins in between lesions (Figure 1) [6, 7].



Figure 1: Pregnant Patient at an early stage of pathology.

We decided to continue the pregnancy, considering the good clinical conditions of fetus and mothers. The patient gave birth with C. to 9 months; the new-born baby was perfectly healthy and after the maternal milk analysis, the baby was also nursed for about 2 months. After the pregnancy, we observed a significant improvement of the clinical condition (extension and suppurative aggressiveness). This behaviour was confirming a correlation with Prolactin levels, but unfortunately, after few months, the pathology was becoming more aggressive and finally we decided to perform a wide excision biopsy instead of needle

biopsy to avoid seeding complication, treating the excision wound bed, strongly bleeding with a gelatin-thrombin matrix topical hemostatic agent (FloSeal) to avoid bleeding and serum flow after a controversial histology (first Warty Carcinoma, then after a "second view" by the Morphopathology of London, hidradenitis suppurativa) [9-11]. The patient, after 6 months of biological medical treatment, including biological therapy with Infliximab, advanced medication, ozone therapy did not answer substantially, getting progressively worse (Figure 2) [12].



Figure 2: Patient 6 months after pregnancy in an advanced stage of pathology.

Therefore, the last chance appears a combined demolitive and plastic surgical treatment. CT with contrast described, in the median and paramedian huge expansive neoformation vegetating broad-based plant, infiltrating the subcutaneous plane to the muscle gluteus maximus. The extension described was from right medium axillary lines to the left one and from lumbar spondyl to the coccyx. The patient was then subjected to a partial excision and partial reconstruction of the defect, and like literature suggests, we started a series of medical treatments. The patient did not answer substantially. Therefore, the last chance appears a combined demolitive and plastic surgical treatment [13]. The advanced clinical conditions of the patient increasing the risk of surgical and anaesthesiological complications, have motivated the patient to refuse the treatment of excision of the lesion and reconstruction of the large defect with free grafts [14]. Macroscopic description: maximum diameter of 4,5x3,2 cm, deep from 2 to 5 cm, with lateral deep margins involved. The histology described a verrucous squamous cell carcinoma, a papillary tumor composed of a well-differentiated squamous epithelium with minimal cytological atypia, covered by a thick keratin layer. The main parameter that then diversifies the cancers from other types of epidermoid carcinoma is the good cytological differentiation through the entire thickness of the neoplasia [15]. We did not approach it before and after surgery with radiotherapy because it could alter the nature of the tumor, determining transformation into anaplastic sense, with higher metastatic potential [16]. Unfortunately, the patient died after 1 year for oncological cachexia.

Conclusion

This very advanced clinical behaviour of the Hidradenitis Suppurativa can present in pregnancy (Prolactin effect), therefore, motivating the interruption of the same one. This case report shows how hormonal activity can anyway influence the development of hidradenitis (more aggressive during pregnancy and less aggressive after). Although the pathogenesis of VC is unknown, it seems that infection with human papillomavirus (HPV) can be involved [17, 18]. In particular, the infection HPV may turn off the expression of some anti-tumor genes such as RB [19, 20]. Inflammatory diseases chronic long duration, such as hypertrophic lichen planus or hidradenitis suppurativa, seem to predispose onset of VC [21, 22]. Rarely, the lesion may arise after many years, when traumatic skin ulcers, varicose, burns or radiodermatitis [23, 24]. An interesting hypothesis could consider the hormonal activity of the pregnancy and the HPV promoting gene regulation (RB) from inflammatory pathology to neoplastic pathology in VC like a model to study.

Ethics Approval and Consent to Participate

Not Applicable.

Consent for Publication

We have had authorization for publication like our Hospital Privacy Policy already require.

Availability of Data and Materials

The datasets used and/or analysed during the current study are available from the corresponding author on reasonable request.

Competing Interests

None.

Funding

None.

Author Contributions

All authors contributed significantly to the present research and reviewed the entire manuscript. DP: Participated substantially in conception, design and execution of the study and in the analysis and interpretation of the data; also participated substantially in the drafting and editing of the manuscript. RR: Participated substantially in conception, design and execution of the study and in the analysis and interpretation of the data; also participated substantially in the drafting and editing of the manuscript. GC: Participated substantially in conception, design and execution of the study and in the analysis and interpretation of the data. FF: Participated substantially in conception, design and execution of the study and in the analysis and interpretation of the data. DB: Participated substantially in conception, design and execution of the study and in the analysis and interpretation of the data. MLV: Participated substantially in conception, design and execution of

the study and in the analysis and interpretation of the data. BC: Participated substantially in conception, design and execution of the study and in the analysis and interpretation of the data. FF: Participated substantially in conception, design and execution of the study and in the analysis and interpretation of the data. GG: Participated substantially in conception, design and execution of the study and in the analysis and interpretation of the data. LD: Participated substantially in conception, design and execution of the study and in the analysis and interpretation of the data. All Authors have read and approve the final manuscript.

Acknowledgements

Not Applicable.

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