



## Benign Peritoneal Melanosis associated with atypical leiomyoma

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### ABSTRACT

Benign Peritoneal Melanosis is an extremely rare condition with only a handful cases in the literature. It is characterized by melanin pigment deposition in the peritoneum. The pathogenesis of Peritoneal Melanosis in particular, the origin of the pigment producing cells, is unclear. We describe a case of Benign Peritoneal Melanosis associated with atypical leiomyoma of the uterus in a 40 year old woman. She presented with increased blood loss during menses and recurrent pain in abdomen since last 3 years. On USG Pelvis, she was diagnosed as a case of fibroid uterus and was posted for hysterectomy. During surgical procedure, India ink colored (Black) pigmentation was seen in the peritoneum and the complete lining peritoneum of the pelvis was seen black. Only the body of the uterus, the fallopian tubes and ovaries were spared. Biopsies of the peritoneum showed pigment in the stroma and pigment laden histiocytic aggregation and ultra structural study found melanosomes in the cytoplasm of histiocytes.

**Keywords:** Benign Peritoneal Melanosis, Hysterectomy, Melanosomes

### INTRODUCTION

Benign Peritoneal Melanosis, a diffuse black pigmentation of peritoneum, is a condition characterized by melanin pigment deposition in the peritoneum, mesentery, appendix surface, pelvic peritoneum and surface of ovary [Kim et al 2002, Jaworski 2003]. It is of unknown origin and it is an extremely rare condition with only handful of cases in the literature.

### SOMMARIO

La Melanosi Peritoneale Benigna è una condizione estremamente rara con pochi casi riportati in letteratura. È caratterizzata da deposizione di melanina nel peritoneo. La patogenesi della melanosi peritoneale ed in particolare, l'origine della produzione delle cellule pigmentate, è non-chiara. In questo articolo descriviamo un caso di melanosi peritoneale benigna associata a leiomyoma uterino atipico in una donna di 40 anni. La paziente ha presentato un aumento della perdita di sangue durante le mestruazioni e dolore ricorrente addominale negli ultimi 3 anni. Con USG della Pelvi, è stata diagnosticata come un caso di fibroma dell'utero ed è stata eseguita l'isterectomia. Durante la procedura chirurgica, con la colorazione di inchiostro di china (nero) il peritoneo e il rivestimento del bacino è diventato nero. Solo il corpo dell'utero, le tube di Falloppio e le ovaie sono stati risparmiati. Le biopsie del peritoneo hanno mostrato pigmentazione dello stroma e aggregazione istiocitaria pigmentata e lo studio ultra-strutturale ha trovato melanosomi nel citoplasma degli istiociti.

### CASE REPORT

A 40 yr old female para 4 living 4, all full term normal vaginal deliveries came to the OPD with chief complaints of increased blood loss during menses since last 1 year and pain in lower abdomen and backache since 1 year. Her LMP was 20 days back and during her present cycles she had a heavy flow of menstrual blood for 6 to 7 days with an interval of every 30 days. This was since last one year. Previous cycles were regular with average blood loss. She was married for 19 years and was using barrier method of contraception. Her last child birth was 15 years. There was no history of any surgery in the past and no relevant medical history. On general examination, all parameters were within normal limits. Per abdomen nothing abnormal was found. Cervix and vagina appeared

healthy; Pap smear was taken which was reported as inflammatory. On p/v examination cervix was downwards backwards, uterus anteverted, 6-8 weeks in size and firm in consistency. Both fornices were free. USG pelvis showed a submucosal to intramural fibroid in anterior wall of the body of uterus measuring 5.2 X 6.0 cms. Pap smear was inflammatory. OT profile was done and decision of hysterectomy was taken in view of symptomatic fibroid uterus.

On opening the abdomen, when we reached the peritoneum, we could see dark picture through the peritoneum as if there was collection of clotted blood. That gave us the suspicion of ruptured chronic ectopic pregnancy or ruptured hemorrhagic ovarian cyst that we might have missed. After opening the parietal peritoneum we could see complete dark black peritoneum covering the inner lining of pelvis. Body of the uterus, ovaries, fallopian tubes were spared upto uterovesical fold anteriorly and posteriorly till rectovaginal fold of peritoneum. Biopsy was taken from peritoneum and subtotal hysterectomy with bilateral salpingo oophrectomy was done. Decision of subtotal hysterectomy was taken as there was dark pigmentation below the utero vesical fold of peritoneum (shown in **Figure 1**). Bilateral salpingo oophrectomy was also considered keeping in view of again any pathology developing in ovaries in future for which laparotomy may be required, as this operative finding was a very rare and unknown entity. Laparotomy was performed over laparoscopy because patient was not affording for laparoscopy and she wanted abdominal hysterectomy. A sample of peritoneum was taken and the specimen was sent for HPR. Patient stood the procedure well and the post op was uneventful.

## MACROSCOPIC FINDINGS OF THE SPECIMEN

There was a flap like structure black in color measuring 2.2X1.3X0.8 cms. Another specimen was of uterus with both adnexa. H & E pictures of peritoneum and underlying connective tissue showed deposition of fine granular black material in the submesothelial fibrous tissue (**Figure 1C & D**). This material stains black with Masson Fontana stain (A Stain for Melanin). On the basis of surgical and histopathological findings, diagnosis of Benign Peritoneal Melanosis was made. Histopathological findings of uterus showed as Atypical Leiomyoma of uterus.



**Figure 1.** Surgical Picture of abdomen showing black colored peritoneum(A&B), Hematoxylin and Eosins(10X) showing fibro adipose tissue with brown black colored melanin pigment(C) Hematoxylin and Eosins(40X) stained section of peritoneal biopsy revealed fibrous connective tissue with deposition of brown black colored pigment in the peritoneum. The pigment stained positive with Masson Fontana stain and negative with Perls stain proving the pigment to be Melanin (D).

## DISCUSSION

Peritoneal melanosis, a diffuse black pigmentation of peritoneum is a very rare condition characterized by melanin pigment deposition in the peritoneum, mesentery, appendix surface, pelvic peritoneum and surface of ovary. It is an extremely rare condition with only handful of cases in the literature. Benign peritoneal melanosis is of unknown origin. Confirmation of the condition is done with peritoneal biopsies.

There are handful case reports showing presentation of Peritoneal Melanosis. Angelopoulos et al in 2013 reported a case of Benign Peritoneal Melanosis in 35 year old woman with symptoms of abdominal and pelvic pain. Diagnosis was done by laparoscopy and confirmed by extensive peritoneal biopsies. Kim et al in 2010 reported a case of peritoneal melanosis associated with mucinous cystadenoma of ovary and adenocarcinoma of colon. In that patient India-ink-colored pigmentation was seen in the peritoneum, in the omentum, and on the surface of the ovary during the surgery. Pigment in the stroma and pigment-laden histiocytic aggregation were seen in biopsies of omentum and peritonium. Likewise, another case of peritoneal melanosis combined with serous cystadenoma of ovary was reported by Kim et al 2002. Follow up of these cases were not reported.

It has been described along with cystic abnormalities of the ovary (serous, mucinous), cystic teratomas of the ovary (Dermoid cysts), colonic tumors, malignant melanomas and rarely with genetic disorders (eg enteric duplication, gastric triplication) [De la Torre 1997, Nada et al 2000, Kim et al 2002, Hefaiiedh et al 2009]. Our patient had no GIT Symptoms. She did not report any ovarian cyst or cyst "accident".

There is no protocol yet mentioned for the follow up of this clinical entity so we decided to give the patient first follow up in 1 month, 2nd follow up in 3 months and then every 6 months. Prognosis of this condition is quite uncertain. History and pattern of disease is unclear due to scarcity of cases. Given associations with ovarian pathology and gastrointestinal malignancies we suggest confirmation of the condition with

peritoneal biopsies and further investigation to exclude sinister pathology. This case was chosen for publication because of its rarity, scary presentation and it could be of research interest

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None

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