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A Case of Sporadic Bilateral Polythelia with High-Grade Myopia

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Author's contribution

The sole author designed, analyzed, interpreted and prepared the manuscript.

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Case Study

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ABSTRACT

Aim: Polythelia is a rare condition. It is known to be associated with urogenital anomalies. Also, there are other types of rare anomalies like hand or mandible anomalies related with this anomaly. However, our case presents the first case of polythelia with ophthalmic abnormalities.

Presentation of Case: A 22-year-old male with congenital bilateral nodular inframammary macules with aesthetic complaints and high-grade myopia. Lesions were excised and concluded as polythelia. Ultrasonic examinations revealed no structural abnormalities in urogenital system.

Discussion: Polythelia is a rare condition with several accompanying abnormalities. Mostly, it is associated with urogenital anomalies. Our case showed no signs of urogenital deformities. Only deformity he had was high-grade myopia which began at infancy.

Conclusion: Our report is the only article that reports the possible association of polythelia with ophthalmic deformities.

Keywords: Polythelia; urogenital anomalies; ophthalmic anomalies.

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1. INTRODUCTION

Polythelia means supernumerary areolar structures and is a rare condition. It's incidence is between 0.4 to 6% in general population [1]. It is the most common supernumerary breast tissue type of all 8 classes that was defined by Kajava in 1915 [2]. It is generally a unilateral anomaly. It is associated with urinary tract anomalies, with a rate of 11 to 30% [3]. Also, several other anomalies have been hypothesized to be related with this anomaly. However, an ocular anomaly was not mentioned before to be related with this entity. This paper present such a case with bilateral polythelia and high-grade myopia.

2. PRESENTATION OF CASE

A 22 year old male patient was administered to the clinic with aesthetic complaint of congenital supernumerary nipple counts below the pectoral region. His familial history revealed no such supernumerary breast abnormalities. Only one of an elder relevant above 70 years old had senile cataract which was operated. He had no history of signs of urinary tract, cardiac or skeletal abnormalities. His only history of a disease was of a high-grade myopia. It was dated back to his childhood and was developed to as high as 11 diopters. Intellectual development was pertinent with his age. Physical examination revealed bilateral papillary structures based on symmetrical macular hyper-pigmented areas with 3 cm in diameter. Ocular examination revealed only myopia with 11 diopters. Blood work and urine analysis showed no functional urinary or cardiac problems. After a full operative written consent, both lesions were excised with close

margins for histopathological examination. Pathology report was concluded as ectopic areolar tissue. Meanwhile, genitourinary anomaly screening was done by ultrasonography (USG) and no structural anomaly, including testicular anomalies, were coincided.

3. DISCUSSION

First described by Leichtenstern in 1878, and classified by Kajava in 1915, supernumerary breast tissue malformations have been described well [4]. Polythelia, one of these malformations had been shown to occur mostly on the line between axilla and pubic region, which is called "the milk line" unilaterally. It occurs seldomly on other parts of the body, and rarely bilateral. [5, 6] Our case demonstrates bilateral polythelia and is a rare case because of this feature.

This group of malformations had been shown to be related with urogenital malformations and malignancies especially [5]. Also, cardiac anomalies, facial and manual skeletal anomalies were shown to be related with this entity [7,8]. In our case, we found only high grade myopia with infancy-onset, instead of urogenital anomalies. To our knowledge, this may be the first report establishing such a relationship. In the literature, no ophthalmological abnormalities with supernumerary breast tissues have been presented, neither as coincidence or associated finding before. On the other hand, our case's myopia had infancy onset. In our opinion, this is a strong indicator of a concomitant congenital anomaly. Therefore, to establish such a relevance, congenital supernumerary breast tissue patients should be consulted to an



Fig. 1. Preoperative frontal view of the case with bilateral ploythelia

ophthalmologist for hidden abnormalities, in the future.

The strongest relationship is with the urogenital anomalies in the literature. USG and urine analysis is a favorable combination for monitoring malformations, functional abnormalities and malignancies [3,9]. Thus, we screened the patient and no such abnormality was encountered. However, as suggested before, we believe it would be wise to monitor this patient lifelong for any urinary tract malignancies and functional disturbances.

4. CONCLUSION

Bilateral polythelia is a rare malformation. Urogenital malformations may accompany to this entity. Our case had only high-grade myopia since his childhood, but no urogenital malformations. This may be the first case report of such a patient.

CONSENT

The author declares that 'written informed consent was obtained from the patient for publication of this case report and accompanying images.

ETHICAL APPROVAL

It is not applicable.

COMPETING INTERESTS

Author has declared that no competing interests exist.

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