# CASE REPORT

#### DOI: 10.5336/caserep.2022-92653

### A Newborn with a Giant Congenital Melanocytic Nevus and Hyperbilirubinemia: What to do Now

Duygu ÇALIŞKAN<sup>a</sup>, <sup>©</sup> Tamer KUYUCU<sup>a</sup>, <sup>©</sup> Mert HATİPOĞLU<sup>b</sup>

<sup>a</sup>Department of Pediatrics, Alanya Alaaddin Keykubat University Alanya Training and Research Hospital, Antalya, Türkiye <sup>b</sup>Department of Emergency Medicine, Alanya Alaaddin Keykubat University Alanya Training and Research Hospital, Antalya, Türkiye

ABSTRACT Giant congenital melanocytic nevi (GCMN) are rare skin lesions that affect approximately 1 in 20,000 live births. GCMN must be followed up closely because of its association with an increased risk of malignant degeneration. We report a newborn with a giant lession of 14 cm in diameter which was located on the chest and multiple satellite lesions were present on the body. The patient required phototherapy due to hyperbilirubinemia. Some studies suggest that phototherapy can increase the incidence of neonatalnevi, and melanocytic nevi is the most important risk factor for the occurrence of skin melanoma. Considering the risk of developing kernicterus, we concluded that the most appropriate solution for the patient was to cover the lesions and apply phototherapy in the prone position.

Keywords: Melanocytic nevus syndrome, congenital; nevus, pigmented; melanoma, cutaneous malignant

About 1% of live births present with melanocytic nevi, which are benign proliferations of melanocytic cells. Although there are various definitions for giant nevus, a covering of 5% of the total body surface area or at least 10-20 cm in diameter is accepted.1 Giant congenital nevi are clinically important because of the risk of malignant transformation, although they are rare lesions seen in 1 in 20,000 births.

### CASE REPORT

A female newborn, from a 28-year-old mother at 38 weeks of gestation through cesarean section with a birth weight of 2,580 g was born. A giant hyperpigmented flat to papular skin lesion with a diameter of 14 cm, covered the chest wall which contained irregularly shaped macules, papules, and nodules with variegated colors and thin hair (Figure 1). Multiple pigmented satellite lesions of size 0.5-1cm were also present over the back, abdomen and lower extremities (Figure 2). Transfontanel and abdominal ultrasonography and x-rays were performed for associated con-

Received: 09 Aug 2022

genital anomalies but they were normal. An magnetic resonance imaging of the spine and brain have been planned. At the postnatal 48th hour, the patient developed jaundice with a total serum bilirubin of 17.5 mg/dL.

Considering the risk of developing kernicterus, we concluded that the most appropriate solution for the patient was to cover the lesions and apply phototherapy in the prone position. After 10 hours of phototerapy the icter regressed and the bilirubin level was 9.8 mg/dL. Pediatric oncology, dermatology and genetics appointments were arranged for the patient and she was discharged. Informed consent was obtained from the family for the case publication.

## DISCUSSION

Melanocytic nevi are benign proliferations of accumulated melanocytic cells in the epidermis or dermis. Giant congenital melanocytic nevi (GCMN) is thought to arise due to a defect in the neuroectoderm during embryogenesis that leads to the irregular growth of melanoblasts, the precursor cells of melanocytes.<sup>2</sup>

Correspondence: Duygu ÇALIŞKAN

Department of Pediatrics, Alanya Alaaddin Keykubat University Alanya Training and Research Hospital, Antalya, Türkiye E-mail: drduyguersoy@gmail.com



2147-9291 / Copyright © 2023 by Türkiye Klinikleri. This is an open access article under the CC BY-NC-ND license (http://creativecommons.org/licenses/by-nc-nd/4.0/).

Accepted: 08 Nov 2022



FIGURE 1: A giant hyperpigmented flat to papular skin lesion at chest.



FIGURE 2: Multiple pigmented satellite lesions.

GCMN is important for its association with malignant melanoma. Swerdlow et al showed patients have a higher risk of melanoma whom have a nevus covering at least 5% of the body surface, of which our patient was also in this category.<sup>1</sup> Studies point to an increased incidence of neoplasia in patients with multiple congenital satellite lesions.<sup>3-5</sup> Our patient has both of these risk factors for developing melanoma. The estimated lifetime-risk for developing melanoma for these patients is 5-10%.<sup>3,6,7</sup>

It has been observed that, sarcomas, liposarcomas, rhabdomyosarcomas, undifferentiated small round cell or spindle cell and neurogenic tumors can also develop from GCMN in addition to malign melanoma. Çalkavur et al reported, a 3 year old patient with a primary diagnosis of round cell sarcoma who later diagnosed as malignant melanoma.<sup>8</sup>

Phototherapy is the first option for treating neonatal jaundice. It is effective and safe in reducing the high serum free bilirubin levels and limiting neurotoxic effects of biluribin. When excessive amounts of free bilirubin cross the blood-brain barrier, it can cause serious damage to the nervous system. Whether phototherapy causes DNA damage is still a controversial issue. Some studies claims that there is no harm of blue light on DNA.<sup>9</sup> On the contrary, other studies claim that phototherapy could cause DNA damage.<sup>10,11</sup>

There are studies showing a correlation between childhood cancers and phototherapy.<sup>12-14</sup> A previous study demonstrated that phototherapy can increase the incidence of neonatal nevi, and melanocytic nevi are the most important risk factor for the occurrence of skin melanoma.<sup>15</sup> Therefore, when children receive phototherapy, the nevi should be closely monitored to prevent the development of melanoma.

At the postnatal 48<sup>th</sup> hour, the patient developed jaundice with a total serum bilirubin of 17.5 mg/dL. Even considering the possibility of DNA damage from phototherapy, we could not exclude the risk of permanent neural damage in the patient and concluded that the most appropriate solution for the patient was to cover the lesions and apply phototherapy in the prone position. After 10 hours of intermittent phototherapy, the biluribin level reduced to around 9 mg/dL.

The purpose of this presentation was to discuss the management of hyperbiluribinemia in a patient with GCMN. It is a challenging case for us because of the risk of kernicterus on one side and the application of phototherapy to a newborn predisposing to malignancy on the other side. It was a difficult dilemma for us to decide as there is not a standard procedure for that situation.

#### Source of Finance

During this study, no financial or spiritual support was received neither from any pharmaceutical company that has a direct connection with the research subject, nor from a company that provides or produces medical instruments and materials which may negatively affect the evaluation process of this study.

### **Conflict of Interest**

No conflicts of interest between the authors and / or family members of the scientific and medical committee members or members of the potential conflicts of interest, counseling, expertise, working conditions, share holding and similar situations in any firm.

#### Authorship Contributions

Idea/Concept: Duygu Çalışkan, Tamer Kuyucu; Design: Duygu Çalışkan; Control/Supervision: Duygu Çalışkan; Data Collection and/or Processing: Mert Hatipoğlu; Analysis and/or Interpretation: Duygu Çalışkan; Literature Review: Duygu Çalışkan; Writing the Article: Duygu Çalışkan; Critical Review: Duygu Çalışkan; References and Fundings: Tamer Kuyucu, Mert Hatipoğlu; Materials: Duygu Çalışkan, Tamer Kuyucu, Mert Hatipoğlu.

### REFERENCES

- Swerdlow AJ, English JS, Qiao Z. The risk of melanoma in patients with congenital nevi: a cohort study. J Am Acad Dermatol. 1995;32(4):595-9. [Crossref] [PubMed]
- Viana AC, Gontijo B, Bittencourt FV. Giant congenital melanocytic nevus. An Bras Dermatol. 2013;88(6):863-78. [Crossref] [PubMed] [PMC]
- Krengel S, Hauschild A, Schäfer T. Melanoma risk in congenital melanocytic naevi: a systematic review. Br J Dermatol. 2006;155(1):1-8. [Crossref] [PubMed]
- Hale EK, Stein J, Ben-Porat L, Panageas KS, Eichenbaum MS, Marghoob AA, et al. Association of melanoma and neurocutaneous melanocytosis with large congenital melanocytic naevi--results from the NYU-LCMN registry. Br J Dermatol. 2005;152(3):512-7. [Crossref] [PubMed]
- Bett BJ. Large or multiple congenital melanocytic nevi: occurrence of cutaneous melanoma in 1008 persons. J Am Acad Dermatol. 2005;52(5):793-7. [Crossref] [PubMed]
- Ka VS, Dusza SW, Halpern AC, Marghoob AA. The association between large congenital melanocytic naevi and cutaneous melanoma: preliminary findings from an Internet-based registry of 379 patients. Melanoma Res. 2005;15(1):61-7. [Crossref] [PubMed]
- Zaal LH, Mooi WJ, Sillevis Smitt JH, van der Horst CM. Classification of congenital melanocytic naevi and malignant transformation: a review of the literature. Br J Plast Surg. 2004;57(8):707-19. [Crossref] [PubMed]
- Çalkavur Ş, Targan Ş, Ortaç R, Serdaroğlu E, Vergin C, Atlıhan F. Konjenital dev melanositik nevüs zemininde gelişen bir pediatrik nodüler malign melanom olgusu [A case of pediatric noduler malign

melanoma developed on basis of the congenital giant melanocytic nevus]. Adnan Menderes Üniversitesi Tıp Fakültesi Dergisi. 2000;1(3):35-7. [Link]

- Gómez-Meda BC, Barros-Hernández A, Guzmán-Bárcenas J, Lemus-Varela Mde L, Zamora-Perez AL, Torres-Mendoza BM, et al. Effects of blue light phototherapy on DNA integrity in preterm newborns. J Photochem Photobiol B. 2014;141:283-7. [Crossref] [PubMed]
- Aycicek A, Kocyigit A, Erel O, Senturk H. Phototherapy causes DNA damage in peripheral mononuclear leukocytes in term infants. J Pediatr (Rio J). 2008;84(2):141-6. [Crossref] [PubMed]
- Tatli MM, Minnet C, Kocyigit A, Karadag A. Phototherapy increases DNA damage in lymphocytes of hyperbilirubinemic neonates. Mutat Res. 2008;654(1):93-5. [Crossref] [PubMed]
- Tyson JE, Miller CC. Whether neonatal phototherapy increases the risk of cancer in children is a disturbing unresolved issue. Evid Based Med. 2017;22(1):39-40. [Crossref] [PubMed]
- Auger N, Laverdière C, Ayoub A, Lo E, Luu TM. Neonatal phototherapy and future risk of childhood cancer. Int J Cancer. 2019;145(8):2061-9. [Crossref] [PubMed]
- Brewster DH, Tucker JS, Fleming M, Morris C, Stockton DL, Lloyd DJ, et al. Risk of skin cancer after neonatal phototherapy: retrospective cohort study. Arch Dis Child. 2010;95(10):826-31. [Crossref] [PubMed]
- Matichard E, Le Hénanff A, Sanders A, Leguyadec J, Crickx B, Descamps V. Effect of neonatal phototherapy on melanocytic nevus count in children. Arch Dermatol. 2006;142(12):1599-604. [Crossref] [PubMed]