Ege Journal of Medicine / Ege Tip Dergisi 2016;55(3):140-142

Lipedematous scalp with obesity in a child Çocukta obezite ile birlikte lipoödematöz skalp Emel Ataş Berksoy¹ Özlem Bağ¹ Tanju Çelik¹ Hülya Tosun Yıldırım² Davi Habif³ Aysel Öztürk¹ ¹Dr. Behçet Uz Children's Hospital, Clinic of Pediatrics, İzmir, Turkey ²Dr. Behcet Uz Children's Hospital, Clinic of Pathology, İzmir, Turkey

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Abstract

An 11-year-old girl applied for having a spongy consistency of swellings in various part of her scalp. The overlying skin and hair growth of these areas appeared normal. There was no evidence of systemic disease except for exogenous obesity. Ultrasonography showed thickening of the subcutaneous tissue on the swelling part of the scalp. Histopathological examination revealed elastic fibre fragmentation on dermis and increased thickness in the subcutaneous fat tissue containing numerous hair follicles. The clinical features and the histopathological changes in the lesion were consistent with a diagnosis of lipedematous scalp, which has previously been described predominantly in adult women.

Keywords: Child, lipedematous scalp, obesity.

Öz

On bir yaşındaki kız çocuğu başının çeşitli bölgelerinde süngerimsi yapıda şişlikler olması nedeniyle başvurdu. Bu bölgelerdeki saç gelişimi ve cilt normal görünüyordu. Eksojen obezite dışında sistemik hastalık bulgusu yoktu. Ultrasonografi şişlik bölgesinde cilt altı doku kalınlığında artış olduğunu gösterdi. Histopatolojik inceleme dermisde elastik liflerde kırılma ve çok sayıda kıl follikülü içeren cilt altı yağ dokusunda kalınlık artışı olduğunu ortaya çıkardı. Klinik özellikler ve lezyondaki histopatolojik değişikler daha önce çoğunlukla yetişkin kadınlarda tanımlanan lipoödematöz skalp tanısı ile uyumlu idi.

Anahtar Sözcükler: Çocuk, lipoödematöz skalp, obezite.

Introduction

Lipedematous scalp (LS) is a rare condition characterized by increased thickness of subcutaneous tissue of the scalp. The exact etiology is unknown, but the characteristic finding is increscent in scalp thickness due to expansion of subcutaneous adipose tissue in the absence of adipose tissue hypertrophy or hyperplasia (1). If there were accompanying hair loss, the condition would have been called lipedematous alopecia (LA) (2). Both of them have been reported mostly in black women in the literature (1,3). Herein we report a white female child with lipedematous scalp associated with obesity.

Case Report

An 11-year-old Turkish female child presented with a fifteen day history of painful swelling of various part of the scalp including the forehead.

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On inspection, she looked fine but she had a moon face and was wearing a hair cover very tightly bound to her head.

The case's body mass index was 29.1(>95 p for age and sex). All the vital signs including blood pressure were normal. Physical examination revealed soft, bogging painful swelling over local areas of parietal and occipital part of the scalp and the forehead. There was no hair loss on these regions (Figure-1a). On palpation, the swellings on the scalp pressed down like a ball, and then returned to their initial shape when pressure was relieved. There was no family history and no medical history. Systemic and neurological examinations were normal. All the laboratory values including thyroid function tests, cortisol, ACTH (adrenocorticotropic hormone) levels, and lipid profile were normal except fasting insulin level (42.93 µIU/mL, indicating insulin resistance). The soft tissue ultrasonography of the scalp showed marked thickening of subcutaneous tissue up to 7 mm on the lesion site. A biopsy was performed on the swelling located at the parietal side of the scalp and its histopathological evaluation proved the thickening of the

subcutaneous tissue of the scalp without adipose tissue hypertrophy or hyperplasia by Von-Gieson staining for elastic fibers, trichrome staining for collagen fibers and Periodic Acid-Schiff Alcian-blue (PAS) for dermal mucin in addition to routine Hematoxylin-Eosin staining (Figure-1b and Figure-1c). The patient was diagnosed as lipedematous scalp with clinical and histopathological findings. There was no evidence of systemic disease except for exogenous obesity. The patient was referred to the pediatric endocrinology clinic for the evaluation of insulin resistance and obesity. Although the patient was followed up for weight control and treated with metformin for insulin resistance, she failed to lose weight and at one year follow-up, scalp swelling continued with no change, with further complaints added as itching and scarring of the scalp.

Ethical approval from the hospital administration and informed consent from the patient were obtained for publishing.



Figure-1.a. Slight swelling observed on the left side of the patient's forehead. b.Epidermis, dermis and thickened subcutaneous fat tissue containing numerous hair follicles (Hematoxylin Eosin x10).
c.Elastic fiber fragmentation on dermis (Von Gieson x10).

Discussion

First described by Cornbleet in 1935, lipedematous scalp is characterized by localized or diffuse spongy thickening of scalp. The spongy swellings are only palpable rather than visible and may be painful. Irritation on the scalp dermis, headache and pruritus on the swellings might accompany. Our patient complained of headache, pain on palpation, sometimes pruritus on the swelling part of the scalp. The reason for these symptoms is not known. Although our patient presented with headache and scalp tenderness, she complained about itching and scarring of the scalp during the follow-up period.

The main etiology is still not known. But the characteristic pathology is scalp thickness resulting from expansion of subcutaneous fat tissue in the absence of

adipose tissue hypertrophy or hyperplasia. Recently, the increase in the reported cases in Asiatic women and even in children would reduce the etiologic role of the racial and the age factor (4).

The histopathological changes include dilated dermal lymphatic vessels, usually normal epidermis, increased thickness of the subcutaneous adipose tissue and dermal edema (2). In our patient, we found elastic fragmentation, coarse collagen bundles and mild perivascular mononuclear cell infiltration in superficial dermis, increased subcutaneous fat tissue with hair follicles and normal dermis. The reason of the dermal lymphatic vessel dilatation is not known. Hormonal changes and the use of tightly bound hair bands as in our case have been implicated for the prevalence in women (5). However, although our patient did not use hair bands anymore, her complaints continued. Obesity (6) or genetic factors (7) may have an effect on lymphatic dilatation which may be the primary pathology. In recent years, the increasing numbers in cases in pediatric literature may be related to the increased prevalence of obesity in childhood. Because of its effects on adipose tissue structure, leptin may have a role for the pathogenesis of LS (6). However, we could not investigate leptin.

In obese patients, it is not enough to explain this entity only with abnormal lipid distribution. In these patients, there may probably be difficulty in lymphatic flow (7). As in other cases in the literature we could not investigate the lymphatic drainage. Beside these; as she was a single child and had no family history, we did not research the genetic basis of the entity.

Literature consists of very rare case reports of LS or LA in children. The first reported case was a nine year Maori girl with LS (8). Zeng YP, et al. (9) reported a Chinese boy with LS associated with heterochromia of scalp hair. Recently Müller, et al. (10) have suggested both of the entities within the group of lipomatosis. They reported a 15-year-old Turkish male with LS according to the current literature. None of the reported cases in the pediatric literature was accompanied by obesity. Up to now there is no treatment for this disorder.

In our opinion, the important point here is to recognize LS which is rarely encountered in the field of dermatology. The majority of children with LS (not LA) may first contact their family physician or pediatrician, as in our case. If the primary care physician can recognize this condition, prevent unnecessary interventions and medications will be prevented.

In conclusion, we suggest that LS may be more widespread among children than reported and detailed investigations on the lymphatic system, leptin levels and genetic studies can help to understand this rare entity.

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