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Case Report

Pathology Section

Melanosis of the Uterine Cervix: A Rare Case Report

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ABSTRACT

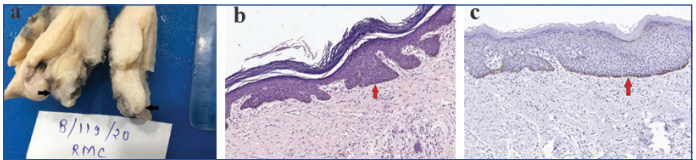
Cervical melanosis is a rare entity in the spectrum of melanocytic lesions of uterine cervix. Melanosis is defined as presence of melanocytes in the basal layer of squamous epithelium causing hyperpigmentation. Authors here by report a case of 57-year-old female who underwent vaginal hysterectomy for third degree utero-vaginal prolapse, showed an incidental gross pathological finding of brownish macular lesion. Histopathological examination showed hyperpigmentation of basal layer without increase in melanocytes. On immunohistochemical examination, basal melanocytes were highlighted by S-100 and HMB 45 immunostains. Thus final diagnosis of cervical melanosis was made. Clinical differentials of cervical pigmented melanocytic lesions include cervical melanomas, blue nevi, congenital or traumatic lesions and melanosis, hence vigilant clinical, gross pathological examination and biopsy is warranted.

Keywords: Biopsy, Hyperpigmentation, Melanocytes, Melanoma

CASE REPORT

A 57-year-old postmenopausal female presented to Department of Gynaecology with complaints of pelvic heaviness and bulging mass per vagina since 2-3 years along with urinary discomfort and constipation. There was no associated co-morbidities, visible hyperpigmented skin lesions or any significant past history. Ultrasonography showed 47×37 mm sized heterogeneously hypoechoic solid anterior wall fibroid of uterus. Per vaginal examination showed third degree utero-vaginal prolapse with grade 1 rectocoele. Cervico-vaginal Pap smear examination was negative for intraepithelial lesion or malignancy. Based on clinical findings and investigations, provisional diagnosis of third degree uterovaginal prolapse with grade 1 rectocoele and uterine anterior wall fibroid was made. Vaginal hysterectomy was performed and sent for histopathological examination. On gross examination ectocervix showed macular light brown coloured pigmented area measuring 2.1×1.8 cm noted over posterior lip of cervix [Table/Fig-1a].

Histopathological examination of macular lesion of cervix revealed keratinised stratified squamous epithelium with mild hyperkeratosis and hypergranulosis. Basal layer showed hyperpigmentation showing deposition of melanin pigment without significant increase in melanocytes. Subepithelial stroma was unremarkable. No dysplasia/malignancy noted. Differential diagnoses of blue nevus and traumatic lesion were ruled out due to absence of stromal melanocytes and haemorrhage/haemosiderin laden macrophages. Melanin bleach successfully removed basal pigmentation confirming it to be melanin. Immunohistochemistry for S100 and HMB-45 immunostains highlighted the basal melanocytes. With these findings diagnosis of cervical melanosis was rendered with changes consistent with prolapse [Table/Fig 1-b,c]. Patient's current status is not available as she was lost to follow-up.



[Table/Fig-1]: (a) Gross photograph of external surface of ectocervix showing light brown coloured pigmented area; (b) Histopathology showing hyperpigmentation of the basal layer of the squamous mucosa (H&E stain, 100x); (c) HMB-45 immunostain highlighting basal melanocytes (IHC, 100x).

DISCUSSION

Melanocytic lesions form important category of visible pigmented lesions of the uterine cervix. These are rare and include the spectrum of benign melanosis, blue nevi (commonest) and malignant melanoma. Other non melanocytic pigmented lesions include endometriosis, haemangioma, haemorrhage in nabothian cysts, granulomatous vasculitis, collection of haemosiderin laden macrophages and multinucleate giant cells surrounding carbon like pigment [1]. Careful microscopic examination will help to differentiate between various pigmented lesions ranging from non melanocytic lesions to harmless benign melanosis to grave melanomas [1].

Melanosis of uterine cervix is benign and rare [1]. It is also being described as synonyms like lentigo, pigmentation, ephelis or melanotic macule of the cervix. Tran TA et al., reported 26 melanocytic lesions among 2118 hysterectomy specimens, in a large prospective study [1]. The study showed blue nevi as commonest while melanosis as the least common entities with an incidence of 1.2% and 0.05%, respectively. Concurrent melanosis of the vagina and cervix is also described in the literature [2]. To the best of the authors knowledge with a thorough review of english literature only 23 other cases of cervical melanosis are reported previously [1,3,4]. Such lesions are commonly missed or misinterpreted in clinical and pathological examination as evidence of previous biopsy [3]. Tran TA et al., noted a learning curve effect in the identification of these pigmented lesions and attributed causes for lower detection rate in the form of, more focus on other significant pathology by residents or pathology assistants [1]. Mudge TJ et al., documented a case of malignant melanoma of the cervix associated with pre-existing area of melanosis [5]. Thus, in uterine cervix whether melanosis is transformed into malignant melanoma like other skin counterparts needs to be studied further. Hence, it emphasises importance of careful meticulous clinical and histopathological examination as an integral part in the management of these lesions.

Melanosis appears to be common in 41-50 years age group with youngest case described at 24 years by Hytiroglou P et al., and oldest being 61 years by Schiller W et al., [1,6,7]. In present case, melanosis was incidentally detected in hysterectomy done for third degree utero-vaginal prolapse. This presentation is similar to that of most of cases reported previously. In remaining cases hysterectomies were performed for complains of menorrhagia, metromenorrhagia, vaginal discharge, incidental finding in screening, pelvic mass, dysmenorrhea, investigation of infertility etc., [1]. A single case of

melanosis developed after cryotherapy for epithelial dysplasia is also described [6]. All these findings favour chronic trauma and irritation as a likely causative factor as suggested by Yilmaz AG et al., [8]. In most of cases, size of the lesion ranged from small macule to large pigmented areas measuring upto 2.5 cm [1,3].

Histopathologically melanosis is represented by basal layer hyperpigmentation of the squamous mucosa with or without increase in number of melanocytes. Some authors refer term letingenes, if number of melanocytes is increased. Almost all of the melanosis cases have shown involvement of ectocervix only with an exception showing changes in squamous metaplastic endocervical gland. Blue nevi are defined by presence of melanocytes in endocervical stroma. Three types of blue nevi are described which include stromal melanocyte focus, mixed type and nevoid type based on presence of slender spindled melanocytes, plump spindled melanocytes and nevoid epithelioid stromal melanocytes respectively [1]. Both melanosis and blue nevi show positivity for S100 and melancytic markers HMB-45 and Melan A. Primary malignant melanomas are distinctive and usually show junctional changes in squamous epithelium with nesting pattern or poorly differentiated lesions with features of anaplasia, high mitotic activity, pigmentation and diffuse strong immunopositivity for S100 and melanocytic markers. To label it as primary, absence of similar lesions elsewhere in body is demonstrated [1,4,9].

Vezzani M and Sola P proposed three possible theories for the origin of melanocytes which include: 1) erratic migration of neural crest elements; 2) melanocytes as a components of malformations i.e., choristoma; and 3) migration of melanocytes from adjacent mucocutaneous areas representing transformation of squamous epithelium to resemble as 'epidermis' as part of local pathological processes like trauma, irritation, prolapse or procedures [10]. As in most of described cases, there was an association with prolapse or local trauma; hence third theory can be aptly applied for melanocyte origin [4].

Chang D et al., suggested possible syndromic associations with the genital melanosis in the form of Laugier-Hunziker syndrome (idiopathic mucocutaneous lenticular pigmentation) associated with oral melanocytic lesions and carney complex associated with atrial myxoma and multiple endocrine neoplasias [4]. But their direct actual association with melanosis of the cervix needs to be studied further.

CONCLUSION(S)

Pigmented melanocytic lesions of uterine cervix are rare and need vigilant clinical and pathological examination. Melanosis of the uterine cervix is extremely rare and must be in the differentials of visible pigmented lesions. Biopsy is of utmost importance to differentiate between benign cervical melanosis and other lesions including melanoma.

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