

# Abdominal wall desmoid tumor during pregnancy: case report and literature review

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## Summary

Desmoid tumors are fibromatous lesions that are the result of abnormal proliferation of myofibroblasts. Despite its benign microscopic appearance and non-metastasizing behavior, tumor infiltrates surrounding tissues and has a high risk of recurrence. Pregnancy-associated desmoid tumors are very rare and optimal management of this tumor is not well established. The authors report a case of a 31-year-old pregnant woman with a large desmoid tumor, which increased rapidly in size and caused worsening symptom of dyspnea. The tumor was successfully removed during a caesarian section, which resulted in an anterior abdominal wall defect. Reconstruction of the abdominal wall defect was performed with a polypropylene mesh. The postoperative recovery of the patient was uneventful. After a follow-up of 44 months, the patient was found to be well and there was no evidence of local recurrence. The authors also reviewed the literature on the world's experience with this tumor and its management during pregnancy. Twelve desmoid tumors arising during pregnancy were reported in the existing literature; the managements were varied and has yet to be defined.

*Key words:* Desmoid tumor; Pregnancy; Abdominal wall.

## Introduction

Desmoid tumors, also known as aggressive fibromatosis or musculo-aponeurotic fibromatosis, are uncommon, benign, soft tissue neoplasms which comprise 0.1% of all the tumors and 3.5% of fibrous tissue neoplasms [1]. These rare tumors can develop in any musculo-aponeurotic structure and can be found in all regions of the human body. They may rise sporadically or in patients with familial adenomatous polyposis (FAP). The incidence of sporadic desmoid tumors has been estimated to be two to five per million people per year; however, it increases almost 1,000 times in patients with FAP [1, 2]. The precise etiology of desmoid tumor is undefined, however, trauma, surgical history, and estrogen hormone levels have been reported to play a crucial role in the pathogenesis, and genetic factors have also contributed [3]. Pregnancy-associated desmoid tumor is even rarer, and there is very limited published research available in this case. The optimal management of this tumor during pregnancy has yet to be defined.

The authors hereby report a large desmoid tumor that was diagnosed and resected successfully within the caesarian section. Previous reports have documented successful surgical management of these tumors during pregnancy with uneventful delivery, however, the size of the specimens they reported are much smaller than in the present case. A caesarian section followed by a successful complete resection

of the tumor, with uneventful postoperative recovery as well as longer-term follow-up. The authors also reviewed published literatures and management experience of this tumor during pregnancy.

## Case Report

A 31-year-old woman (gravida 2, para 1) at 34 weeks of estimated gestation was transferred to the present department because of difficult breathing. The symptom was mainly caused by an abdominal mass that had grown very rapidly. At 21 weeks gestation, in a primary hospital, it was misdiagnosed to be a uterine leiomyoma by ultrasound examination. The ultrasonography revealed a solid hypo-echoic mass measuring 12×15 cm in the anterior wall of the uterine. Seven weeks later, another ultrasonography was performed and it indicated that the mass had grown to 23.3×11.4×17.6 cm. The fetus was also evaluated and noted to be viable. The doctor advised her to receive a mass removal, however, the patient refused to undergo surgery. She began to experience symptoms of increased dyspnea. The symptoms were very severe at her 34 weeks of estimated gestation. Ultrasonic evaluation showed a large mass of 36×19.5×33 cm in size. From this result, there was a rapid growth of the mass, almost tripled in size compare to the prior result at 13 weeks gestation. This patient was transferred to the present department because of the limited medical resources at the primary hospital.

The patient was admitted, an ultrasonography confirmed a 35×35×14 cm mass located in the abdominal cavity anteriorly to the uterine. The myometrium of the anterior wall was measured to be six mm in thickness. The evaluation of the fetus showed that the baby was in a good condition. The authors suspected the mass to be a large leiomyoma or a sarcoma of the uterine. Given the

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Figure 1. — Intraoperative photograph: the uterine was first covered by the tumor, after successful child delivery, it was extracted from the pelvic cavity and sutured.



Figure 3. — Macroscopic appearance of the desmoid tumor.

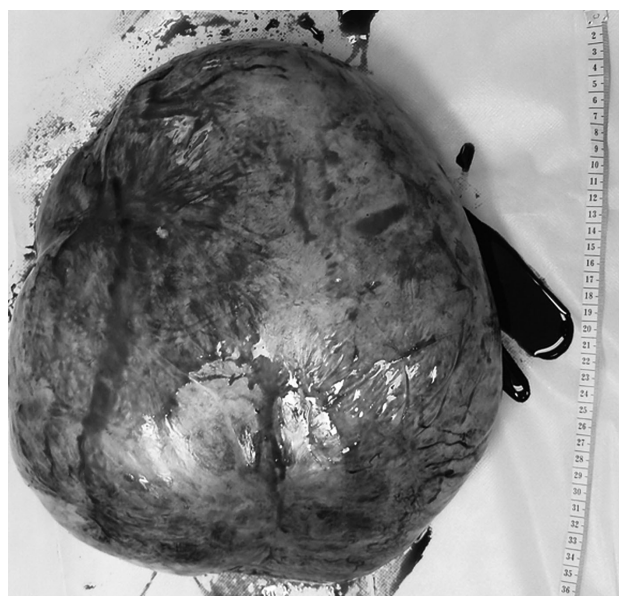


Figure 2. — Macroscopic appearance of the desmoid tumor.

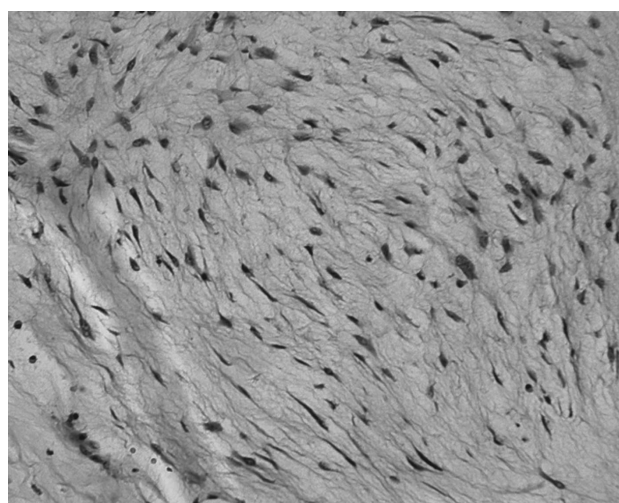


Figure 4. — Histological micrograph of the mass. The tumor consists of a proliferation of spindle cells without significant atypia or pleomorphism (H&E stain ×200).

rapid growth of this tumor and the worsened symptoms of dyspnea, a decision was taken to resect the tumor within the caesarian section and not to wait until the pregnancy came to term spontaneously. The surgery was performed at 35 weeks of estimated gestation.

A vertical incision of the lower abdominal was chosen. The authors found a large, hard, incompressible tumor in front of the peritoneum. The tumor seemed to originate from the anterior abdominal wall muscles and covered the uterine corps and the lower uterine segment. No adhesions were found between the tumor and the uterus. The tumor was so large that its superior border reached the xiphoid process, while its right side reached the right posterior axillary line and its left side reached the left anterior axillary line. The inferior border of the tumor reached the pubic symphysis.

Many vessels were found in the surface of the tumor. The authors used retractors to expose the lower uterine segment and then successfully preceded with caesarean section. A normal male infant with an Apgar score of 10 and a body weight of 2,300 g was extracted. After successful child delivery and closure of the uterine tissue, the authors extended the incision to the xiphoid process. However, due to the large tumor size, it was difficult to expose the entire tumor, so they made another transverse incision, from the upper side of the umbilical round to the left anterior axillary line. Then, a local complete resection with a macroscopically tumor-free margin was performed. The peritoneal and rectus abdominis involved were also dissected during the operation that resulted in an anterior abdominal wall defect measuring 10×5 cm in size. Reconstruction of the abdominal wall defect was performed

Table 1. — Summary of reports on desmoid tumor during pregnancy.

Study (year)	Patient (n)	Tumor location	Time of diagnosis	Tumor size	Intervention	Outcome	Ref.
De Cian <i>et al.</i> (1998)	1	Caesarean section scar	First trimester	8×5cm	Resection with mesh reconstruction during cesarean delivery	No recurrence	18
Kunieda <i>et al.</i> (1999)	1	Xiphoid and costal chondrium	Third trimester	10×11.5×11.5 cm (0.395 kg)	Conservative management	Wide excision postpartum; no recurrence	25
Gherman <i>et al.</i> (1999)	1	larynx	Second trimester	2.3×1.2×1.6 cm	Subtotal excisional biopsy at 21 weeks	Complete regression 9 weeks postpartum	28
Firoozmand <i>et al.</i> (2001)	1	Mesentery	Second trimester	17×14×10 cm (1.12kg)	Second resection with ileostomy	Preterm delivery	43
Molelekwa <i>et al.</i> (2004)	1	Abdominal wall	First trimester	3.2×6.5 cm	Conservative management	Caesarean section at 36 weeks with resection 1 month postpartum	19
Durkin <i>et al.</i> (2005)	1	Abdominal wall	First trimester	18.5×15×9 cm	Resection with mesh reconstruction at 20 weeks	Successful vaginal delivery at 39 weeks	9
Arshad <i>et al.</i> (2008)	3	Abdominal wall	Case 1: third trimester Case 2: not mentioned Case 3: first trimester	17×15 cm 15×15 cm (1.2kg) 25×20 cm (4.3kg)	Conservative management	Resection with mesh reconstruction postpartum; no recurrence	20
Viriyaroj <i>et al.</i> (2009)	1	Abdominal wall	Second trimester	28×21×18 cm (4.9 kg)	Conservative management	Complete resection postpartum; no recurrence	26
Johner <i>et al.</i> (2009)	1	Abdominal wall	Third trimester	Not mentioned	Resection with primary reconstruction postpartum	Successful vaginal delivery at 39 weeks	3
Krentel <i>et al.</i> (2012)	1	Caesarean section scar	Third trimester	3-4 cm in diameter	Resection during cesarean delivery	No recurrence	27
Present case	1	Abdominal wall	Second trimester	35×30×14 cm (7.1 kg)	Resection with mesh reconstruction during cesarean delivery at 35 weeks	No recurrence	

with a polypropylene mesh, which was sutured to the excision edge of the anterior abdominal wall fascia and muscles. Intraoperative blood loss was approximately 2,500 ml, thus, the patient received blood transfusion (Figures 1-3).

The pathological report demonstrated that the specimen measuring 35×30×14 cm in size and 7.1 kg in weight. The tumor was encapsulated and margin-free. The histologic specimens consisted of interlacing bundles of fibrous tissues and benign fibroblasts with moderate cellularity. The appearance of which was consistent with desmoid tumor (Figure 4).

The postoperative recovery of the patient was uneventful. She was discharged ten days after the surgery. However, the infant was transferred to neonatal intensive care unit because of hyaline membrane disease caused by preterm labor. The patient was noted to be well and there was no evidence of local recurrence after 44 months of follow-up.

## Discussion

Desmoid tumor is a benign, locally aggressive neoplasm that arises from fascial or musculo-aponeurotic tissue. It is characterized by proliferation of fibroblasts but without the cytological feature of malignancy [4]. Although these tumors are benign, they can infiltrate the surrounding vital structures or organs, which may result in significant local morbidity and even death [5]. This tumor was first described

by MacFarlane in 1832, and the term of “desmoid” was applied by Muller in 1838 [6]. Most of them occur sporadically, while 5% arise in association with FAP [7]. The incidence in the general population is two to five cases per million people. Patients with FAP have a 1,000-fold increase risk for desmoid tumors as compared to the general population [1, 2]. Desmoid tumor may occur in the extremities (most commonly around limb girdles), the abdominal wall (most frequently diagnosed in women), and bowel mesentery (most commonly associated with FAP)[7].

The definitive etiology of desmoid tumors is currently unknown; however, endogenous or exogenous estrogen exposure has been shown to be a risk factor regarding the pathogenesis of this tumor. First, women are more frequently affected with an incidence ratio of female to male of 5:1 [2, 8]. Second, women during their reproductive age, pregnancy, and those taking contraceptive pills have a higher incidence of desmoid tumors [9]. Third, spontaneous regression of this tumor has been observed in women after menopause or oophorectomy [4, 10]. Finally, anti-estrogens agents such as tamoxifen have been reported to be effective in vitro [11, 12] and in vivo [13-15]. Risk factors like prior trauma or surgery and genetic pre-



disposition are also contributed to the pathogenesis of this tumor [3, 16].

Pregnancy-associated desmoid tumor is even less common. By now, the available literatures, reporting desmoid tumor during pregnancy, are sporadic case reports (Table 1). There is a lack of general recommendations for the treatment of pregnancy-associated desmoid tumor because of the small number of cases available in literature, as well as the lack of randomized and prospective studies concerning the direct comparisons of different treatment approached. Management of patients with desmoid tumor during pregnancy is complicated and some issues remain controversial. It is not only due to the tumor itself, but also because of confounding obstetrical considerations. Embryo safety has to be considered while deciding therapeutic approaches during pregnancy. Currently, the main controversies focus on the role and timing of surgery, and the safety and value of non-operative therapies.

A simple observation is a reasonable management option for selected patients. According to the literature, 10% of desmoid tumor resolved spontaneously, 30% underwent cycles of progression and resolution, 50% remained stable after diagnosed, and 10% progressed rapidly [6, 17]. Given its inherent morbidity, some authors suggested a wait-and-see policy for suitable patients. A number of reports regarding conservative management for desmoid tumor during pregnancy have been published. De Cian *et al.* documented a case where an abdominal wall desmoid tumor was diagnosed in a 42-year-old woman at 12 weeks of gestation. The tumor measured 8×5 cm in size. The patient was examined clinically every month, while the tumor was carefully measured every two months by ultrasonography and remained unchanged during the last six months of gestation. At last, the tumor was resected via a Pfannenstiel incision during concomitant caesarean section at 37 weeks gestation [18]. Molelekwa *et al.* reported a case in which a desmoid tumor was diagnosed in a 42-year-old woman during her first trimester. The tumor measured 3.2×6.5 cm in size. Regular fortnightly scans were performed and showed no significant growth of the tumor. The tumor was excised with clear margins one month after caesarean delivery [19]. Arshad *et al.* reported three patients who presented with a large anterior abdominal wall desmoid tumor diagnosed during their pregnancy. All three patients received a “wait-and-see” policy until the tumors were excised successfully postpartum [20]. Viriyaroj *et al.* reported a 17-year-old woman presenting with a desmoid tumor at her suprapubic region during the fifth month of gestation. She completed a full-term pregnancy and delivery by caesarean section. The tumor had grown very rapidly after delivery and was finally completely excised. In these cases, the patients were asymptomatic or with few or mild symptoms and the tumors were not so large. Most important, the conditions of the fetus were not influenced by the tumor. Therefore, pa-

tients with small desmoid tumor which is not encroaching on any nearby structures, especially not compressing the gravid uterus and endangering the fetus could be observed closely to complete a full-term pregnancy. These patients should be followed clinically and with modern imaging methods to assess the increase of size of these tumors. Ultrasonography and magnetic resonance imaging (MRI) are usually recommended. Different treatment approaches should be considered after successful delivery depending on the risk factors like age, tumor size, and location.

Surgery has a key role in the management of abdominal desmoid tumors. Wide radical local excision has been described as the optimal primary treatment. The role of surgical management for desmoid tumor has been questioned because of its inherent morbidity and its biological behavior. Desmoid tumor with a lack capsule or displays non-palpable spread along muscles and fascial planes makes it difficult for surgeons to estimate the tumor extent at operation. It can result in microscopic positive surgical margins and incomplete tumor removal, which possibly explains their high recurrence rate even after a presumably adequate resection [17, 21, 22]. The timing of the surgery is also controversial. Early surgical intervention entails risks to the fetus and potential obstetric problems related to resection and reconstruction of the abdominal defect. Severe pain had been reported during pregnancy due to shearing of an abdominal wall mesh graft [23]. Furthermore, a caesarean section would be technically troublesome in such a case, and vaginal delivery would be considered inadvisable within one or two years after the prosthetic implant [24]. To date, most of the literature has documented the diagnosis and resection of an abdominal desmoid tumor during caesarean section or postpartum [3, 18-20, 25-27]. However, only few reports regarding desmoid tumor resection during pregnancy have been published in the recent literatures. Gherman *et al.* reported a case of desmoid tumor of the larynx complicating pregnancy, which caused a symptom of progressively worsening vocal hoarseness and subtotal excision was performed at 21 weeks gestation. The residual tumor was monitored until the labor was induced at 36 weeks gestation because of oligohydramnios. Endoscopic evaluation and biopsy at nine weeks postpartum revealed regression of tumor [28]. Firoozmand *et al.* described an abdominal tumor in a pregnant woman, which caused worsened symptoms of difficult defecation and interference with fetal growth. The pelvic desmoid tumors measuring 17×14×10 cm was successfully excised at 23 weeks gestation, but the patient required a diverting ileostomy, and she subsequently entered preterm labor at 27 weeks [29]. Durkin *et al.* documented a case where a desmoid tumor was diagnosed at the first trimester. The tumor increased significantly in size and worsened the patient's symptoms of pain and abdominal fullness. The patient un-

derwent a successful en bloc resection of her desmoid tumor as well as abdominal wall reconstruction with polytetrafluoroethylene mesh during her 22<sup>nd</sup> week of gestation. Subsequent to her surgery, the patients completed a full-term pregnancy without complication and proceeded with an uneventful transvaginal delivery at 39 weeks [9]. These reports demonstrated that symptomatic desmoid tumors can be resected during pregnancy, even those requiring abdominal reconstruction. Surgical intervention has to be the first choice in desmoid tumors that cause significant symptoms, increased rapidly in size, compressing the uterus, and interfering with fetal growth. The decision to subject the mother and fetus to the potential morbidity of immediate surgery was weighed against the potential circumstance that further delay in treatment may have resulted in an inability to achieve tumor-free margin.

Radiation therapy had been used as an adjuvant treatment for patients with positive surgical margin as well as those who are poor candidates for surgery or with unresectable disease. Some investigators [30-32] reported better control of recurrent disease in patients with no residual tumor treated with adjuvant radiation therapy while some other groups [33, 34] failed to make similar observations. However, radiation therapy is contraindicated during pregnancy because it can lead to abortion, stillbirth and fetal malformation. The role of radiotherapy postpartum has yet to be defined.

A variety of systemic therapies have been investigated in non-pregnant patients. The aim is to induce remission, to prevent complications and disease recurrence, and to reduce morbidity [6]. Non-steroidal, anti-inflammatory drugs such as indomethacin, sulindac, and anti-estrogens such as tamoxifen have been considered as first line pharmacological therapies [26, 35, 36]. Cytotoxic drugs such as doxorubicin, dacarbazine, actinomycin-c, methotrexate, vinblastine, and vinorelbine have been found to have some activities in symptomatic patients, unresectable and clinically aggressive desmoid tumors which do not respond to conventional treatment [37-39]. Nevertheless, a medical intervention exposes the fetus to the potentially harmful effects of drugs, it is also contraindicated during pregnancy. To the knowledge of the present authors, there are no pregnant patients with desmoid tumor who have been treated with these drugs during pregnancy. The role of pharmacological therapy in postpartum period has yet to be established. Early delivery of the fetus may be necessary to facilitate tumor regression or to allow a treatment with radiotherapy or pharmacological therapies in selected patients.

Desmoid tumors are reported to have a high recurrence rate even after radical surgical excision due to the infiltrative growth pattern. The recurrence rate following resection of abdominal wall desmoid tumors are approximately 50% [40]. There is an increased risk of recurrence after a primary operation in patients with positive surgical mar-

gins of resection, whereas age, sex, site, size, or number of previous recurrences had no significant value on the likelihood of recurrence [37, 41, 42]. Unfortunately, there are no literatures, which have evaluated the recurrence rate of pregnancy-associated desmoid tumors following surgical excision specifically. However, based on reviewed documentations, it was noticed that abdominal wall desmoid tumors that arise during pregnancy seemed to have a lower recurrence rate compared with the other types of desmoid tumor. The reasons are not clear yet it may be associated with the postpartum drop of estrogen level.

## Conclusion

Desmoid tumor that arises during pregnancy is rare and the optimal management has not yet been well established. Simple observation is a reasonable management option for asymptomatic patients while surgical intervention has to be the first choice for patients who have significant symptoms or tumors increased rapidly in size, compressing the uterus and interfering with fetal growth. However, radiotherapy and pharmacological therapy are contraindicated during pregnancy, the role of postpartum radiotherapy, and pharmacological therapy remains controversial. The management of desmoid tumors diagnosed during pregnancy is complicated and the treatment must be individualized.

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