CASE REPORT

Vaginal delivery after hemipelvectomy and pelvic radiotherapy for chondrosarcoma

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SUMMARY

Chondrosarcoma of the proximal femur is a rare malignant disorder in women of (pre-) childbearing age, for which a radical resection through a hemipelvectomy could be indicated. We describe a case of a 36-year-old primigravida with a hemipelvectomy (2004) who had a history of radiotherapy of the pelvic and uterine regions after an atypical cartilaginous tumour. After an uncomplicated pregnancy, she had a spontaneous rupture of the membranes at 41+2 weeks and an uncomplicated vaginal delivery with physiological development of the infant. On the basis of the literature review, vaginal delivery after hemipelvectomy should be advocated and discussed with the patient and involved care providers. Women with radiotherapy in the pelvic and uterine areas have an increased risk of stillbirth, placental attachment disorders, impaired fetal growth, fetal malposition and preterm labour, but no association with prolonged rupture of the membranes has been described.

BACKGROUND

Chondrosarcoma is a malignant tumour with hyaline cartilage differentiation and accounts for 20% of tumours of the skeleton, the second most common primary tumour after osteosarcoma.^{1 2} The pelvic bones and proximal femur are the most common sites affected. Chondrosarcoma generally occurs in adults, with a peak incidence between 50 and 80 years and a slightly higher occurrence in men. On the basis of histopathology (nuclear staining/hyperchromasia, nuclear size and cellularity), chondrosarcomas are graded on a scale from 1 to 3, with increasing histopathological abnormalities and decreased prognosis. An atypical cartilaginous tumour, formally known as grade 1 chondrosarcoma, grows relatively slowly, has cells whose histological appearance is quite similar to cells of normal cartilage, and has much less aggressive, invasive and metastatic properties. Grades 2 and 3 are increasingly faster-growing cancers, with more varied and abnormal-looking cells, and are much more prone to infiltrate surrounding tissues, lymph nodes and organs. Some, but not all, authorities and medical facilities assign a 'Grade 4' to the most anaplastic, undifferentiated cartilage-derived tumours. The 5-year survival of grade 1 patients is 89–99%, and for grade 2 and grade 3 patients it is 53%.^{1 2} Grade 1 has a local recurrence rate of 9%.²

Billroth performed the first hemipelvectomy in 1891, though the patient only survived merely hours. The first successful hemipelvectomy was

performed in 1895 by Girard. The malignant tumours of the bone or soft tissue of pelvis osteosarcoma, chondrosarcoma and Ewing's sarcoma are the most common indications for the procedure.³ Figure 1 schematically illustrates three subtypes of hemipelvic resection approaches.³ Type I only includes the ilium, or the ilium and gluteal muscle (type Ia), or the ilium with a portion of the sacrum (type I/S). Type 2 is a resection of the periacetabular region, which can include the hip joint (type IIa). Type III constitutes a (partial) resection of the complete os ischium and os pubis.

Little is known about pregnancy outcomes in patients after hemipelvectomy, in particular with a history of radiotherapy in the lower abdominal region. We describe a case report with a review of the literature.

CASE PRESENTATION

In 1997, the patient had relapsing chondromatosis of the left femur, which subsequently developed into a chondrosarcoma grade I for which local resection of the left trochanter minor was indicated in 2002. Owing to local recurrence, this was repeated in 2003 and followed by a left hemipelvectomy type I with partial removal of the os ilium in 2004. For abdominal wall reconstruction, a Marlex mesh was placed. Postoperative radiotherapy started 7 weeks after hemipelvectomy. The

Figure 1 Subtypes of hemipelvectomy depending on resection sites, as presented in Wedemeyer and Kauther.³ Type I: ilium only; type IA: ilium and gluteal muscles; type I/S: ilium with a portion of the sacrum; type II: periacetabular region; type IIa: periacetabular region including the hip joint; type III: parts or complete ischium and pubis.

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ovaries were not transposed but recognised as organs at risk on the planning CT scan. The left lateral pelvis (whole operated area) was irradiated up to a dose of 50 Gy in 25 fractions, five times per week, using a three-dimensional conformal plan with anteroposterior parallel opposed 6 MV photon beams. This was followed by two separate boost fields to the bony resection margins. A dose of 10 Gy in five fractions was applied to the left pubic bone and to the left iliac wing, both using a three-field wedged beam set-up, avoiding the right ovary. The left ovary was irradiated up to the full dose, whereas the right ovary and tuba were spared, with a mean dose of 2.8 Gy. The uterus received a mean dose of 24.3 Gy and a maximum dose of 59.2 Gy, while the right side of the uterus was relatively spared. Relapsing infections and fistulas, hindering the prosthetic fitting, complicated primary recovery. The patient mobilises nowadays primarily with two elbow crutches and occasionally using a wheelchair. She works in public service and is an elected member of the municipality council and a former Paralympic coxed four rower. Bi-yearly bone scintigraphy and annual MRIs have shown no sign of local recurrence or metastasis.

A 36-year-old primigravida with a left hemipelvectomy presented at AD 14+3 weeks after spontaneous conception for prenatal care and clinical delivery. After an uncomplicated pregnancy, she had spontaneous rupture of the membranes at 41+2 weeks and delivered vaginally with physiological development of the infant. Placental abnormalities indicative of chorioamnionitis with a fetal response were observed in pathological examination of the placenta. Owing to suspected respiratory problems after birth, the infant was admitted to the medium care unit for 6 days, after which he was discharged in good health.

INVESTIGATIONS

Before pregnancy, in January 2012, a pelvic MRI was performed (figure 2). This MRI showed no evidence of disease and no contraindications for pregnancy or vaginal delivery.

At 14+3 weeks, the chromosomal abnormalities risk was estimated to be low based on the nuchal translucency test and levels of pregnancy-associated plasma protein-A and human chorionic gonadotropin levels. Structural ultrasound AD at 19+3 weeks showed no fetal abnormalities and a posteriorly localised placenta in fundo. Ultrasounds during pregnancy showed normal fetal growth (50th centile) and amniotic fluid. Prenatal laboratory results were unremarkable.

At 33+3 weeks, a bimanual pelvic examination was performed to assess possible restrictions for vaginal delivery. The right spina iliaca was normal. On the left side, at the location where usually ossal parts of the pelvis would be positioned, dense soft tissue likely to arise from muscle or connective tissue was felt. The fetal head was already engaged at this point in time, and the cervix was situated anteriorly and softened. No dilation was detected. In conclusion, there were no obstructions for a normal vaginal delivery.

DIFFERENTIAL DIAGNOSIS

Considerations during care were: marginally elevated risk of placenta abruption due to radiotherapy and elevated risk of pelvic instability and muscle herniation due to pelvic surgeries and association muscle manipulation.

TREATMENT

The pregnancy was uncomplicated and the fetus developed normally. On the basis of patient preference, MRI and consultations with the patient's hemipelvectomy orthopaedic surgeon, pelvic

floor-specialised gynaecologists and her rehabilitation physician, she was counselled for a vaginal delivery.

At 41+2 weeks, the patient presented with spontaneous rupture of the membranes with clear amnion fluid, for which expected management was suggested. After a total duration of 26 h, the patient developed spontaneous contractions. No raised temperature or other signs of infection were observed during this period. The first stage of labour was 6 h and 32 min; subsequently, the second stage of labour had a total duration of 32 min, after which the infant was born in the left occipitoanterior position. Computer-assisted cardiotocography indicated a good fetal condition throughout all phases of labour.

Active management of the third stage of labour was provided with oxytocin. The placenta was delivered 8 min later with a total blood loss of 500 mL. The subtotal sphincter rupture (grade IIIA) of the perineum was repaired.

OUTCOME AND FOLLOW-UP

The placenta was 495 g, with a paracentral insertion, two windings and a true knot at 45 cm. Despite there being no macroscopic anomalies, microscopic analysis showed a locally severe chorioamnionitis expanding into the chorial plate with micro-abscesses, indicative of a fetal response to inflammation.

The infant was a baby boy who weighed 3900 g (p50–80) and had an Apgar score of 7/7 due to persistent grunting and subcostal retractions. Umbilical venous pH was 7.29–12. The infant was admitted to the medium care unit on suspicion of a perinatal infection for antibiotic treatment for 6 days, after which he was discharged in good health. The mother recovered well without any sphincter rupture complications and stayed in the hospital until her child finished her course of antibiotics; they were discharged together and have been in good health since. During pregnancy, the patient mobilised increasingly with a skip chair. After delivery, a second skip chair was placed on the first floor of the residence to facilitate infant care, which was more convenient from a seated position. She continued to mobilise primarily with two elbow crutches. She resumed work, and was re-elected as a member of the municipality council in March 2014.

DISCUSSION

We present a case of a successful pregnancy and vaginal delivery after a hemipelvectomy and pelvic area radiotherapy. The combination of these treatments has been described only once by Nuss and Lee in 1967,⁴ though this pregnancy resulted in a caesarean section instead of a successful vaginal delivery as in the present case.

Pregnancy after hemipelvectomies

Table 1 presents an overview of the other cases presented in the literature,^{4–15} which includes the pregnancies of 15 women with a hemipelvectomy based on soft tissue and bone malignancies between 1926 and 2004. Of the 21 postoperative pregnancies, 15 were uneventful singleton pregnancies, two uneventful twin pregnancies, one second-trimester spontaneous abortion of twins, and two complicated by preeclampsia. Seventeen pregnancies ended in a vaginal birth, of which there were four in breech position and four with forceps extraction. Five pregnancies resulted in a caesarean section due to intrauterine growth restriction, prevention of stress or pain on the surgical scar, breech position, pelvic distortion and repeat caesarean section. The pregnancies resulted in 21 healthy neonates. This review illustrates that there is no evidence to support preference for a caesarean section over a vaginal delivery for women with a

Figure 2 (A and B) Preconceptional transversal MRIs of the pelvis.

hemipelvectomy without pelvic bone distortion after bone remodelling or a high risk of scar herniation. Rather, given the caesarean section associated risks (of infection, haemorrhage, injury to pelvic organs, future placental implantation abnormalities and increased risk of future caesarean delivery), vaginal delivery should be advocated.

Placenta function after radiotherapy

Table 2 presents a literature review of the pregnancy outcomes of women who received radiotherapy in childhood or early adulthood, including a quantification of effects.^{16–28} Radiotherapy in the abdominal and pelvic region is associated with an increased risk of low birth weight, prematurity,

Table 1 Overview of hemipelvectomy cases in the literature

Study	Maternal clinical information (age at diagnosis, diagnosis, treatment)	Pregnancy history in relation to hemipelvectomy (time since operation)	Pregnancy	Outcome
Judin ^{5*}	36 years, chondrosarcoma of the right femur, right hemipelvectomy in 1923	8 vaginal deliveries before hemipelvectomy, one pregnancy postoperative (1 year)	Uneventful	Spontaneous delivery, male 4200 g
Brockunier ⁶	20 years, chondrosarcoma of the right femur, right hemipelvectomy in 1957	2 postoperative pregnancies (1 year, 47 months)	Uneventful	g1=Spontaneous delivery, male 3160 g g2=Prolapsing cord causing fetal distress, low forceps delivery, male 3090 g
Cregan and Burslem ⁷	15 years, desmoid tumour in the left thigh, left hemipelvectomy in 1957	1 pregnancy postoperative (4 years)	Uneventful	Spontaneous delivery, complete breech, Maurice-Smillie-Viet manoeuvre, male 3380 g
	19 years, osteogenic sarcoma, left hemipelvectomy in 1955	1 pregnancy postoperative (4 years)	Uneventful	Caesarean section because of stress in the hemipelvectomy scar. Laevorotation of the uterus through 90°, female 3100 g
Ben-Aderet ^{8*}	27 years, echinococcal cyst in the left ilium, left hemipelvectomy, date unknown	1 pregnancy postoperative (2 years)	Uneventful, twins	Caesarean section for 'incipient hernia' of the scar, female 3052 g and male 2800 g
Nuss and Lee ⁴	25 years, myxoid liposarcoma in the left thigh, radiotherapy (6000 rad to the midplane of the thigh) and left hemipelvectomy in 1962	4 pregnancies postoperative, (18 weeks pregnant at time of diagnosis and treatment, 17 months, 31 months and 40 months)	g1=post-treatment severe atelectasis and tracheostoma, preeclampsia in third trimester; g2=uneventful, g3=premature rupture membranes, amnionitis, mid-second trimester abortion of twins; g4=uneventful	g1=Induction of labour, low forceps delivery, female 2140 g, stillborn g2=double foot-ling partial breech, female 2977 g g3=Spontaneous delivery, non viable twins g4=spontaneous delivery, double footling partial breech, male 3570 g
Holzaepfel ⁹	28 years, synovial fibrosarcoma of the right thigh. Right hemipelvectomy in 1969.	2 pregnancies postoperative (4 weeks gestation at diagnosis and treatment, 21 months)	g1=uneventful g2=uneventful	g1=induction of labour, low forceps extraction, female 2700 g g2=premature spontaneous delivery with footling breech, unknown
Inmon and Bledsoe ^{10*}	21 years, fibrosarcoma of the left femur, left hemipelvectomy in 1951	2 pregnancies postoperative (2 years, 11 years)	g1=uneventful g2=uneventful	g1=spontaneous delivery, sex unknown, 2700 g g2=spontaneous delivery, sex unknown, 2940 g
Stavarakas and Sanders ^{11*}	18 years, giant cell tumour of the pelvis, left hemipelvectomy in 1967	1 pregnancy postoperative (13 years)	Preeclampsia	Low forceps delivery, male 3742 g
Bergh <i>et al</i> ¹²	15 years, osteogenic sarcoma, left hemipelvectomy in 1977	1 pregnancy postoperative (10 years)	Uneventful	Spontaneously in labour, stagnation for which artificial rupture of membranes and pitocin augmentation, female 3005 g
Snyder and Thomas ^{13*}	18 years, rhabdomyosarcoma of the left upper leg, left hemipelvectomy in 1985	1 pregnancy postoperative (2 years)	g1=Uneventful, twins	g1=caesarean section, male 2430 g, female 3480 g
	22 years, osteogenic sarcoma, left hemipelvectomy in 1977	1 pregnancy preoperative, 1 pregnancy postoperative (3 years)	Uneventful	Repeat caesarean section, male 2080 g
Chihara <i>et al</i> ¹⁴	11 years, Ewing sarcoma left pelvic bone, chemotherapy and limb-sparing left hemipelvectomy	1 pregnancy postoperative (10 years)	Uneventful	Caesarean section because of pelvic distortion, male 2345 g
Heetkamp <i>et al</i> ¹⁵	28 years, chondrosarcoma of the right femur, right hemipelvectomy in 1998	1 pregnancy postoperative (9 years)	Uneventful	Spontaneous delivery, male 3550 g
Browne (2014)	20 years, chondrosarcoma of the left femur, radiotherapy pelvic and uterine regions, left hemipelvectomy 7 years post diagnosis in 2004	1 pregnancy postoperative (9 years)	Uneventful, prolonged rupture of membranes	Spontaneous delivery, two windings and true knot, chorioamnionitis, male 3900 g

*Full text not available, information inferred from subsequent publications.

Table 2 Overview of review of the literature on abdominal and pelvic irradiation and pregnancy outcomes

Study	Study population	Irradiation site	Outcomes
Chiarelli <i>et al</i> ¹⁶	Ontario Cancer Registry (1964–1988), 594 pregnancies	Abdomen and pelvis	Low birth weight (OR=3.64, 95% CI 1.33 to 9.96, dose–response) Premature low birth weight (OR=3.29, 95% CI 0.97 to 11.1, dose–response) Perinatal infant mortality (OR=2.41, 95% CI 0.50 to 11.5) No difference in spontaneous abortions and congenital abnormalities
Green <i>et al</i> ¹⁷	National Wilms Tumor Study Group (1969–1999), 427 pregnancies	Flank	Preterm delivery/threatened labour (OR=2.36, 95% CI 0.93 to 6.02 for >25 Gy) Malposition of fetus (OR=6.26, 95% CI 1.50 to 36.57 for >25 Gy radiation) Prematurity (OR=4.07, 95% CI 1.74 to 9.90 for >25 Gy) Lower birth weight (3.036±805 g vs 3.245±620 g, p=0.02 for Rx vs non-Rx women) Congenital malformations (10.5% Rx vs 3.2% for non-Rx, p=0.054)
Green <i>et al</i> ¹⁸	Childhood Cancer Survival group (1970–1986), 4029 pregnancies	Pelvis	Low birth weight (OR=1.85, 95% CI 1.07 to 3.18)
Signorello <i>et al</i> ¹⁹	Childhood Cancer Survivor Study (1968–2002), 2201 pregnancies	Uterus	Preterm delivery (OR=3.5, 95% CI 1.5 to 8.0 for >500 cGy with dose–response. Not significant in postmenarche radiation) Low birth weight (OR=6.8, 95% CI 2.1 to 22.2 for >500 cGy, with dose–response) Small for gestational age (OR=4.0, 95% CI 1.6 to 9.8 at >500 cGy with dose–response)
Winther <i>et al</i> ²⁰	Danish Cancer Registry (1950–1984), 1479 pregnancies	Not specified	Increased risk of spontaneous abortions (proportion ratio (PR)=1.58, 95% CI 1.2 to 2.2, especially with Rx to ovaries/uterus PR=2.8, 95% CI 1.7 to 4.7) No difference in live birth or stillbirths
Mueller <i>et al</i> ²¹	4 population-based cancer registries of Surveillance, Epidemiology and End Result (SEER) Program of the National Cancer Institution, US (1974–2000), 1898 pregnancies.	Not specified	No increased risk of gestational age of <37 weeks or low birth weight with Rx-only or Rx-surgery combination. Increased risk of radiotherapy in combination with chemotherapy (Relative Risk (RR)=2.14, 95% CI 1.27 to 3.63). Aggregated data ('any radiotherapy'): increased risk of <37 weeks gestation (RR=1.57, 95% CI 1.19 to 2.06) and low birth weight (RR=1.38, 95% CI 1.03 to 1.85)
Reulen <i>et al</i> ²²	British Childhood Cancer Survivor Study (1940–1991), 4113 pregnancies	Abdomen	Miscarriage (OR 1.4, 95% CI 1.0 to 1.9 overall, OR=1.9, 95% CI 1.1 to 3.2 for >12 weeks gestation) Premature birth OR=3.2, 91% CI 2.1 to 4.7) Low birth weight (OR 1.9, 95% CI 1.1 to 3.2) No increased risk for stillbirth
Green <i>et al</i> ²³	National Wilms Tumor Study Group (1969–2006), 1021 pregnancies	Flank	Hypertensive disorders in pregnancy (24% with >35 Gy, compared to 12.3 in total Rx population) Fetal malposition (10% with >35 Gy, 4.3% in total Rx population) Premature labour (30% at >35 Gy, 10.2% in total Rx population) No difference in congenital abnormalities.
Lie Fong <i>et al</i> ²⁴	The Netherlands (2000–2005) 9071 pregnancies (40 cancer survivors)	Abdomen	Birth weight (Rx: 2503±1026 vs 3266±705, p=0.02) Gestational age (Rx: 34.9±4.3 vs 39.2, p=0.001)
Madanat-Harjuoja <i>et al</i> ²⁵	Finnish cancer and birth registries (1987–2006), 1309 pregnancies	Abdomen/pelvis	Preterm delivery (OR=3.81, 95% CI 2.02 to 7.19)
Signorello <i>et al</i> ²⁶	Childhood Cancer Survivor Study (1968–2002), 4946 pregnancies	Uterus and ovaries radiation	Stillbirth and neonatal death (OR 9.1, 95% CI 3.4 to 24.6) at doses >10 Gy. Higher risk when Rx was before menarche
Sudour <i>et al</i> ²⁷	Childhood cancer (1975–2004), 84 pregnancies	Pelvis	Pelvic radiotherapy vs abdominal radiotherapy: higher incidence of prematurity and low birth weight
Madanat–Harjuoja <i>et al</i> ²⁸	Medical Birth Register (1987–), 3513 female patients with cancer 8636 sibling control group	Information not stratified by treatment modality	–

malposition of the fetus, stillbirth and neonatal death, with a dose–response relationship observed and a higher incidence of adverse outcome with younger or premenarche age. This is supposed to be a consequence of uterine damage consisting of uterine damage, with reduced blood flow, reduced endometrial thickness and less elasticity of uterus musculature.^{29–30} The risks

associated with radiotherapy in the abdominal and pelvic area did not occur in our patient. In fact, she delivered an infant at 41+2 weeks, at normal weight (50–80 centile). This may be a reflection of an age well beyond menarche at the time of therapy (20 years), despite a radiation dose at which all complications have been described (between >5 and 35 Gy).^{17–19, 23–26}

Patient's perspective

The fact that I got pregnant spontaneously, shortly after we had a wish for a child, gave us/me a strong feeling of trust. Trust in the strength and capability of my body to grow new life. The assistance given by the hospital was very good, we felt that we could discuss and ask everything, and this supported our good feeling of trust. Considerations in the treatment were well discussed. We experienced a warm contact between my oncological surgeon and gynaecologist. In the Revalidation Centre, a special bandage was made to support my stomach but it appeared that even without it, my pelvis was able to carry the baby.

In the beginning of my pregnancy, I was convinced that a natural birth would be impossible. Yet, after a while and a second opinion with a gynaecologist who guided a similar case, I began to believe that a normal birth would be possible. And so it happened. After an easy-going pregnancy, the delivery was not enjoyable—but it was very nice to do it the 'natural' way. We look back at a pleasant period.

Learning points

- Vaginal delivery after hemipelvectomy should be advocated and discussed with the patient and involved care providers.
- Hemipelvectomy is associated with an increased risk of fetal malposition due to the changed pelvic anatomy. Yet the reduced pelvic bone restriction also facilitates the delivery of a breech fetus.
- Pelvic and abdominal radiation results in a higher risk for stillbirth, placental attachment disorders, impaired fetal growth, fetal malposition and preterm labour.
- A dose-response relation with radiotherapy and complications is observed. Most complications are described in radiotherapy ranges of >25 to 35 Gy. Fewer adverse outcomes are observed in women who received radiotherapy after the onset of the menarche.

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