Original Article

Reflex sympathetic dystrophy in pregnancy: nine cases and a review of the literature


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Received 26 November 1998; accepted 10 February 1999

Abstract

Objective: To better understand the diagnosis of reflex sympathetic dystrophy of the lower extremities in pregnant women. Subject: Disease analysis using a retrospective series of nine cases and a review of the literature (57 patients and 159 sites of reflex sympathetic dystrophy). Results: This disorder should be considered in any painful pelvic girdle syndrome or lower extremity pain. The hip is involved in 88% of cases. Symptoms develop in the third trimester of pregnancy, between the 26th and the 34th weeks. Magnetic resonance imaging (MRI) provides an early, accurate, and very specific diagnosis, although standard radiography continues to be the first-line diagnostic tool. Fracture occurs in 19% of patients. The etiology and pathophysiology remain unclear, although pregnancy itself appears to play a significant role in this disease. Although locoregional mechanical factors partly explain reflex sympathetic dystrophy, hypertriglyceridemia appears to be a risk factor. This disorder develops independently, but the conclusion of pregnancy appears to be necessary for cure. Reflex sympathetic dystrophy does not appear to affect the course of the pregnancy. Indications for cesarean delivery remain obstetrical and should be discussed when a fracture is involved. Simple therapeutic management using gentle physical therapy provides rapid and complete recovery in 2–3 months. Conclusion: Reflex sympathetic dystrophy during pregnancy remains poorly understood and underestimated. Only joints of the inferior limbs are involved. MRI appears to be the best diagnostic tool. Pathogenesis remains unclear. Fractures are not rare. Treatment should be non-aggressive.

Keywords: Pregnancy; Reflex sympathetic dystrophy; Rheumatology; Sudeck’s syndrome

1. Introduction

Pelvic or lower extremity pain during pregnancy poses certain diagnostic difficulties. Although pregnancy can alter the course of some inflammatory disorders, it can also lead to the emergence of mechanical joint diseases, such as reflex sympathetic dystrophy of the lower extremities. Reflex sympathetic dystrophy (RSD) is a type of arthropathy combining a painful syndrome with locoregional trophic disturbances. This poorly understood disorder, which is frequently deceptive and sometimes clinically misleading in the pregnant woman, was first described in 1959 by Curtiss and Kincaid [1]. They first described three cases of transient demineralization of the hip during pregnancy. Lequesne [2], in 1968, described the same pathology and called it transient osteoporosis or neurotrophic rheumatism. Later, other terms were used: migratory osteolysis, reflex sympathetic dystrophy syndrome, algoneuropathy, and Sudeck’s atrophy or syndrome [3–5]. Currently these very similar clinical disorders are grouped as RSD in the American English medical literature. So far only case reports have been published in the literature. Therefore we were prompted to analyse in a
series of nine cases of RSD: (a) the characteristics of the patients, (b) the relation between the evolution of the pregnancy and the occurrence of RSD and (c) the outcome of the pregnancy.

2. Materials and methods

The clinical cases were compiled retrospectively over a period of 3 years, from May 1994 to May 1997, from four French services: the Rheumatology Service at the Argenteuil Hospital Center, and the maternity wards at Pontoise Regional Hospital Center, Lariboisière University Hospital Center in Paris, and Poissy Hospital Center. Only cases with certain diagnosis (based on clinical examination, paraclinical diagnostic tools and evolution after treatment and delivery) were involved in this series. During this time, about 8000 deliveries were recorded in the three maternity wards.

3. Results

Nine cases of RSD occurring during pregnancy were found (Table 1). The average age of the patients was 36±3.5 years (range, 30–43). Only two patients were primiparas. The average interval from conception to the appearance of symptoms was 28.1±2.2 weeks (range, 25–32). There were eight singleton pregnancies and one spontaneous triplet pregnancy. The only abnormal sign observed before the onset of the RSD was excessive weight gain, without gestational diabetes, for six of the patients (weight gain superior to 12 kg). In two other cases the total weight gain was only 3 kg, but there was significant initial obesity. In one case the weight gain was not recorded.

Any of the joints of the lower extremity may be involved. The hip was most frequently involved: in seven patients and in 11 of 21 involved joints (52%). Bilateral involvement was observed in four cases. There was no apparent predominant side: seven cases occurred on the right and six on the left. A joint group may become involved, at the outset or later on. Thus, in one case, the involvement of one hip even occurred postpartum.

The diagnosis of RSD was made at the outset in only one patient. In six cases the initial diagnosis was sciatic-type pain, and in two cases, phlebitic pain.

Clinically, the mechanical symptomatology appeared gradually, and the clinical signs were nonspecific. Functional impairment, sometimes total, was commonly found. Limping was observed in eight cases. Pain was always present, often poorly localized, and of variable severity. In only two cases did we find symptomatology characteristic of an inflammatory phase. In three cases we noted a ‘clinostatic’ syndrome (that is, defined by weakness of the lower limb in the supine position contrasting with none or only slight weakness upon standing, a normal neurological examination and full passive mobility of the hip). When a reduction in joint mobility was present, it was mild, in contrast with the apparent severity of the impairment.

The diagnosis was made using standard radiography in eight cases, sometimes after several weeks’ delay. In most cases the X-rays were initially normal but subsequently revealed areas of patchy bone demineralization in the involved joint. In the hip, the femoral head sometimes appeared ‘ghostly’. The joint space remained intact in all cases (Fig. 1). The radiological findings did not always correspond to the clinical signs. Magnetic resonance imaging (MRI) was performed in four cases and revealed a characteristic image each time, thus providing an early diagnosis. There was low signal intensity on T1-weighted images and high signal intensity on T2-weighted images in the affected joints (Figs. 2 and 3).

Treatment was symptomatic until delivery, combining non-weight-bearing with rest and non-narcotic analgesics. Physical therapy was gradually undertaken and was carried out only to the point of pain. After delivery, six patients began a calcitonin-based treatment (Calsyn® synthesis Calcitonin SPECIA® Laboratory Paris France 100 IU/day for 8 days, then 100 IU/1 3 times a week for 3 weeks). Independent of the treatment undertaken, clinical recovery was rapidly obtained, with an average time to recovery of

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## Table 1

<table>
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<tr>
<th>Age</th>
<th>P</th>
<th>Date</th>
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<th>Side</th>
<th>Recovery</th>
<th>Delivery term</th>
<th>Weight (PERC)</th>
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<td>4</td>
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<td>Hip+knee</td>
<td>R</td>
<td>6</td>
<td>V, 38,5WA</td>
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<tr>
<td>(1998)</td>
<td>30</td>
<td>1</td>
<td>30</td>
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<td>7</td>
<td>CESAR, 38 WA</td>
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<tr>
<td></td>
<td>43</td>
<td>4</td>
<td>26 1/2</td>
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<td></td>
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<td>29</td>
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<td>*</td>
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<tr>
<td></td>
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<td>24</td>
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<td>8</td>
<td>26/PP</td>
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<td>L/R</td>
<td>3</td>
<td>V, 40 WA</td>
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<tr>
<td></td>
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<td>6</td>
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<td>25</td>
<td>Ankle</td>
<td>L</td>
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<td>CESAR, 34,5 WA</td>
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<tr>
<td></td>
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<td>5</td>
<td>30</td>
<td>Hips</td>
<td>R+L</td>
<td>10</td>
<td>V, 38 WA</td>
</tr>
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</table>

* P, parity; date, date of sign apparition in weeks of amenorrhoea; R, right; L, left; recovery, in weeks; PP, post-partum, *, unknown; V, vaginal delivery; CESAR, caesarean section; WA, weeks of amenorrhoea; weight, in grams.
Fig. 1. Standard radiograph: homogeneous demineralization of the right femoral head, with joint space intact.

Fig. 2. MRI of the hips, T1-weighted images (coronal sections): left hip, fuzzy contour of homogeneous low signal intensity in the femoral neck; right hip, discrete area of low signal intensity in the inferior femoral head.
9 ± 7.3 weeks (range, 3–26). The time to radiographic normalization was longer.

All of the fetuses were in the cephalic presentation and were full-term at birth, except for the triplet pregnancy (34 weeks and 2 days). Seven deliveries were vaginal and only one was by forceps. There were two cesarean deliveries, one for cephalopelvic disproportion and the other for a triplet pregnancy complicated by preeclampsia. The first triplet was born in the cephalic presentation. The average weight of the newborns was 3497 ± 512 g (range, 2550–4020 g), excluding the triplet pregnancy. Six newborns had birth weights above the 75th percentile according to Leroy’s curves [6].

4. Discussion

The prevalence of RSD in pregnant women is unknown. According to series of RSD [3,7–11], pregnancy accounts for 0.5–33% of the cases of RSD reported in women and seems to be the etiology found most frequently among them. The incidence of RSD during pregnancy is very likely underestimated [12–14]. It is likely that many mild cases of pelvic pain remain undiagnosed, particularly in the obstetrical field, due to poor understanding of this disorder as well as the fact that it spontaneously resolves.

According to our results, the clinical symptomatology is nonspecific, aside from its insidious, gradual onset. This onset contrasts with that of various other etiologies, particularly of vertebropelvic origin such as fracture, surgery [14]. In cases involving the hip, there is usually mechanical pain. It is located in the inguinal area and sometimes radiates to the anterior thigh or knee. Functional impairment with unsteady support causes limping. In severe forms, whether uni- or bilateral, walking with crutches is difficult, even impossible. This severity often contrasts with the moderate limitation of passive movement found upon examination, which often (but not always) demonstrates a slight restriction of movements at the extremes of the range of motion [14]. In cases of more distal involvement, edema and cutaneous vasomotor dysfunction are more marked and are often the primary symptoms. A clinostatic syndrome may be seen [2]. Three patients in our series were unable to raise their heel from the bed in the decubitus position. In the great majority of cases, these phenomena appear for no apparent reason, with no history of recent trauma, and there is frequently a specific pain-relieving position. The pain is exacerbated upon standing and improves in the decubitus position. During pregnancy, the affected joints are exclusively located in the lower extremities. There has been only one case of shoulder involvement, described by Hunder in 1968 [14]. It appeared, however, 3 months after delivery, which makes pregnancy unlikely as the cause. Joint
involvement is not always synchronous. Exacerbation of pain immediately following delivery is possible. Recurrences in subsequent pregnancies have been described [14,15].

A review of the literature uncovered 57 reported cases, involving 159 sites (Tables 2 and 3) [1–3,5,7,8,13–27]. The patients’ average age was 32 years (range, 20–43 years). The peak occurrence of RSD and the onset of symptoms was at 6 months (range 24–32 weeks) (Fig. 4). The patients were primarily primiparas (57% of 49 usable cases).

The location of the lesions reveals various tendencies. Of 57 patients, 51 presented with at least one involved hip (88%). Of the 159 lower extremity sites, 86 (54%) involved the hip, with bilateral involvement in almost one-third of the cases. The knee was involved in 40 cases (25%) and an ankle or foot in 33 cases (21%). The entire lower extremity was involved in 13 cases (8%) (Fig. 5). Contrary to what has customarily been reported, the incidence of involvement of the two hips is the same [1,4,12–14,24].

In our experience we did not found any fracture. But RSD can be complicated by fractures (19%). The fracture may be displaced, nondisplaced or impacted; it occurs in a pathological, demineralized, fragile bone. There is usually a minimal, if any, history of trauma. The fracture may be located at the level of the femoral head, the femoral neck or the pelvis, involving the pubic rami. Surgery is seldom required, but even when it is indicated, tissue and bone healing occur within the normal time periods. Bone affected by RSD heals as fast and as well as normal bone. Some authors have even described simultaneous complete healing of the fracture and resolution of the reflex sympathetic dystrophy syndrome [1,7,8,15–18,20,22,23].

Laboratory examination is relatively unhelpful. The sedimentation rate and hydroxyprolinuria do not provide any diagnostic clarification [1,12,13,15,16,18,21]. The only significant change is hypertriglyceridemia. Acquaviva et al. [9], in 765 cases of RSD, noted that hypertriglyceridemia was significantly more frequent in cases of RSD involving the lower extremities, particularly the hip. Like Amor [28], they believe that this laboratory change constitutes a risk factor without being a determining component of the disorder. However, hypertriglyceridemia

Table 2
Review of the literature

<table>
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<tr>
<th>Author</th>
<th>N</th>
<th>Age</th>
<th>Term</th>
<th>Localisation</th>
<th>Side</th>
<th>Complication</th>
<th>Recovery</th>
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<td>3</td>
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<td>R</td>
<td>Neck fracture</td>
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<td>De Seze (1960)</td>
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<td>Foot</td>
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<td>R</td>
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<td>R/L</td>
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<td>Hip</td>
<td>R</td>
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<td>L</td>
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<td>L</td>
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<td></td>
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<td></td>
<td>Hip</td>
<td>L</td>
<td></td>
<td>12</td>
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N, number of patients; R, right; L, left; term, term of first manifestation in weeks of amenorrhea; PP, post-partum; IL, inferior limb; recovery, in weeks; *, unknown.
is common during pregnancy. It begins early and is significantly elevated even in the first trimester. It continues to increase during pregnancy and reaches a maximum at the end of pregnancy [29]. In our series, the eight patients in whom a lipid profile was performed (specifically, serum triglyceride levels) demonstrated a significant hypertriglyceridemia that subsided in the postpartum period.

Magnetic resonance imaging provides an early diagnosis, with characteristic images. The T1-weighted images show regional low signal intensity and T2-weighted images show regional high signal intensity. These changes appear within 48 h after the onset of symptoms and normalize in 6–8 months [7,8,16,19,21,31]. This is currently the key study for the diagnosis of RSD when faced with an unclear clinical picture, particularly during pregnancy [7,19,21,27,31].

Standard radiography is helpful in making the diagnosis in the great majority of cases. The radiological lesions, however, appear late, 3–6 weeks after the clinical symp-

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### Table 3
**Review of the literature**

<table>
<thead>
<tr>
<th>Author</th>
<th>N</th>
<th>Age</th>
<th>Term</th>
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<th>Side</th>
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</tbody>
</table>

* N, number of patients; R, right; L, left; term, term of first manifestation in weeks of amenorrhea; PP, post-partum; IL, inferior limb; recovery, in weeks; *, unknown.
seems to be a determining factor for recovery [4,21]. Specific treatment of RSD is recommended. It seems [3,13±15,18,19,21,24,30].

Conclusion of the pregnancy average of 2±3 months. According to our series, non-symptoms in the days following delivery is common postpartum period. Complete resolution was obtained in an faster than in other contexts; however, a rebound in was suficient. Calcitonin was occasionally helpful in the ly after delivery. In addition, this type of RSD resolves rest, gentle physical therapy, and non-narcotic analgesics uncomplicated RSD resolves spontaneously and complete- In our series, treatment consisting of non-weight-bearing layed. Treatment shortens the time to recovery, although women appears to be multifactorial.

weeks or months. The radiographic normalization is de- pregnancy that have yet to be elucidated. RSD in pregnant occurred in the first trimester and resolved after voluntary explained. Chigira [4], for example, reported a case which microthrombi and phlebitis in the bone, which could itself itself, an indication for a cesarean delivery. The involved combination of these various disturbances may lead to produce or maintain autonomic dysfunction.

in early stage of disease, bone scintigraphy provides diagnosis, revealing homogeneous uptake in the area of the involved joints. Multifocal involvement may also be demonstrated by total body scanning. The areas of increased uptake persist more than 6 months and thus provide a retrospective diagnosis. This study is less specific than radiography or MRI, however, and is con-traindicated during pregnancy [7,13,16,19,21,24,28].

Rarely performed, pathological examination of synovial or bone biopsies reveals nonspecific inflammation or lesions of vascular origin, with hemorrhagic medullary stasis in combination with foci of fibrosis, and in the trabeculae, accelerated turnover with a combination of osteoclastic resorption and osteoblastic regeneration [4,16,18,20].

No author has discussed the possible obstetric implic- ations. Type of delivery is only mentioned for 33 cases. Twenty-six patients had vaginal deliveries; four of those required forceps. No breech deliveries have been reported. Seven cesareans were reported: two due to cephalopelvic disproportion, two for orthopedic indications and three for which no indication was noted. These data do not provide any definite conclusions but it appears that RSD in pregnant women does not cause dystocia, nor is it associ- ated with an increase in premature births.

According to our findings, in any case, RSD of the lower extremities in pregnant women is not, in and of itself, an indication for a cesarean delivery. The involved joints should, of course, be manipulated with care during delivery to avoid fracture.

The differential diagnosis of RSD includes sciatica, neoplasia, inflammatory disorders, tuberculosis, infection, aseptic necrosis of the femoral head [7,12,13,16,18,19,30], the osteoligamentous or Lacomme’s syndrome [13,18,19,30], symphysis disjunction [13], phlebitis [14], or osteomalacia.

The course of RSD of the hip during pregnancy is generally benign, with resolution of the pain in several weeks or months. The radiographic normalization is de- layed. Treatment shortens the time to recovery, although uncomplicated RSD resolves spontaneously and complete- ly after delivery. In addition, this type of RSD resolves faster than in other contexts; however, a rebound in symptoms in the days following delivery is common [3,13–15,18,19,21,24,30]. Conclusion of the pregnancy seems to be a determining factor for recovery [4,21].

Although there are relatively few published cases, the association of RSD and pregnancy does not appear to be random. Pregnancy may well have a causative role, given the exclusive involvement of the lower extremities, particularly the hip, the clinical course including rapid re- covery after delivery, and the possible recurrence in subsequent pregnancies [8,13].

Although various hypotheses have been put forth, the pathogenesis of RSD in pregnancy remains unclear. Curtiss and Kincaid [1] have suggested the possibility of intermittent mechanical compression on the maternal left obturator nerve by the fetal head. Although animal studies involving compression or section of the obturator nerve in bitches have not supported this hypothesis, these experiments were performed in nonpregnant bitches. Moreover, they purported to explain the greater involvement of the left hip by the greater prevalence of the left occipitoiliac position in the cephalic presentation. We have noted, however, that both hips are equally likely to be involved and that bilateral hip or even bilateral lower extremity involvement may occur. On the other hand, no breech deliveries have been reported. Nevertheless, in pregnant women, intermittent compression of one or both obturator nerves might induce autonomic dysfunction.

The exclusive involvement of the lower extremities would seem to argue for locoregional causes. During pregnancy, significant weight gain, hyperlordosis and the weight of the infant alter the mechanical conditions and multiply the microtrauma at the level of the femoral head and neck. These sensory stimuli may irritate the autonomic system. This well-known mechanism for RSD induces spasm of the arteriole or the precapillary sphincter. These alterations cause backward filling of capillaries, with passive dilation of venules as well. This maintains vascular stasis by a neurologically induced mechanism in addition to the vascular stasis induced by the compression on the inferior vena cava by the gravid uterus [32,33]. The combination of these various disturbances may lead to microthrombi and phlebitis in the bone, which could itself produce or maintain autonomic dysfunction.

Furthermore, hypertriglyceridemia appears to be widely recognized as a risk factor in RSD of the hip [9,13,28].

However, despite these hypothesis, clinical occurrence of RSD during the first trimester of pregnancy cannot be explained. Chigira [4], for example, reported a case which occurred in the first trimester and resolved after voluntary termination of the pregnancy. That may be an isolated case, or it may be a case that involves factors related to pregnancy that have yet to be elucidated. RSD in pregnant women appears to be multifactorial.

In our series, treatment consisting of non-weight-bearing rest, gentle physical therapy, and non-narcotic analgesics was sufficient. Calcitonin was occasionally helpful in the postpartum period. Complete resolution was obtained in an average of 2–3 months. According to our series, non-specific treatment of RSD is recommended. It seems logical to summarize: non-weight-bearing rest, non-nar-
cotic analgesics, measures to counter venous stasis. It is best not to attempt early rehabilitation in RSD. By avoiding any rough or painful motion, physical therapy and careful mobilization can be helpful in the initial stage. Remaining pain-freer avoids nociceptive stimulation that might perpetuate the RSD. It also serves as a preventive measure against fractures [18]. Physical therapy in terms of cold treatments and assisted active mobilization are frequently combined. For treatment of pain, several regiments have been recommended such as: calcitonin, beta-blockers, corticosteroids [14,22]. However, Calcitonin is routinely contraindicated in pregnancy and local corticosteroids provide very little benefit.

5. Conclusion
Our results emphasize the underestimated rate of RSD during pregnancy. Only joints of the inferior limbs are involved. MRI appears to be the best means of confirming the diagnosis. The pathogenesis remains unclear. A better understanding of this disease will enable us to avoid diagnostic errors, procedures, and aggressive treatment, which are sometimes harmful and always unwelcome during pregnancy.

6. Condensation

Reflex sympathetic dystrophy of the lower extremities is a diagnosis to be considered in any pregnant woman with pelvic girdle or lower extremity pain. The diagnosis can be confirmed by nuclear magnetic resonance imaging. Treatment is symptomatic.

Acknowledgements

We specially thank Jo Ann Cahn for translation.

References


