West Middlesex University Hospital

**DISCLAIMER:** Results of database and or Internet searches are subject to the limitations of both the database(s) searched, and by your search request. It is the responsibility of the requestor to determine the accuracy, validity and interpretation of the results.

**Date:** 8 January 2018
**Sources:** Medline, Embase.

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1.
**Complications of pregnancy and labour in women with Klippel-Trenaunay syndrome: a nationwide cross-sectional study.**
Horbach SE; Lokhorst MM; Oduber CE; Middeldorp S; van der Post JA; van der Horst CM.
Ovid MEDLINE(R) Epub Ahead of Print, In-Process & Other Non-Indexed Citations, Ovid MEDLINE(R) Daily, Ovid MEDLINE and Versions(R)
BJOG: An International Journal of Obstetrics & Gynaecology. 124(11):1780-1788, 2017 Oct.
[Journal Article]
UI: 28432715
OBJECTIVE: To evaluate complications of pregnancy, including thromboembolism, in women with extensive vascular malformations associated with Klippel-Trenaunay syndrome (KTS).
DESIGN: Nationwide cross-sectional study.
SETTING: Two tertiary expert centres and the Dutch Klippel-Trenaunay patient organisation.
SAMPLE: Adult women with KTS.
METHODS: Patients with KTS were invited to participate in a comprehensive online survey about their obstetric history. Reference data on pregnancy outcomes and complications of non-diseased women were collected from population-based cohorts from the literature.
MAIN OUTCOME MEASURES: Prevalence of complications, specifically venous thromboembolism and postpartum haemorrhage.
RESULTS: Sixty women completed the survey. Seventeen patients did not conceive, of whom three refrained from pregnancy because of KTS. A total of 97 pregnancies and 86 deliveries were reported in 43 patients. KTS-related symptoms were aggravated during pregnancy in 43% of patients. Deep vein thrombosis was present in 5.8% and pulmonary embolism was present in 2.3% of pregnancies, which was extremely high compared with the reference population ($P < 0.0001$), with a relative risk of 108.9 (95% confidence interval, 95% CI 46.48-255.03) and 106.2 (95% CI 26.97-418.10), respectively. Severe postpartum haemorrhage (PPH) occurred in 11% of KTS pregnancies, compared with 5.8% of pregnancies in the reference population (relative risk, RR 1.81, 95% CI 0.97-3.37, $P = 0.06$).
CONCLUSIONS: Our data suggest that women with KTS have a significant risk of venous thromboembolic events, severe postpartum haemorrhage, and aggravation of KTS symptoms during pregnancy, and in early postpartum period. Obstetricians should counsel patients about these risks in the preconception phase. Antithrombotic prophylaxis should be considered in the obstetric management of patients with KTS.
TWEETABLE ABSTRACT: High risk of complications during pregnancy and labour in women with Klippel-Trenaunay syndrome.

Version ID

1
Status
In-Data-Review
Authors Full Name
Horbach, S E; Lokhorst, M M; Oduber, C E; Middeldorp, S; van der Post, J A; van der Horst, C M.
Institution
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van der Horst, C M. Department of Plastic, Reconstructive, and Hand Surgery, Academic Medical Centre (AMC), University of Amsterdam, Amsterdam, the Netherlands.
Date Created
20170422
Year of Publication
2017

**2.**

**A 39-Year-Old Pregnant Woman with Pulmonary Emboli on Long Term Anticoagulation.**

Mehta V; Bhatia K; Dave AM; Depew ZS.

Ovid MEDLINE(R) Epub Ahead of Print, In-Process & Other Non-Indexed Citations, Ovid MEDLINE(R) Daily, Ovid MEDLINE and Versions(R)

Cureus. 9(6):e1356, 2017 Jun 15.

[Journal Article]

UI: 28721324

We present the case of a 39-year-old pregnant woman with Klippel-Trenaunay syndrome (KTS). We demonstrate the risks of multiple, co-existing pro-thrombotic states (pregnancy, KTS), discuss complications of KTS (deep venous thromboembolisms and pulmonary emboli) and highlight general and disease-specific preventive measures against venous thromboembolic events (VTE). KTS is a rare condition and it's co-existence with pregnancy and VTEs is rarer still.

Version ID

1

Status

PubMed-not-MEDLINE

Authors Full Name

Mehta, Vishisht; Bhatia, Karishma; Dave, Amanda M; Depew, Zachary S.

Institution

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Date Created

20170719

Year of Publication

2017

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Link to the External Link Resolver:

NHS LinkResolver

**3.**

**A case of Klippel-Trenaunay syndrome (KTS) in pregnancy.**

Myers B., Tam J., Siddiqui F.

Embase
Thrombosis Research. Conference: 7th International Symposium on Women's Health Issues in Thrombosis and Haemostasis. Spain. 151 (Supplement 1) (pp S132), 2017. Date of Publication: March 2017.
[Conference Abstract]
AN: 619997536

A 29-year old Caucasian woman with known KTS was referred into the haematology- obstetrics clinic at 24 weeks. Anomaly scan and MRI were performed and showed extreme vascular uterine wall of 7.6cm in depth. (see picture). The fetus was at high risk of intra-uterine growth retardation and fetal death, and our patient was at high risk of antepartum/post-partum haemorrhage and high cardiac output failure. MDT decision was to proceed to an elective caesarean section and hysterectomy. 5L of red blood cell was crossed-match preoperatively. Under general anaesthetic, bilateral internal iliac artery balloons were inserted, baby delivered via a long midline laparotomy, and a hysterectomy performed. Bleeding from the uterine wall was controlled with soft bowel and vascular clamps and deep tension sutures. Despite this, total blood loss was 6.5L. Cell salvage was used and patient was transfused, transferred to intensive for monitoring and made an uneventful recovery. KTS is a rare condition with few case reports in pregnancy. Morbidity of the disease is associated with vascular anomalies resulting in rectal haemorrhage, haematuria or peri-operative bleeding in surgeries that compromise intra-abdominal organs. There is also an increased risk of thromboembolism such as deep vein thrombosis and pulmonary embolism. The thrombotic risk and haemorrhagic risks are further exacerbated by normal physiological changes in Multidisciplinary approach is key in managing these patients. Thoughtful preconceptual counselling, systematic intrapartum care and MRI imaging are likely to reduce mortality and morbidity. These aspects were key to improving the safety of delivery in this case.

Status
CONFERENCE ABSTRACT

Institution
(Myers, Tam, Siddiqui) University Hospitals of Leicester, Leicester, United Kingdom (Myers) United Lincolnshire Hospitals, Lincoln, United Kingdom

Publisher
Elsevier Ltd
Date Created
20171231
Year of Publication
2017

**4.**

**Pregnancy in women with Klippel-Trenaunay syndrome: Report of three pregnancies in a single patient and review of literature.**

Keerthana K., Keepanasseril A., Maurya D.K., Kadambari D., Sistla S.

Embase
Obstetric Medicine. 10 (4) (pp 177-182), 2017. Date of Publication: 01 Dec 2017.
[Article]
AN: 619492604

Klippel-Trenaunay syndrome is characterised by vascular abnormality which increases the risk of thromboembolism and haemorrhage. Physiological changes in pregnancy pose an increased risk to these complications. Being an uncommon disorder, there is limited literature about the management of women with pregnancy and Klippel-Trenaunay syndrome. We report in detail two of three pregnancies in a woman with Klippel-Trenaunay syndrome who had repeated episodes of haematochezia leading to anaemia, managed with Argon laser Photo-Coagulation in pregnancy and also reviewed the complications and the management of pregnant women with Klippel-Trenaunay syndrome.



Status
Embase

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Publisher
SAGE Publications Inc. (E-mail: claims@sagepub.com)
Date Created
20171208
Year of Publication
2017

**5.**

**A case of Klippel Trenaunay Syndrome in pregnancy.**

Tam J., Siddiqui F., Sayers R., Myers B.

Embase

British Journal of Haematology. Conference: 57th Annual Scientific Meeting of the British Society for Haematology. United Kingdom. 176 (pp 115-116), 2017. Date of Publication: March 2017. [Conference Abstract]

AN: 615322615

We report of a case of pregnancy in a patient with Klippel-Trenaunay syndrome (KTS). Our patient presented with a large uterine haemangioma and was managed under obstetrics, haematology, radiology and vascular surgeons throughout the pregnancy. A healthy neonate was delivered electively at 30-week gestation. Her bleeding risk was extremely high and the patient lost 6.5 L of blood postpartum. Patient underwent hysterectomy postpartum as planned and received prophylactic low molecular heparin post partum. The patient was a 29-year old Caucasian woman with known KTS. She was seen in haematology- obstetrics clinic and was counseled about the supportive care required for the condition. She was advised to receive low molecular heparin during the antepartum period and postpartum period. Anomaly scan and MRI at 24 weeks of gestation showed an extreme vascular uterine wall of 7.6 cm in depth. The fetus was at high risk of intra-uterine growth retardation and fetal death. Our patient was at extremely high risk of antepartum and post-partum haemorrhage and significant risk of high cardiac output failure. MDT decision was to proceed to an elective caesarian section and hysterectomy. 5 L of red blood cell was crossed-match preoperatively. The patient was sedated under general anaesthetic, and bilateral internal iliac artery balloons were inserted. The baby was delivered via a long midline laparotomy, and a hysterectomy performed. Bleeding from the uterine wall was controlled with soft bowel clamps, vascular clamps and multiple deep tension sutures to stop the bleeding. Despite this, total blood loss was 6.5 L. Cell salvage was used and patient was transfused. She was transferred to intensive for monitoring and made an uneventful recovery. KTS is a rare condition with few case reports in pregnancy. Morbidity of the disease is associated with vascular anomalies resulting in rectal haemorrhage, haematuria or peri-operative bleeding in surgeries that compromise intra-abdominal organs. There is also an increased risk of thromboembolism such as deep vein thrombosis and pulmonary embolism. The thrombotic risk and haemorrhagic risks are further exacerbated by normal physiological changes in pregnancy such as increased venous pressure, leg oedema, venous stasis and cardiac output. Multidisciplinary approach is the key in managing these patients. Thoughtful preconceptual counseling, along with methodical and systematic intrapartum care including MRI imaging are likely to reduce mortality and morbidity. These aspects were key to improving the safety of delivery in this case.

Status

CONFERENCE ABSTRACT

Institution

(Tam) Haematology, United Kingdom (Siddiqui) Obstetrics and Gynaecology, United Kingdom (Sayers) Vascular Surgery, University Hospitals of Leicester NHS Trust, United Kingdom (Myers) Haematology, University Hospitals of Leicester NHS Trust, Leicester, United Kingdom (Myers) Haematology, United Lincolnshire Hospitals NHS Trust, Lincoln, United Kingdom

Publisher

Blackwell Publishing Ltd

Date Created

20170414

Year of Publication
2017

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Link to the External Link Resolver:
NHS LinkResolver

**6.**
**Diffuse Venous Malformation of the Uterus in a Pregnant Woman with Klippel-Trenaunay Syndrome Diagnosed by DCE-MRI.**
Yara N; Masamoto H; Iraha Y; Wakayama A; Chinen Y; Nitta H; Kinjo T; Aoki Y.
Ovid MEDLINE(R) Epub Ahead of Print, In-Process & Other Non-Indexed Citations, Ovid MEDLINE(R) Daily, Ovid MEDLINE and Versions(R)
Case Reports in Obstetrics and Gynecology. 2016:4328450, 2016.
[Journal Article]
UI: 27006845
Background. We experienced a rare case of a pregnant woman with Klippel-Trenaunay syndrome complicated with diffuse venous malformation of the uterus. This is the first report on the usefulness of dynamic contrast-enhanced-MRI for the diagnosis of diffuse venous malformation of the uterus. Case Presentation. A 23-year-old woman presented with convulsions and talipes equinus position of both lower limbs at 11 weeks of gestation. At 27 weeks, ultrasonography demonstrated tubular echolucent spaces throughout the myometrium. Dynamic MRI at 37 weeks revealed that the myometrial lesion was enhanced slowly and showed homogeneous enhancement even on a 10min delayed image. Taken together with unilateral foot hypertrophy, varices, and port-wine stain, the patient was diagnosed as having Klippel-Trenaunay syndrome complicated with diffuse venous malformation of the pregnant uterus. The patient underwent elective cesarean section because of severe dystonia. The lower uterine segment was thickened and heavy venous blood flow was observed at the incision. Histological diagnosis of the myometrial biopsy specimen was venous malformation. Conclusions. Both diffuse venous malformation and Klippel-Trenaunay syndrome during pregnancy can involve considerable complications, in particular, massive bleeding during labor. Women who suffer from this syndrome should be advised about the risk of complications of pregnancy.
Status
PubMed-not-MEDLINE
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Authors Full Name
Yara, Nana; Masamoto, Hitoshi; Iraha, Yuko; Wakayama, Akihiko; Chinen, Yukiko; Nitta, Hayase; Kinjo, Tadatsugu; Aoki, Yoichi.
Institution
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Aoki, Yoichi. Department of Obstetrics and Gynecology, Graduate School of Medicine, University of the Ryukyus, 207 Uehara, Nishihara, Okinawa 903-0215, Japan.
PMID
https://www.ncbi.nlm.nih.gov/pmc/articles/PMC4783561
Date Created
20160323
Year of Publication
2016

**7.**

**Klippel-trenaunay syndrome, pregnancy, and pulmonary embolism: When prophylaxis is not enough!.**

Mehta V., Bhatia K., Kandasamy V.V., DePew Z.

Embase
Chest. Conference: CHEST 2015. Montreal, QC Canada. Conference Publication: (var.pagings). 148 (4 MEETING ABSTRACT) (no pagination), 2015. Date of Publication: October 2015.
[Conference Abstract]
AN: 72132784

INTRODUCTION: Klippel-Trenaunay Syndrome (KTS) is a rare syndrome comprised of capillary, venous and lymphatic malformations associated with bony/soft-tissue overgrowth affecting a limb or limbs [1]. Deep veinous thrombosis (DVT) is a known potential complication of KTS, but pulmonary embolism (PE) is relatively rare [2]. Herein we describe a postpartum patient with KTS who developed PE despite prophylactic measures. CASE PRESENTATION: A 39 year old pregnant woman with history of KTS and associated pelvic varicosities delivered her child by emergent cesarean section following placental abruption due to a retroplacental clot. Her history included prior pulmonary embolism (PE) at age 16 with subsequent placement of an IVC filter. Her antenatal course was uncomplicated and she received a low dose aspirin and 40 mg low molecular weight heparin (LMWH), both daily, throughout her pregnancy. LMWH was resumed 6 hours after delivery. On post-operative day 2 she complained of acute severe shortness of breath, and was tachycardic with an oxygen saturation of 94% and non-tender lower limbs. A chest CT angiogram revealed extensive bilateral PE's. ECG showed a classic S1Q3T3 pattern. Transthoracic echocardiogram showed RV dilatation with reduced systolic function and positive McConnell's sign. Troponin I was elevated at 1.27 ng/mL (normal <= 0.04 ng/mL). LMWH was increased to therapeutic dosing and warfarin therapy was begun. Upper and lower extremity venous duplex studies were completed and negative for DVT. The patient remained hemodynamically stable and transitioned to the outpatient setting. DISCUSSION: KTS and pregnancy are both risk factors for DVT/PE. The coexistence of these two prothrombotic states can cause the PE to break through the prophylaxis. PE appears to be rare in KTS [2,3] and reports of KTS complicated by pregnancy are scarce. It has been suggested that aspirin and heparin use during pregnancy in patients with KTS may reduce thromboembolic disease [1]. This did not bear out in our patient, however, Rebarber et al [1] described a case in which therapeutic LMWH was used during pregnancy without thromboembolic complications. Additionally, our patient's IVC filter was not protective, potentially due to clot bypassing it via the collateral vasculature that develops in the vascular malformations of KTS. CONCLUSIONS: Patients with KTS need lifelong anticoagulation and/or an IVC filter. Those with additional risk factors predisposing them to thromboembolic disease should have these issues duly addressed and should be monitored closely for DVT/PE.

Status
CONFERENCE ABSTRACT

Institution
(Mehta, Bhatia, Kandasamy, DePew) Creighton University, Omaha, NE, United States

Publisher
American College of Chest Physicians
Date Created
20151226
Year of Publication
2015

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Link to the External Link Resolver:
NHS LinkResolver

**8.**
**Obstetrical management of pregnancy in patients with klippel-trenaunay-weber syndrome.**
Basurto-Diaz D., Arroyo-Lemarroy T., Flores-Mendoza H., Hernandez-Nieto C.A., Soto-Fuenzalida G.
Embase
International Journal of Gynecology and Obstetrics. Conference: 21st FIGO World Congress of Gynecology and Obstetrics. Vancouver, BC Canada. Conference Publication: (var.pagings). 131 (pp E459), 2015. Date of Publication: October 2015.
[Conference Abstract]
AN: 72070240
Objectives: To present the obstetrical management of two patients with diagnosis of Klippel-Trenaunay-Weber (KTW) syndrome; as well as their evolution throughout gestation and puerperium. KTW is a rare congenital disorder that consists of vascular-cutaneous nevi, varicose veins, venous malformations, soft and hard-tissue hypertrophy, all of which affects one or more limbs. The morbidity of this disease is associated with the presence of vascular abnormalities. In this subset of patients, pregnancy is not encouraged because it places the woman in a high obstetrical risk and potential complications. There are few cases reported this disease associated with gestation. Method: Consultation and revision of medical records of these two patients with the diagnosis of Klippel-Trenaunay-Weber syndrome (KTW); both of which received close follow up during prenatal care and anticoagulation at prophylactic doses additional to aspirin during gestation and puerperium. Obstetrical management in both cases resulted in delivery by means of cesarean section. Revisions and analysis of both medical records was undertaken with a thorough literature-associated revision. Results: Case 1) 19-year-old woman with KTW syndrome and 38.5- week pregnancy complicated with gestational diabetes mellitus and preeclampsia. C-section was performed (male - 3340 gr, Apgar 8/9). Case 2) 23-year-old primigravid, with KTW, 37-week gestation and type 1 intrauterine growth restriction. Anticoagulation with 100mg daily aspirin and 20 mgs of subcutaneous enoxaparin, both started during the second trimester. A C-section was performed because of a breech presentation (female - 2430 gr, Apgar 8/9 respectively. During surgery of both patients multiple uterine tortuous varicose veins where encountered. These two patients coursed an uneventful puerperium. Conclusions: The expected physiological changes undertaken during gestation can exacerbate the complications of this syndrome. There is doubt of the benefit of anticoagulation therapy either with aspirin or low-molecular-weight heparins in this subset of patients. The most commonly reported complication in these patients is coagulopathy including deep vein thrombosis. There is currently no evidence to establish C-section as a method of delivery. During puerperium distinct recommendations point toward profilactic anticoagulation therapy. As of date, no other cases are reported of KTW syndrome in a pregnant woman with either gestational diabetes mellitus, preeclampsia or intrauterine growth restriction.

Status
CONFERENCE ABSTRACT
Institution
(Basurto-Diaz, Arroyo-Lemarroy, Flores-Mendoza, Hernandez-Nieto, Soto-Fuenzalida) Instituto Tecnologico y De Estudios Superiores De Monterrey, Monterrey, Nuevo Leon, Mexico
Publisher
Elsevier Ireland Ltd
Date Created
20151106
Year of Publication
2015

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Click here for full text options

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NHS LinkResolver

**9.**
**Klippel-trenaunay syndrome: A rare diagnosis with implications for pregnancy.**
Russo M.L., Grundy M.E., Ehsanipoor R.M., Blakemore K.J.
Embase
Reproductive Sciences. Conference: 62nd Annual Scientific Meeting of the Society for Gynecologic Investigation, SGI 2014. San Francisco, CA United States. Conference Publication: (var.pagings). 22 (pp 350A), 2015. Date of Publication: March 2015.
[Conference Abstract]
AN: 71848141
INTRODUCTION: Klippel-Trenaunay syndrome (KTS) is characterized by port-wine stain, venous and/or lymphatic malformation and associated soft tissue/bone hypertrophy. Women with KTS face substantive risk for thromboembolic and hemorrhagic events in pregnancy. Given the rarity of this condition, little is known about the optimal management in pregnancy. Our objective was to investigate the pregnancy complications, management strategies and outcomes in patients with KTS. METHODS: This was an IRB-approved case series of all pregnancies in which there was maternal KTS at our institution, as identified by ICD9 codes. Medical records were reviewed from 1993 to 2014. We recorded history of thrombosis, anticoagulation, location of vascular malformations, relevant imaging studies, details of delivery and neonatal outcomes in currently pregnant and delivered patients. RESULTS: Seven pregnancies (5 patients) were included in this case series. Two patients had extensive vulvovaginal malformations that precluded an attempt at vaginal delivery. There were 2 successful vaginal deliveries and 5 Cesarean sections. Anesthesia used was epidural (N=4) and general endotracheal anesthesia (N=3). Diagnostic imaging influenced management in four cases. Post-operative complications included EBL greater than 1L (N=3), wound hematoma (N=2), pelvic hematoma (N=1) and post-partum pulmonary embolism (N=1). There were no cases of fetal intrauterine growth restriction or neonatal KTS. CONCLUSIONS: Women with Klippel-Trenaunay syndrome can have successful pregnancies with good maternal and neonatal outcomes. Patients often require anticoagulation

during and after pregnancy. A multidisciplinary approach to their care is essential to optimize these outcomes. Antenatal diagnostic imaging with MRI is a useful tool in delivery planning, as these patients are at high risk for bleeding complications.
Status
CONFERENCE ABSTRACT
Institution
(Russo, Grundy, Ehsanipoor, Blakemore) Gynecology and Obstetrics, Johns Hopkins University, School of Medicine, Baltimore, MD, United States
Publisher
SAGE Publications Inc.
Date Created
20150418
Year of Publication
2015

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NHS LinkResolver

**10.**
**Klippel-Trenaunay-Weber syndrome-associated arterial and venous malformations in the lower uterine segment.**
Bouchard-Fortier G; El-Chaar D; Hawrylyshyn P; Kingdom J; Lyons E.
Ovid MEDLINE(R) Epub Ahead of Print, In-Process & Other Non-Indexed Citations, Ovid MEDLINE(R) Daily, Ovid MEDLINE and Versions(R)
Journal of Obstetrics & Gynaecology Canada: JOGC. 36(8):665-6, 2014 Aug.
[Case Reports. Journal Article]
UI: 25222156
Status
MEDLINE
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Lyons, Elliott. Department of Obstetrics and Gynaecology, University of Toronto, Toronto ON.
Date Created

20140916
Year of Publication
2014

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NHS LinkResolver

**11.**
**Klippel-Trenaunay syndrome and gestational trophoblastic neoplasm.**
Sreenivasan P., Kumar S., Kumar K.K.
Embase
Indian pediatrics. 51 (9) (pp 745-746), 2014. Date of Publication: 01 Sep 2014.
[Article]
AN: 603854094
BACKGROUND: Klippel-Trenaunay syndrome is a non-heritable venous malformation with bone and soft tissue hypertrophy and cutaneous nevi. CASE CHARACTERISTICS: Neonate with Klippel Trenaunay syndrome born to a mother with past history of Gestational trophoblastic neoplasm. OBSERVATION: Antenatally, a fetal vascular malformation was identified ultrasonologically at 29 weeks gestation. Acute myeloid leukemia was diagnosed in mother at 33 weeks gestation. MESSAGE: A rare association of Klippel Trenaunay syndrome and gestational trophoblastic neoplasm with the possible role of either hyperglycosylated Human Chorionic Gonadotropin or chemotherapy as a link is highlighted.
PMID
25228614 [http://www.ncbi.nlm.nih.gov/pubmed/?term=25228614]
Institution
(Sreenivasan, Kumar, Kumar) Department of Pediatrics, Government Medical College, Thiruvananthapuram, Kerala. Correspondence to: Dr Priya Sreenivasan, Assistant Professor of Pediatrics, Government Medical College, Thiruvananthapuram, Kerala. priyavineed@yahoo.co.in
Date Created
20150421
Year of Publication
2014

Link to the Ovid Full Text or citation:
Click here for full text options

Link to the External Link Resolver:
NHS LinkResolver

**12.**

**Successful management of pregnancy in an African woman with Klippel Trenaunay syndrome.**

Kemfang Ngowa J.D., Dobgima Pisoh W., Motzebo Mbouopda R., Ngassam A., Fokou M., Kasia J.-M.

Embase
Pan African Medical Journal. 16 (no pagination), 2013. Date of Publication: 2013.
[Article]
AN: 370324780

Klippel Trenaunay syndrome (KTS) is a rare congenital disease characterized by a triad of cutaneous hemangioma, varicose veins and bone or soft tissue hypertrophy. Cases of pregnancy complicated by KTS are rare. There is an increased risk of thrombo-embolic disease and hemorrhage during pregnancy. Both obstetric and anesthetic management of KTS in pregnancy can be rather complicated. We present a successful management of pregnancy in an African woman with KTS at Yaounde General Hospital, Cameroon. © Jean Dupont Kemfang Ngowa et al.

PMID
24772228 [http://www.ncbi.nlm.nih.gov/pubmed/?term=24772228]
Status
EMBASE

Institution
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(Motzebo Mbouopda) Anesthesiology Unit, Yaounde General Hospital, P.O. Box 5408, Yaounde, Cameroon
(Fokou) Surgery Unit, Yaounde General Hospital, P.O. Box 5408, Yaounde, Cameroon

Publisher
African Field Epidemiology Network (PO Box 12874, Kampala, Uganda)
Date Created
20131216
Year of Publication
2013

**13.**

**Klippel Trenaunay syndrome and pregnancy associated with post partial thyroidectomy and gestational diabetes mellitus: A case report.**

De S., Nagandla K., Kanagasabai S., Woh T.S.

Embase
Internet Journal of Gynecology and Obstetrics. 17 (3) (no pagination), 2013. Date of Publication: 2013.
[Article]
AN: 370251607

Pregnancy associated with Klippel Trenaunay syndrome is rare. Few cases have been reported in the medical literature. In this case report, we describe a patient who had partial thyroidectomy seven years prior to the pregnancy associated with gestational diabetes. She was monitored in the second half of the pregnancy by two weekly blood sugar profile, thyroid function test and serial ultrasound. The soft tissue hypertrophy restricted the movement of her entire left lower limb which made flexion of the hip and knee joint severely painful. Hence she was planned for an elective caesarean. Unfortunately she came with preterm prelabor rupture of membranes at 36 weeks of gestation and had an emergency caesarean. She had an uneventful intra and post operative period and was discharged on the fifth post-op day. This case report illustrates that women with Klippel Trenaunay syndrome with systematic management approach are likely to have an uneventful pregnancy. 

Status
EMBASE

Institution
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Publisher
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**14.**

**Klippel-trenaunay syndrome and a successful pregnancy: A case report.**

Bentham L., Scarr C., Al-Sayegh H., Singh J.

Embase
BJOG: An International Journal of Obstetrics and Gynaecology. Conference: RCOG World Congress 2013. Liverpool United Kingdom. Conference Publication: (var.pagings). 120 (pp 29-30), 2013. Date of Publication: June 2013.
[Conference Abstract]
AN: 71135671

Aim To present a rare case of a successful pregnancy and delivery of a 21-year-old woman with a longstanding diagnosis of Klippel- Trenaunay Webber Syndrome. Methods Review of case notes of the patient who was managed in a large district hospital. Background Klippel-Trenaunay Webber Syndrome (KTS) is a rare congenital condition, first described in 1900 by French physicians Maurice Klippel and Paul Trenaunay. The aetiology is, as yet, unknown, and displays sporadic incidence. KTS is characterised by a clinical triad of cutaneous capillary malformations - commonly a capillary haemangioma ('port-wine stain'), bone and soft tissue hypertrophy - commonly affecting one limb and varicose veins or venous malformations. Although there is a wide clinical variability common complications include chronic venous stasis (and associated risk of venous thromboembolism), visceral bleeding and orthopaedic complications such as gait disturbances and scoliosis. Discussion This 21-year-old primiparous women was successfully managed during her pregnancy under consultant-led care with multi-disciplinary input - namely anaesthetic opinion regarding analgesia options in labour and haematological opinion due to the high thromboembolic risk. KTS manifested in this patient as scoliosis, left sided hemihypertrophy and mild learning disability. She was delivered by emergency caesarean section for failure to progress, giving birth to a healthy male infant. She suffered 1000 mL estimated blood loss intraoperatively due to multiple prominent vessels on the lower segment of her uterus and slight uterine atony. She had an uneventful postoperative recovery and was discharge with 6 weeks of prophylactic low molecular weight heparin due to her high risk of venous thromboembolism. Conclusion Pregnant patients with complex medical conditions such as KTS require integrated multi-professional involvement to ensure high-quality patient-centred care. Early involvement of appropriate specialities allowed the composition of a robust birth plan for this patient, facilitating this successful outcome.

Status
CONFERENCE ABSTRACT

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20130816
Year of Publication
2013

NHS LinkResolver

**15.**

**Management of a Klippel-Trenaunay syndrome in pregnant women with mega-cisterna magna and splenic and vulvar varices at birth: a case report.**

Atis A; Ozdemir G; Tuncer G; Cetincelik U; Goker N; Ozsoy S.

Ovid MEDLINE(R) Epub Ahead of Print, In-Process & Other Non-Indexed Citations, Ovid MEDLINE(R) Daily, Ovid MEDLINE and Versions(R)
Journal of Obstetrics & Gynaecology Research. 38(11):1331-4, 2012 Nov.
[Case Reports. Journal Article]
UI: 22612590

Klippel-Trenaunay syndrome (KTS) is a rare, sporadic and complex malformation characterized by the clinical triad of: (i) capillary malformation (port-wine stain); (ii) soft tissue and bone hypertrophy or occasionally, hypertrophy of one lower limb; and (iii) atypical lateral varicosity. The maternal and fetal risks associated with pregnancy in women with KTS are proportional to disease severity, which can be exacerbated by pregnancy. Complications include bleeding, disseminated intravascular coagulation, thromboembolic events and pain. Here, we report the case of a pregnant woman with KTS who had an uneventful pregnancy, labor and postpartum course, but had splenic and large vulvar vein varices. The obstetrical course of women with KTS varies. Management is largely conservative and multidisciplinary approaches form the mainstay for managing these patients based on their symptoms.



Status
MEDLINE

Authors Full Name
Atis, Alev; Ozdemir, Gunseli; Tuncer, Gulden; Cetincelik, Umran; Goker, Nimet; Ozsoy, Sibel.
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Atis, Alev. Obstetrics and Gynecology Clinic Department of Genetics, Sisli Etfal Training and Research Hospital, Istanbul, Turkey. alevatis@mynet.com

Date Created
20121030
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2012

**16.**

**Paraplegia in pregnancy: a case of spinal vascular malformation with Klippel-Trenaunay syndrome.**

Demir CF; Yildiz M; Ozdemir H; Kapan O; Bozgeyik Z; Berilgen S; Aygun B; Erol FS.

Ovid MEDLINE(R) Epub Ahead of Print, In-Process & Other Non-Indexed Citations, Ovid MEDLINE(R) Daily, Ovid MEDLINE and Versions(R)

Spine. 37(19):E1218-20, 2012 Sep 01.

[Case Reports. Journal Article]

UI: 22576072

STUDY DESIGN: A case report.

OBJECTIVE: To present improvement of paraplegia due to spinal vascular malformation after delivery with no intervention.

SUMMARY OF BACKGROUND DATA: Pregnancy has been reported rarely in patients with Klippel-Trenaunay syndrome (KTS). A combined case of spinal arteriovenous malformations (AVMs) within KTS has not been reported before.

METHODS: A case report of KTS with paraplegia is presented and the pertinent literature is then reviewed.

RESULTS: A pregnant woman who was diagnosed with KTS in childhood presented with newly developed paraplegia due to spinal AVMs. Magnetic resonance image showed a vascular malformation at the T9-T12 levels without evidence of spinal cord hemorrhage. Diagnostic angiography that was performed after delivery revealed a high-flow arteriovenous fistula with AVM.

CONCLUSION: Pregnancy complicated by paraplegia is thought to be secondary to venous engorgement and to the resulting spinal cord ischemia in this case. Her neurological symptoms and signs gradually improved over the few days after cesarean delivery.

Status

MEDLINE

Authors Full Name

Demir, Caner Feyzi; Yildiz, Mustafa; Ozdemir, Hasan; Kapan, Oktay; Bozgeyik, Zulkif; Berilgen, Said; Aygun, Banu; Erol, Fatih Serhat.

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Date Created

20120830

Year of Publication

2012

**17.**
**Klippel-Trenaunay syndrome complicating pregnancy.**
Gonzalez-Mesa E., Blasco M., Anderica J., Herrera J.
Embase
BMJ Case Reports. (no pagination), 2012. Date of Publication: 2012.
[Article]
AN: 365527842
The Klippel-Trenaunay syndrome is a rare congenital disorder that affects one or more limbs. It is characterised by cutaneous vascular nevi, venous malformations and hypertrophy of soft tissues and bone. There are very few cases reported in pregnant women, so the level of uncertainty is high when it appears during gestation. It is a disease that increases obstetric risk and can exacerbate complications, mainly thromboembolic and haemorrhagic. We report below the case of a pregnant woman diagnosed with this syndrome and the multidisciplinary management held in our centre. 
PMID
22854239 [http://www.ncbi.nlm.nih.gov/pubmed/?term=22854239]
Status
EMBASE
Institution
(Gonzalez-Mesa, Blasco, Anderica, Herrera) Department of Obstetrics and Gynecology, Carlos Haya University Hospital, Malag, Spain
Publisher
BMJ Publishing Group (Tavistock Square, London WC1H 9JR, United Kingdom)
Date Created
20120910
Year of Publication
2012

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Click here for full text options

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**18.**

**Klippel-Trenaunay syndrome, pregnancy and the liver: An unusual interplay.**

Samonakis D.N., Oustamanolakis P., Manousou P., Kouroumalis E.A., Burroughs A.K.

Embase
Annals of Gastroenterology. 25 (4) (pp 365-367), 2012. Date of Publication: 2012.
[Article]
AN: 366040914

Klippel-Trenaunay syndrome is a rare congenital syndrome characterized by capillary malformations, soft tissue and bone hypertrophy, and varicose veins. There is a well-established risk for thrombotic complications in these patients. A case of a young patient diagnosed post partum with the very rare liver involvement is presented. The complex clinical course, the multidisciplinary management and the long-term outcome are discussed. © 2012 Hellenic Society of Gastroenterology.

Status
EMBASE

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(Samonakis, Manousou, Burroughs) The Royal Free Sheila Sherlock Liver Centre, University Department of Surgery, Royal Free Hospital Hampstead, London, United Kingdom (Samonakis, Oustamanolakis, Kouroumalis) Department of Gastroenterology and Hepatology, University Hospital of Heraklion, Crete, Greece

Publisher
Hellenic Society of Gastroenterology (67, Democratias Ave., Psychico 15451 N, Greece)
Date Created
20121122
Year of Publication
2012

**19.**

**Klippel-Trenaunay-Syndrome and pregnancy: Is there a relationship?.**

Scholz U., Oppermann J., Siegemund A., Schobess R.

Embase
Hamostaseologie. Conference: 56. Jahrestagung der Gesellschaft fur Thrombose- und Hamostase - Forschung e. V., GTH. St. Gallen Switzerland. Conference Publication: (var.pagings). 32 (1) (pp A63), 2012. Date of Publication: 2012.
[Conference Abstract]
AN: 70867193

The Klippel-Trenaunay-syndrome (KTS) is a rare congenital anomaly with various expression and unknown etiology. It is characterized by capillary and venous malformations and hypertrophy of bone and soft tissue. During pregnancy the hemostatic balance is physiological impared and the complications of patients with KTS include bleeding, disseminated intravascular coagulation, thrombembolic events and pain. Patients and methods:The obstetrical course of three patients were analysed. Documentation included thrombophilic markers, course of pregnancy, complication and outcome of the patients and the newborns. Results: The clinical symptoms and the maternal and fetal risks were associated with the severity of disease. Patient 1 with a second thrombophilic marker (Prothrombinmutation, heterozygot, G20210A) showed a massive activation of the coagulation system during the first trimester. Low molecular heparin was used in prophylactic dose with an excellent effect on the impaired coagulation system and the outcome. In patient 2 the coagulation system showed no abnormalities but in an antenatal ultrasound a massive vascular malformation was detected. The planned sectio caesarea (reason: position of the child) were performed without complicationPatient 3 also showed vascular malformations of the uterus wall. The spontan delivery was without complications. All three patents had a thrombosis prophylaxis during the 4 weeks after delivery. Conclusion: The maternal and fetal risks associated with pregnancy in women with KTS are a multifactorial process. Beside the changes of the hemostasis the expansion of the vascular malformation should be investigated to avoid complications.

Status
CONFERENCE ABSTRACT

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Publisher
Schattauer GmbH
Date Created
20120915
Year of Publication
2012

**20.**

**Successful Management of Pregnancy Complicated by Klippel-Trenaunay Syndrome Using MR Angiography-Based Evaluation.**

Tanaka R; Fujita Y; Ishibashi Hiasa K; Yumoto Y; Hidaka N; Fukushima K; Wake N.

Ovid MEDLINE(R) Epub Ahead of Print, In-Process & Other Non-Indexed Citations, Ovid MEDLINE(R) Daily, Ovid MEDLINE and Versions(R)
Case Reports in Obstetrics and Gynecology. 2011:723467, 2011.
[Journal Article]
UI: 22567516

Klippel-Trenaunay syndrome (KTS) is a rare congenital disease, and extensive cutaneous hemangiomas and abnormal venous vessels are characteristic. In our case, to manage her pregnancy with KTS, whole-body MRA was performed before delivery. A 29-year-old woman was referred at 28 weeks because of prominent vulvovaginal varicosities due to KTS. At 35 weeks, hypertrophy and multiple venous varicosities of her leg as well as massive vulvovaginal varicosities became prominent with a normal coagulation profile. Systematic MRAs revealed hemangiomas and varicosities in the right leg, the lower abdomen, and the pubic region, while no obvious AVM was detected around the bronchial tube and spine. We decided to deliver her baby by cesarean section at 37 weeks under general anesthesia, and a healthy baby was delivered. No blood transfusion was required. Prophylaxis against thrombosis was performed after the operation. She was discharged with her baby. Her vulvovaginal varicosities shrunk considerably one month later.

Status
PubMed-not-MEDLINE

Authors Full Name
Tanaka, Reiko; Fujita, Yasuyuki; Ishibashi Hiasa, Kana; Yumoto, Yasuo; Hidaka, Nobuhiro; Fukushima, Kotaro; Wake, Norio.
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PMID
https://www.ncbi.nlm.nih.gov/pmc/articles/PMC3335629
Date Created
20120508
Year of Publication
2011

**21.**

**Assisted reproduction in a patient with Klippel-Trenaunay syndrome: Management of thrombophilia and consumptive coagulopathy.**

Martin J.R., Pels S.G., Paidas M., Seli E.

Embase
Journal of Assisted Reproduction and Genetics. 28 (3) (pp 217-219), 2011. Date of Publication: March 2011.
[Article]
AN: 51208719

Klippel-Trenaunay Syndrome (KTS) is a rare, sporadic triad of congenital malformations involving an extensive port wine stain, soft tissue or bone hypertrophy and underlying venous and/or lymphatic malformation involving an extremity. Pregnancy is known to exacerbate KTS complications and can put women at increased obstetrical risk due to deep venous thrombosis and other thromboembolic events. Here we report a case of a patient with KTS who achieved a pregnancy through in vitro fertilization (IVF) using her own eggs and a gestational surrogate in the setting of hypercoagulability and chronic consumptive coagulopathy. © 2010 Springer Science+Business Media, LLC.

PMID
21188495 [http://www.ncbi.nlm.nih.gov/pubmed/?term=21188495]
Status
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(Martin, Pels, Paidas, Seli) Department of Obstetrics, Gynecology and Reproductive Sciences, Yale University, School of Medicine, 333 Cedar Street, New Haven, CT 06520-8063, United States
Publisher
Springer New York (233 Spring Street, New York NY 10013-1578, United States)
Date Created
20110622
Year of Publication
2011

**22.**
**Klippel-trenaunay syndrome and pregnancy.**
Gungor Gundogan T; Jacquemyn Y.
Ovid MEDLINE(R) Epub Ahead of Print, In-Process & Other Non-Indexed Citations, Ovid MEDLINE(R) Daily, Ovid MEDLINE and Versions(R)
Obstetrics & Gynecology International. 2010:706850, 2010.
[Journal Article]
UI: 21209709
Klippel-Trenaunay syndrome is a rare congenital vascular disorder, and only few cases have been described in pregnancy. We describe two cases, in one patient without complications, the other patient developed postpartum deep venous thrombosis.
Status
PubMed-not-MEDLINE
Authors Full Name
Gungor Gundogan, Tugba; Jacquemyn, Y.
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Gungor Gundogan, Tugba. Department of Obstetrics and Gynaecology, Antwerp University Hospital UZA, Wilrijkstraat 10, 2650 Edegem, Belgium.
PMID
https://www.ncbi.nlm.nih.gov/pmc/articles/PMC3010666
Date Created
20110106
Year of Publication
2010

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**23.**

**Peripartum cardiomyopathy and Klippel-Trenaunay syndrome.**

Carta G., De Lellis V., Di Nicola M., Kaliakoudas D.

Embase
Clinical and Experimental Obstetrics and Gynecology. 37 (2) (pp 155-157), 2010. Date of Publication: 2010.
[Article]
AN: 359074337

Klippel-Trenaunay syndrome (KTS) is a rare congenital disorder of unknown etiology characterized by venous malformations or varicose veins, cutaneous capillary malformation and hypertrophy of soft tissues with limb (usually asymmetric lower extremity) involvement. Peripartum cardiomyopathy (PPCM) is characterized by rapid onset heart failure during the final month of pregnancy or within five months of delivery, in the absence of identifiable risk factors or previous heart disease. The aim of this study was to illustrate the correlation between the KTS and the onset of PPCM in women with twin pregnancies. Our case is a 35-year-old woman, gravida II para I, with KTS, twin pregnancy and PPCM. We can assume that, as the heart of a women with KTS usually works with a low preload reserve due to the widespread venous varicosities, if a significant increase in preload occurs, it may lead to the onset of cardiac dilatation and thus PPCM.

PMID
21077513 [http://www.ncbi.nlm.nih.gov/pubmed/?term=21077513]
Status
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(Carta, De Lellis, Di Nicola, Kaliakoudas) Department of Obstetrics and Gynecology, University of L'Aquila, Italy

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Date Created
20100709
Year of Publication
2010

**24.**

**Cervical prolapse during pregnancy and Klippel-Trenaunay syndrome.**

Minguez J.A., Auba M., Olartecoechea B.

Embase

International Journal of Gynecology and Obstetrics. 107 (2) (pp 158), 2009. Date of Publication: November 2009.

[Article]

AN: 50566712

PMID

19576584 [http://www.ncbi.nlm.nih.gov/pubmed/?term=19576584]

Status

EMBASE

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Publisher

Elsevier Ireland Ltd (P.O. Box 85, Limerick, Ireland)

Date Created

20091211

Year of Publication

2009



**25.**

**A case of recurrent vaginal bleeding in pregnancy: The antenatal management of a patient with Klippel-Trenaunay syndrome.**

Tirlapur S., Abughazza O., Downes E.

Embase
International Journal of Gynecology and Obstetrics. Conference: 19th FIGO World Congress of Gynecology and Obstetrics. Cape Town South Africa. Conference Publication: (var.pagings). 107 (pp S427), 2009. Date of Publication: October 2009.
[Conference Abstract]
AN: 70230519

Presentation: A 39 year-old Caucasian woman presented to casualty at 14 weeks of gestation with a sudden onset of unprovoked, profuse vaginal bleeding. Anomaly ultrasound scans revealed no fetal abnormalities, although a hypertrophied, vascular uterus was noted, with no signs of fibroids. After a third episode of heavy vaginal bleeding, a male infant was born in good condition, by emergency Caesarian section for antepartum haemorrhage and preterm labour at 29 weeks gestation. This was her sixth pregnancy. She suffered two first trimester miscarriages. Her first pregnancy was uneventful with an instrumental delivery at term. Throughout her second pregnancy she suffered vaginal bleeding and delivered at 35 weeks by Caesarian section for breech presentation. In her third pregnancy she experienced mild antepartum bleeding throughout and delivered at 32 weeks by emergency Caesarian section for preterm labour. Discussion: Klippel-Trenaunay Syndrome is a rare congenital vascular disease causing cutaneous capillary and venous malformations and soft tissue hypertrophy. Identified complications include cellulitis, bleeding, thromboembolic events and secondary infection. To minimise complications, patients can be treated antenatally with anticoagulant treatment and Caesarian sections may be performed if there are highly vascularised changes to the cervix and lower uterine segment. Klippel-Trenaunay is an unusual cause of antepartum haemorrhage and may lead to an expedited delivery. Between 2003 and 2005 36% of all maternal deaths in the United Kingdom due to haemorrhage were attributed to antepartum haemorrhage. Sub-standard care is a huge issue. Early recognition and management is vital to save mothers' lives.

Status
CONFERENCE ABSTRACT

Institution
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Elsevier Ireland Ltd
Date Created
20100809
Year of Publication
2009

**26.**

**Anesthetic and obstetric considerations in a parturient with Klippel-Trenaunay syndrome.**

Sivaprakasam MJ; Dolak JA.

Ovid MEDLINE(R) Epub Ahead of Print, In-Process & Other Non-Indexed Citations, Ovid MEDLINE(R) Daily, Ovid MEDLINE and Versions(R)

Canadian Journal of Anaesthesia. 53(5):487-91, 2006 May.

[Case Reports. Journal Article]

UI: 16636034

PURPOSE: To explain the considerations governing the anesthetic management of pregnant patients with Klippel-Trenaunay syndrome (KTS). Klippel-Trenaunay syndrome is a congenital vascular disease characterized by cutaneous hemangiomas, venous varicosities, and limb hypertrophy; and is associated with both hemorrhagic and thrombotic complications. The importance of this diagnosis, including the presence of neuraxial vascular anomalies, is often under-appreciated by both obstetric and anesthesia providers. While regional anesthetic management of patients with KTS has been discussed by others, we present a case in which regional anesthesia presented an unwarranted risk to the patient.

CLINICAL FEATURES: An obese, 18-yr-old parturient with a fetus in the breech position underwent Cesarean delivery at 35 weeks gestation secondary to evolving preeclampsia. Unfortunately, no neurovascular imaging of this patient's spine was available. The patient underwent an attempted external cephalic version, a failed obstetric induction, and, ultimately, a Cesarean delivery under general anesthesia. The resulting infant was without any stigmata of KTS. Both mother and infant did well during the course of their hospitalization, and were discharged home without incident.

CONCLUSIONS: The posterior cutaneous hemangiomas of KTS may be associated with underlying epidural and subdural vascular malformations. Disruption of these vascular anomalies during regional anesthesia may lead to neuraxial hematoma formation, which may be further compounded by a consumptive coagulopathy observed in some cases of KTS. If neuraxial vascular anomalies cannot be ruled out radiographically, regional anesthesia should be avoided. Additionally, regardless of the anesthetic technique chosen, the coagulation profile of these patients should be verified for signs of coagulopathy.

Status

MEDLINE

Authors Full Name

Sivaprakasam, Michael J; Dolak, James A.

Institution

Sivaprakasam, Michael J. Department of Anesthesiology, St. Edward's Mercy Medical Center, Fort Smith, AR, USA.

Date Created

20060425

Year of Publication

2006

**27.**
**Klippel-Trenaunay-type syndrome in pregnancy. [Review] [46 refs]**
Stein SR; Perlow JH; Sawai SK.
Ovid MEDLINE(R) Epub Ahead of Print, In-Process & Other Non-Indexed Citations, Ovid MEDLINE(R) Daily, Ovid MEDLINE and Versions(R)
Obstetrical & Gynecological Survey. 61(3):194-206, 2006 Mar.
[Case Reports. Journal Article. Review]
UI: 16490119
INTRODUCTION: Klippel-Trenaunay-Type Syndrome (KTTS) is a rare congenital anomaly with variable expression and an unknown etiology characterized by capillary and venous malformations and hypertrophy of bone and soft tissue. Pregnancy has been rarely reported in patients with KTTS and since 1989 there have been only 13 case reports of pregnancy in women with KTTS reported in the literature. Concurrent pregnancy is associated with adverse perinatal outcomes. To the best of our knowledge this is the second reported, and largest, series of cases.
STUDY DESIGN: After a thorough review of the literature, the medical records of four obstetrical patients with KTTS were reviewed.
RESULTS: The obstetrical course of women with KTTS varies. Complications include bleeding, DIC, thromboembolic events, and pain.
CONCLUSIONS: The maternal and fetal risks associated with pregnancy in women with KTTS are proportional to the severity of disease, which can be exacerbated by pregnancy. Thoughtful preconceptional counseling, along with methodical and systematic intrapartum and postpartum care are keys to reducing mortality and morbidity. [References: 46]
Status
MEDLINE
Authors Full Name
Stein, Susan R; Perlow, Jordan H; Sawai, Shirley K.
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Stein, Susan R. Department of Obstetrics & Gynecology, Banner Good Samaritan Medical Center, Phoenix, Arizona 85006, USA. SRSTEIN@aol.com
Date Created
20060221
Year of Publication
2006

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**28.**

**Obstetric management of Klippel-Trenaunay syndrome.**

Rebarber A; Roman AS; Roshan D; Blei F.

Ovid MEDLINE(R) Epub Ahead of Print, In-Process & Other Non-Indexed Citations, Ovid MEDLINE(R) Daily, Ovid MEDLINE and Versions(R)

Obstetrics & Gynecology. 104(5 Pt 2):1205-8, 2004 Nov.

[Journal Article]

UI: 15516454

BACKGROUND: Klippel-Trenaunay syndrome is a rare congenital disease characterized by extensive cutaneous vascular malformations, venous varicosities, focal abnormalities of the deep venous system, and underlying soft tissue or bony hypertrophy. Given the rarity of the disease, there is little information available to counsel patients with Klippel-Trenaunay syndrome regarding obstetric outcome.

CASES: We report our experience with 3 patients in whom Klippel-Trenaunay syndrome complicated 4 pregnancies. Successful delivery of a healthy infant at or beyond 36 weeks of gestation was achieved in all pregnancies. One of the 4 pregnancies was complicated by pulmonary embolism.

CONCLUSION: Klippel-Trenaunay syndrome was once thought to be a contraindication to pregnancy. With careful management, successful pregnancies can be achieved.

Status

MEDLINE

Authors Full Name

Rebarber, Andrei; Roman, Ashley S; Roshan, Daniel; Blei, Francine.

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Date Created

20041101

Year of Publication

2004

**29.**
**Klippel-Trenaunay syndrome and pregnancy.**
Hergesell K., Kroger K., Petruschkat S., Santosa F., Herborn C., Rudofsky G.
Embase
International Angiology. 22 (2) (pp 194-198), 2003. Date of Publication: June 2003.
[Article]
AN: 36869391
General recommendations on how to deal with pregnancy in patients with Klippel-Trenaunay syndrome (KTS) are rare. We describe the case of a 32-year-old female with KTS, involving the head and the left arm and leg, delivering a healthy female child, and are reviewing the recent literature. The risk to deliver an ill child is low in women with KTS. At the end of the 1st trimester a sonographic investigation can exclude angiodysplastic alterations of the fetus. If the fetus shows changes compatible with KTS, a termination can be discussed because the risk of fatal complications after delivery is high. During pregnancy the careful monitoring of coagulopathic disorders is necessary. Prior to delivery an MR-scan may be useful to detect angiodysplastic vascular structures next to the spinal cord, pelvic structures or the lower abdominal wall, which might complicate peridural anesthesia or caesarean section.
PMID
12865887 [http://www.ncbi.nlm.nih.gov/pubmed/?term=12865887]
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Publisher
Edizioni Minerva Medica S.p.A. (Corso Bramante 83-85, Torino 10126, Italy)
Date Created
20030729
Year of Publication
2003

**30.**
**The Klippel-Trenaunay syndrome in pregnancy.**
Watermeyer S.R., Davies N., Goodwin R.
Embase
BJOG: An International Journal of Obstetrics and Gynaecology. 109 (11) (pp 1301-1302), 2002.
Date of Publication: 01 Nov 2002.
[Article]
AN: 36164760
PMID
12452471 [http://www.ncbi.nlm.nih.gov/pubmed/?term=12452471]
Status
EMBASE
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(Watermeyer, Davies) Dept. of Obstetrics and Gynaecology, University Hospital of Wales, Cardiff, United Kingdom (Goodwin) Department of Dermatology, University Hospital of Wales, Cardiff, United Kingdom
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Publisher
Blackwell Publishing Ltd (9600 Garsington Road, Oxford OX4 2XG, United Kingdom)
Date Created
20030213
Year of Publication
2002

**31.**

**Epidural analgesia in an obstetric patient with Klippel-Trenaunay syndrome.**

Dobbs P; Caunt A; Alderson TJ.
Ovid MEDLINE(R) Epub Ahead of Print, In-Process & Other Non-Indexed Citations, Ovid MEDLINE(R) Daily, Ovid MEDLINE and Versions(R)
British Journal of Anaesthesia. 82(1):144-6, 1999 Jan.
[Case Reports. Journal Article]
UI: 10325854

We describe the use of epidural analgesia for vaginal delivery of a parturient with Klippel-Trenaunay syndrome in whom the use of repeated magnetic resonance imaging during her obstetric care allowed us to see deep haemangiomata. This also allowed the safe sitting of an epidural catheter at L1-2 to provide analgesia for labour and delivery. Klippel-Trenaunay syndrome and the anaesthetic implications of the congenital vascular abnormalities and potential coagulopathy are discussed.

Status
MEDLINE

Authors Full Name
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Dobbs, P. Department of Anaesthetics, Royal Hallamshire Hospital, Sheffield, UK.

Date Created
19990520
Year of Publication
1999

**32.**
**Klippel-Trenaunay-Weber syndrome in pregnancy and at delivery.**
Andreasen K.R., Tabor A., Weber T.
Embase
Journal of Obstetrics and Gynaecology. 19 (1) (pp 78-79), 1999. Date of Publication: 1999.
[Article]
AN: 29074259
Status
EMBASE
Institution
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Publisher
Informa Healthcare (69-77 Paul Street, London EC2A 4LQ, United Kingdom)
Date Created
19990218
Year of Publication
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**33.**
**Sonographic demonstration of widespread uterine angiomatosis in a pregnant patient with Klippel-Trenaunay-Weber syndrome.**
Richards D.S., Cruz A.C.
Embase
Journal of Ultrasound in Medicine. 16 (9) (pp 631-633), 1997. Date of Publication: September 1997.
[Article]
AN: 27494586
Klippel-Trenaunay-Weber syndrome is a rare genetic condition manifesting localized hemangiomas and venous varicosities. A variety of complications have occurred in the few pregnancies that have been reported in women affected by this condition. In one previous case report prenatal ultrasonography demonstrated abnormal uterine vascularity in a patient with Klippel-Trenaunay-Weber syndrome, and in that case the abnormal vessels were restricted to the lateral lower uterine segment. We report a case in which widespread uterine angiomatosis was present in a pregnant patient with Klippel-Trenaunay-Weber syndrome.
PMID
9321785 [http://www.ncbi.nlm.nih.gov/pubmed/?term=9321785]
Status
Embase

Institution
(Richards, Cruz) Dept. of Obstetrics and Gynecology, University of Florida, College of Medicine, Gainesville, FL, United States (Richards) University of Florida, College of Medicine, P.O. Box 100294, Gainesville, FL 32610-0294, United States
Publisher
John Wiley and Sons Ltd (Laurel)
Date Created
20051125
Year of Publication
1997

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**34.**
**Klippel-Trenaunay-Weber syndrome associated with fetal growth restriction.**
Fait G., Daniel Y., Kupferminc M.J., Gull I., Peyser M.R., Lessing J.B.
Embase
Human Reproduction. 11 (11) (pp 2544-2545), 1996. Date of Publication: 1996.
[Article]
AN: 27007474
Klippel-Trenaunay-Weber syndrome is a rare congenital deep-vein malformation. Pregnancy in patients with this syndrome is rare and only a few cases have been reported. Known obstetrical risks in pregnant patients with this syndrome include bleeding from angiomata in the genitalia, and coagulation disturbances. We present a 31 year old woman with this syndrome who, on two occasions, delivered small-for-gestational-age neonates. This may have been due to placental insufficiency caused by angiomatosis related to the syndrome.
PMID
8981152 [http://www.ncbi.nlm.nih.gov/pubmed/?term=8981152]
Status
EMBASE
Institution
(Fait, Daniel, Kupferminc, Gull, Peyser, Lessing) Department Obstetrics Gynecology 'A', Serlin Maternity Hospital, Tel Aviv Sourasky Medical Center, PO Box 7079, Tel Aviv 61070, Israel
Publisher
Oxford University Press (Great Clarendon Street, Oxford OX2 6DP, United Kingdom)
Date Created
19970113
Year of Publication
1996

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NHS LinkResolver

**35.**
**Kasabach-Merritt coagulopathy complicating Klippel-Trenaunay-Weber syndrome in pregnancy.**
**Neubert AG; Golden MA; Rose NC.**
Ovid MEDLINE(R) Epub Ahead of Print, In-Process & Other Non-Indexed Citations, Ovid MEDLINE(R) Daily, Ovid MEDLINE and Versions(R)
Obstetrics & Gynecology. 85(5 Pt 2):831-3, 1995 May.
[Case Reports. Journal Article]
UI: 7724128
BACKGROUND: Klippel-Trenaunay-Weber syndrome is a sporadic genetic syndrome characterized by localized hemangiomas, venous varicosities, and asymmetric osseous hypertrophy of the ipsilateral extremities. Most commonly seen in association with hemangiomas, Kasabach-Merritt syndrome is defined by the presence of thrombocytopenia and a consumptive coagulopathy.
CASE: A 22-year-old primigravida with a prior diagnosis of Klippel-Trenaunay-Weber syndrome presented for genetic counseling and delivery management at 37 weeks' gestation. Large varicosities of the vulva required cesarean delivery. Multiple hemangiomas in the right lower quadrant of the abdomen necessitated the use of a left paramedian cutaneous incision. The patient subsequently developed Kasabach-Merritt syndrome and required the transfusion of blood products as well as heparin and aminocaproic acid therapy for her postoperative management.
CONCLUSION: Klippel-Trenaunay-Weber syndrome in pregnancy is rare. The potential for a refractory coagulopathy presenting as Kasabach-Merritt syndrome should be considered in any patient who presents with extensive hemangiomas.
Status
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Authors Full Name
Neubert, A G; Golden, M A; Rose, N C.
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Neubert, A G. Department of Obstetrics and Gynecology, University of Pennsylvania Medical Center, Philadelphia, USA.
Date Created
19950522
Year of Publication
1995

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**36.**
**Pregnancy complicated by the Klippel-Trenaunay syndrome: A case report.**
Pollack R.N., Quance D.R., Shatz R.M.
Embase
Journal of Reproductive Medicine for the Obstetrician and Gynecologist. 40 (3) (pp 240-242), 1995. Date of Publication: 1995.
[Article]
AN: 25093008
A primigravida with the Klippel-Trenaunay syndrome was admitted to the hospital at 34 weeks' gestation with a complaint of right calf pain. Superficial thrombophlebiris was diagnosed, and she was treated with compresses and analgesia. Speculum examination failed to reveal the presence of lower genitourinary tract arteriovenous malformations. Color flow mapping of the uterus did not identify any arteriovenous malformations. The patient delivered vaginally at term, and the postpartum course was unremarkable. Pregnancies complicated by the Klippel-Trenaunay syndrome are at increased risk of adverse perinatal outcomes, related primarily to the increased risk of hemorrhagic diathesis. The mode of delivery should be considered carefully in all attempt to minimize the risk to both mother and fetus.
PMID
7776315 [http://www.ncbi.nlm.nih.gov/pubmed/?term=7776315]
Status
EMBASE
Institution
(Pollack, Quance, Shatz) Division of Maternal-Fetal Medicine, Department of Obstetrics/Gynecology, Bikur Cholim Hospital, P.O.B. 492, Jerusalem, Israel
Publisher
Donna Kessel (8342 Olive Boulevard, St. Louis, Missouri 63132-2814, United States)
Date Created
19950330
Year of Publication
1995

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**37.**

**Pregnancy in a patient with the Klippel-Trenaunay-Weber syndrome: A case report.**

Verheijen R.H.M., Van Rijen-De Rooij H.J.M., Van Zundert A.A.J., De Jong P.A.

Embase
European Journal of Obstetrics Gynecology and Reproductive Biology. 33 (1) (pp 89-94), 1989.
Date of Publication: 1989.
[Article]
AN: 19248642

A rare case of pregnancy in a patient with the syndrome of Klippel-Trenaunay-Weber is described. The arterio-venous anomalies in this patient originally occurred in her right leg, but in the course of her first pregnnacy she also developed circumscript angiomatosis at the left and right side of the uterus. Her pregnancy was uneventful. However, because of prominent vascular changes in the cervix and lower uterine segment, there appeared to be a cephalo-pelvic discongruency. For this reason a Cesarean section was performed at term. Postoperatively, during heparinization, she had signs of abdominal bleeding, which could be controlled conservatively. There were no signs of intravascular coagulation or cardiac decompensation. A review of the scarce literature on diffuse uterine angiomatosis and angiomatosis occurring as a result of the syndrome of Klippel-Trenaunay-Weber is given. The clinical course and the risks of a pregnancy with this condition are discussed. It is concluded that the angiomatosis based on the syndrome of Klippel-Trenaunay-Weber is less hazardous than a diffuse angiomatosis of the uterus without this disease, the main risk being diffuse intravascular coagulation at or after delivery.

PMID
2553510 [http://www.ncbi.nlm.nih.gov/pubmed/?term=2553510]
Status
EMBASE

Institution
(Verheijen, Van Rijen-De Rooij, Van Zundert, De Jong) Department of Obstetrics and Gynecology, Catharina Hospital, Eindhoven Netherlands

Publisher
Elsevier Ireland Ltd (P.O. Box 85, Limerick, Ireland)
Date Created
19891103
Year of Publication
1989

**38.**
**Klippel-Trenaunay syndrome with complications during pregnancy.**
Fishman A., Paldi E.
Embase
Harefuah. 116 (3) (pp 147-148), 1989. Date of Publication: 1 Feb 1989.
[Article]
AN: 19416141
In Klippel-Trenaunay syndrome there are congenital vascular malformations, usually involving the limbs. It is characterized by extreme varicose veins, bone and soft tissue hypertrophy and pigmentary skin changes in the affected limbs. We report a 29-year-old pregnant woman with this syndrome. The combination of the physiologic changes which occur in the vascular system and in blood coagulation during pregnancy, and the malformations of this syndrome is very unusual. During the patient's pregnancy there was significant worsening of the typical complications. There was extreme dilatation of varicose veins in the left leg, from the ankle up to the posterior aspect of the buttock. The involved area was swollen and edematous, and there were signs of superficial thrombophlebitis. Because we were unable to rule out deep vein thrombosis in the affected limb, prophylactic anticoagulant treatment was given during the 3rd trimester and into the early puerperium. Only then did the complications begin to resolve, but return to the prepregnant state was incomplete.
PMID
2541057 [http://www.ncbi.nlm.nih.gov/pubmed/?term=2541057]
Date Created
19890619
Year of Publication
1989

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**39.**
**Nonimmune hydrops fetalis associated with angioosteohypertrophy (Klippel-Trenaunay) syndrome.**
Mor Z., Schreyer P., Wainraub Z., Hayman E., Caspi E.
Embase
American Journal of Obstetrics and Gynecology. 159 (5) (pp 1185-1186), 1988. Date of Publication: 1988.
[Article]
AN: 19002452
A case of nonimmune hydrops fetalis in association with angioosteohypertrophy (Klippel-Trenaunay) syndrome is reported for the first time.
PMID
2847530 [http://www.ncbi.nlm.nih.gov/pubmed/?term=2847530]
Status
EMBASE
Institution
(Mor, Schreyer, Wainraub, Hayman, Caspi) Department of Obstetrics and Gynecology, Assaf Harofe Medical Centre, Zerifin 70300 Israel
Publisher
Mosby Inc. (11830 Westline Industrial Drive, St. Louis MO 63146, United States)
Date Created
19890105
Year of Publication
1988

40.
**Pregnancy and labour in a woman with the Klippel-Trenaunay syndrome.**
Powolny M.
Embase
Ginekologia Polska. 57 (3) (pp 195-197), 1986. Date of Publication: 1986.
AN: 16086233
PMID
3015747 [http://www.ncbi.nlm.nih.gov/pubmed/?term=3015747]
Status
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Institution
(Powolny) Z Oddzialu Polozniczo-Ginekologicznego Centralnego Szpitala Klinicznego MSW, Warszawa Poland
Date Created
19860821
Year of Publication
1986

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1 "angioosteohypertrophy syndrome".mp. [mp=ti, ab, hw, tn, ot, dm, mf, dv, kw, fx, nm, kf, px, rx, an, ui, sy] (1910)
2 "Klippel-Trenaunay".mp. [mp=ti, ab, hw, tn, ot, dm, mf, dv, kw, fx, nm, kf, px, rx, an, ui, sy] (3206)
3 1 or 2 (3885)
4 pregn$.mp. [mp=ti, ab, hw, tn, ot, dm, mf, dv, kw, fx, nm, kf, px, rx, an, ui, sy] (1945422)
5 3 and 4 (224)
6 remove duplicates from 5 (153)
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