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Case Report

UNILATERAL NEVOID TELANGIECTASIA

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ABSTRACT
Unilateral nevoid telangiectasia (UNT), is a rare vascular skin lesion which occur in unilateral dermatomal distribution in cervical, trigeminal and upper thoracic regions. UNT may be congenital or acquired. In this report our aim is to present a case with UNT that is first developed during pregnancy. A 32-year-old woman who is 24 weeks pregnant presented with a complaint of reddish blotch located on her left upper arm, shoulder and chest. She reported that the reddish blotch was present for 2 weeks. There is no itching and history of exposure to sunlight. She received progesterone 250 mg intramuscular injection only once at 9th week of pregnancy. She was using oral multivitamins and iron. Her personal and family history was unremarkable. Dermatological examination and dermoscopy revealed widespread telangiectasia located in her left shoulder region. She was diagnosed as UNT developed during pregnancy. UNT is an uncommon vascular skin lesion that has two forms: congenital and acquired. Whereas congenital forms are mostly seen in men, acquired forms are mostly seen in female. Even so etiopathogenesis is still not clear, UNT may relate to grade of estrogen and progesterone in body. At second trimester of pregnancy, our patient developed UNT lesions in her left upper arm, shoulder, and chest as a result of increased estrogen levels.

Key words: Acquired, nevoid, nevus, telangiectasia, vascular

ÖZET
Tek taraflı nevoid telanjiyektazi (UNT), servikal, trigeminal ve üst torasik bölgelerde tek taraflı dermatomal dağılımda ortaya çıkan nadir bir vasküler deri lezyonudur. UNT doğuştan veya edinilmiş olabilir. Bu yazıda amacımız, hamilelik sırasında ilk kez gelişen UNT'li bir olgu sunmaktır. 24 haftalık gebe olan 32 yaşında kadın olgu, sol üst kol, omuz ve göğüste 2 haftadır mevcut kırmızımsı leke şikayeti ile başvurdu. Güneş ışığına maruz kalma veya herhangi bir kaşıntı öyküsü yoktu. Gebeliğin 9. haftasında sadece bir kez progesteron 250 mg intramüsküler enjeksiyon almıştı. Oral multivitamin ve demir kullanıyordu. Kişisel ve aile öyküsünde özellik yoktu. Dermatolojik muayene ve dermoskopide sol omuz bölgesinde yaygın telanjiektaziler saptandı. Bu bulgularla hastaya hamilelik sırasında gelişmiş UNT tanısı konuldu. UNT, iki formu olan nadir görülen bir vasküler cilt lezyonudur: konjenital ve edinsel. Konjenital formlar çoğunlukla erkeklerde görülürken, edinsel formlar çoğunlukla kadınlarda görülür. Etiyopatogenezi hala net olmamakla birlikte, UNT vücutta östrojen ve progesteron düzeyleri ile ilişkili olabilir. Hastamızın gebeliğin ikinci trimesterinde, artmış östrojen düzeylerinin bir sonucu olarak sol üst kol, omuz ve göğüsde UNT lezyonları geliştirdiği düşünüldü.

Anahtar kelimeler: Edinsel, nevoid, nevüs, telanjiektazi, vasküler

INTRODUCTION

Unilateral nevoid telangiectasia(UNT) is a rarely seen vascular disorder that was first described by Blaschko in 1899.[1] UNT is distributed in a linear and unilateral pattern that can be either in a congenital or acquired form.[2] Congenital ones are mostly seen in males and acquired ones mostly seen in women.[1,2] Acquired UNT may be associated with hyperestrogenic conditions, liver diseases, skin related conditions and there may be no association (Table 1).[3] Exact pathogenesis of this disorder is unknown. It is mainly located in trigeminal, cervical, and upper thoracic regions.[1]

Table 1. Characteristics of acquired and congenital types of UNT.

Acquired		Congenital
Mostly in women		
Associated with	No association	
Estrogen related Pregnancy Puberty in women Adrenarch in men Childhood in women	Hemodynamic disturbance	Mostly seen in man
Liver related Cirrhosis Alcholism Liver metastasis from Carcinoid tumor Hepatitis C infection Hepatitis B infection	unknown epidermal angiogenic factor	Congenital, appears during or after the neonatal period
Skin related pyogenic granuloma polymophous light eruption	Neuronal alteration or anormalies of perivascular connective tissue	Inherited in an autosomal dominant trait
Others Hyperthyroidism After chemotherapy	Increased concentration of adrenergic receptors in lesional skin	May be influenced by maternal estrogen

CASE REPORT

A 32-year-old woman who is 24 weeks pregnant presented with a complaint of reddish blotch located on her left upper arm, shoulder and chest. The patient's

weight is 78 kg and her height is 158 cm with a BMI of 31.2. She reported that the reddish blotch was present for two weeks. There is no itching and history of exposure to sunlight. She received progesterone 250 mg intramuscular injection only once at 9th week of pregnancy. She was using Decavit and Santofer as a drug. Her personal and family history was unremarkable. Dermatological examination and dermoscopy revealed widespread telangiectasia located in her left shoulder region (Figure 1 and 2). She was clinically diagnosed as UNT first developed at second trimester of pregnancy.

Figure 1. Patient presented with a complaint of reddish blotch located on her left upper arm, shoulder, and chest. Dermoscopy revealed widespread telangiectasia located in her left shoulder region.

DISCUSSION

We presented a case with UNT first developed at 22th week of first pregnancy. UNT may be of acquired and congenital types and the characteristics are shown in table 1.[1] Although the exact pathogenesis of UNT is unknown some authors proposed that it may be related to hyper-estrogenic conditions[2-6] According to some authors a localized enhance in estrogen or progesterone receptors caused by gene abnormality occur as a somatic mosaicism in the affected skin[1-6] UNT associated with hyperestrogenism is mainly related to pregnancy, puberty, use of oral contraceptives, hepatitis or other liver diseases. [1, 5] The pathogenesis of estrogen or progesterone induced UNT is proposed to develop as a result of endothelial receptor supra-expression found in cutaneous lesion compared to normal skin

or hypersensibility of the present estrogen and progesterone receptors found in lesional skin.[1] High estrogen may lead to endothelium-dependent vascular dilatation by increasing nitric oxide and prostacyclin levels and additionally blocking the vascular smooth muscle contraction.[6]

Estrogen hormone is steadily increased at second trimester of pregnancy and decreased at the end of gestation.[7] There are two case report articles published on appearance of UNT during pregnancy; one is a 35 year old woman and her lesions began during the last 6 months of gestation.[3] The other patient is a 33 year old women and her lesions appeared at 32nd week of gestation.[6] The biopsy in one of the reported pregnant case revealed presence of progesterone and absence of estrogen receptors in the affected skin.[6] In these two cases the lesions were reported to disappear in a few months after the delivery. [3,6] According to changes of the hormones in gestation period we assumed that UNT in our case is developed as a result of increased estrogen levels in the second trimester. [7]

When a physician confronts a case with UNT a careful clinical history of patient and his family could reveal clues about its clinical type. Dermoscopy and skin biopsy may be helpful. During the physical examination of patient whole skin and systemic physical examination may reveal clues whether it is a syndromic one. After establishment of the diagnosis of UNT some laboratory tests must be ordered evaluating liver functions, hormones in female patients (estrogen, progesterone), hepatitis B and C serology and thyroid functions.[1] Typically acquired UNT suddenly will get well.[2,3] Our case was follow up for 14 months. Right now the patient is in 10th month of postpartum and her lesions are discolored like %80. She has just a little lesions like little points in her chest and it's not problem for her. Pulse dye laser is the first choice for the treatment of UNT. [1-4]

CONCLUSION

UNT is an uncommon vascular skin lesion that has two forms; congenital and acquired. Whereas congenital forms are mostly seen in men, acquired forms are mostly seen in female. Even so etiopathogenesis is still not clear, UNT may be related to the increased estrogen or progesterone levels in body. At second trimester of pregnancy, our patient developed UNT lesions in her left upper

arm, shoulder and chest as a result of increased estrogen levels.

REFERENCES

1. Ţăranu T, Condurache O, Toader MP et al. Acquired idiopathic unilateral nevoid telangiestasia. Romanian J Functional & Clinical, Macro- & Microscopical Anatomy & of Anthropology vol XVI-Nr.1 – 2017.
2. Derrow AE, Adams BB, Timani S, et al. Acquired unilateral nevoid telangiectasia in a 51-year-old female. Int J Dermatol 2008; 47: 1331-3.
3. Guedes R, Leite L. Unilateral nevoid telangiectasia: a rare disease? Indian J Dermatol 2012; 57: 138-40.
4. Oliveira A, Velho G, Sanches M, et al. Unilateral nevoid telangiectasia--report of two cases. Int J Dermatol 2014; 53: e32-3.
5. Tanglertsampan C, Chanthothai J, Phichawong T. Unilateral nevoid telangiectasia: case report and proposal for new classification system. Int J Dermatol 2013; 52: 608-10.
6. Michelerio A, Rivetti N, Bolcato V, et al. Unilateral nevoid telangiectasia: a condition unmasked by hyperoestrogenism? Eur J Dermatol 2017; 1; 27: 439-41.
7. John E.Hall. Guyton and Hall Textbook of Medical Physiology, Pregnancy and Lactation, 12th edition 2011, pp: 1007.