



Case Report

Juvenile cystic adenomyoma - A rare cause of refractory dysmenorrhoea



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ABSTRACT

Introduction: This is a case report of Juvenile Cystic Adenomyoma (JCA) which is very rare disease. Here, our purpose is to describe this rare cause of refractory secondary dysmenorrhoea along with its whole diagnostic work up & to evaluate laparoscopic management for treatment of JCA in terms of symptomatic relief & recurrence.

Materials and Methods: Laparoscopic excision of cystic lesion was performed & the mass was sent for histopathological examination(HPE). Diagnosis of JCA was confirmed by HPE. Patient has been followed up for four times to assess for relief of dysmenorrhoea & recurrence of adenomyosis.

Results: In all four follow up visits, the patient had not complained of dysmenorrhoea & there was no evidence of adenomyosis.

Conclusion: In Conclusion, JCA is a rare cause of severe secondary dysmenorrhoea & therefore, it should be kept in mind when an adolescent girl presents with severe dysmenorrhoea refractory to medication. Laparoscopic removal is the best in terms of feasibility & preservation of fertility with resolution of symptoms.

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1. Introduction

Adenomyosis is a condition where there is presence of heterotopic endometrial glands and stroma in myometrium with adjacent smooth muscle hyperplasia. Adenomyoma is term used when diameter of the mass is >5mm. Juvenile cystic adenomyoma (JCA) is cystic lesion & it is of earlier onset than usual time of onset of adenomyosis in women. Here in, we report a rare case of cystic adenomyoma which was treated in a 20 year old unmarried female patient with complaint severe dysmenorrhoea since menarche that was refractory to analgesics & conservative management. The whole diagnostic work up of the patient & laparoscopic removal of the lesion have been mentioned.

2. Case Report

A 20 year old unmarried female patient came to gynaecology OPD with complaint of severe dysmenorrhoea since past 7 years. The patient gave history of onset of menarche at 13 years of age with onset dysmenorrhoea from the beginning of the menarche. She had menstrual bleeding at regular intervals & moderate in quantity with severe lower abdominal, pelvic & backpain. Patient was nulligravida with no history of sexual exposure. Patient had history of medical treatment with NSAIDs, Sevista & Dinogest with no relief of symptoms.

On general examination, patient had no pallor/edema/icterus/clubbing/cyanosis/lymphadenopathy. Vitals were within normal limits. There was not any abnormality detected on respiratory, cardiovascular & neurological system examination.

On per abdominal inspection, there was no positive finding. On palpation, a soft, tender mass of approximately

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3× 4 cm in size was felt at right iliac fossa with no guarding and/or rigidity. There was no change in size of mass with respiration or on cough stimulus. On auscultation, there was not any gurgling sound or bruit detected. A detailed gynaecological examination could not be performed because the patient was virgin.



Fig. 1:



Fig. 2:

2.1. Investigations

The computed Tomography scan demonstrated a 19× 22 mm size cystic lesion in uterine myometrium at right side of body & in fundal region. A 3D-Pelvic Ultrasonography detected 25×22× 15 mm size thick walled cystic lesion in right lateral wall of the body of the uterus. Based

on clinical examination & imaging finding, differential diagnosis which we thought were unicornuate uterus with hematometra in a non communicating rudimentary horn, cystic degeneration of fibroid, congenital uterine cyst & intramyometrial hydrosalpinx.¹

A diagnostic & therapeutic laparoscopy for excision of the mass under general anaesthesia was planned after normal reports of all the necessary preoperative investigations with written, informed consent from the patient for the procedure. The laparoscopy was performed through an 11mm primary port & 3 accessory ports of 5mm in size. On laparoscopic examination, around 4× 4 cm size buldge was present on right lateral wall of uterus.

2.2. Laparoscopic procedure

An incision kept on buldging site at right lateral wall of uterus. Chocolate coloured fluid drained out from the cavity of the lesion. With the help of myometrial screw, endometrial cyst enucleated out from the myometrial wall. Myometrial wall sutured with vicryl 2-0. Hemostasis was achieved. The endometrial cyst (Figure 1) was removed from the left accessory port. All port sites closed with vicryl no.1 by vertical mattress.

2.3. Outcome & follow up

Histopathology confirmed the diagnosis of Juvenile Cystic Adenomyoma (Figure 2). Patient experienced complete relief of dysmenorrhoea during menses after laparoscopic surgery. There was no evidence of adenomyosis on follow up visits.

3. Discussion

Juvenile cystic adenomyoma is a very rare condition with around 43 cases reported in literature.² The most common presenting complaint is severe dysmenorrhoea.² These symptoms are thought to be due to intracyclic bleeding with progressive increase in cyst size. Juvenile cystic adenomyoma should be suspected when uterine cyst is associated with severe dysmenorrhoea in young girls.³ Juvenile cystic adenomyoma mimics uterine malformation that is uterine unicornis with hematometra in non communicating rudimentary horn.³ The best investigation to exclude other differential diagnosis in virgin patient is MRI.^{2,3}

In Conclusion, JCA is a rare cause of severe secondary dysmenorrhoea & therefore, it should be kept in mind when an adolescent girl presents with severe dysmenorrhoea refractory to medication. Laparoscopic removal is the best in terms of feasibility & preservation of fertility with resolution of symptoms.

3.1. Source of funding

None.

3.2. Conflict of interest

None.

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