Case Report

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Trichoblastoma in a Newborn: A Case Report

Bir Yenidoğanda Trikoblastom: Olgu Sunumu

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ABSTRACT

Trichoblastoma is a benign skin tumor that arises from the hair germ, which is the precursor of the hair follicle. It is most frequently observed on the scalp and face. The occurrence of trichoblastoma in the pediatric population is extremely rare. As far as our research, we have not come across any reported case of trichoblastoma in newborns in the English literature. A 10-day-old male newborn was referred to our clinic because of swelling under the right neck since his birth. Physical examination revealed a painless, firm and mobile mass, approximately 3 cm in size on the right submandibular region. Magnetic resonance imaging showed a smooth contoured mass with diffuse contrast enhancement. Tru-cut biopsy results suggested that the tumor was composed of epithelial and myoepithelial component. Total mass excision was performed and the final pathological result was a trichoblastoma. The patient was discharged on the second day following the surgery and the subsequent two-year follow-up examination showed no signs of wound complications, indicating successful healing. Trichoblastoma should be taken into account in newborns with neck masses.

Keywords: Trichoblastoma, trichoepithelioma, newborn, neck mass, case report

ÖZ

Trikoblastoma, kıl folikülünün öncüsü olan kıl germinden kaynaklanan benign bir deri tümörüdür. En sık saçlı deride ve yüzde görülür. Pediatrik popülasyonda trikoblastoma oluşumu oldukça nadirdir. Araştırabildiğimiz kadarıyla İngilizce literatürde şimdiye kadar yenidoğanlarda bildirilen herhangi bir trikoblastoma olgusuna rastlamadık. On günlük erkek yenidoğan, doğduğundan beri var olan sağ boyun altında şişlik nedeniyle kliniğimize yönlendirildi. Fizik muayenede sağ submandibuler bölgede yaklaşık 3 cm boyutunda ağrısız, sert ve hareketli kitle tespit edildi. Manyetik rezonans görüntülemesi, yaygın kontrast artışına sahip düzgün konturlu bir kitle ile uyumluydu. Tru-cut biyopsi tümörün epitelyal ve miyoepitelyal bileşenlerden oluştuğunu gösterdi. Total kitle eksizyonu yapıldı ve nihai patolojik rapor trikoblastoma olarak sonuçlandı. Hasta postoperatif ikinci gün taburcu edildi ve sonraki iki yıllık takiplerinde herhangi bir komplikasyon saptanmadı. Boyun kitlesi ile başvuran yenidoğanlarda trikoblastom akılda tutulmalıdır.

Anahtar Kelimeler: Trikoblastoma, trikoepitelyoma, yenidoğan, boyun kitlesi, olgu sunumu

Introduction

Trichoblastomas are extremely rare in childhood and information about these lesions in the literature is primarily derived from studies conducted on adult patient groups. According to general consensus trichoblastoma originates from the hair germ, the precursor of the hair follicle. It is most commonly found as a solitary nodule on the scalp and face. It is mostly diagnosed in the fourth and fifth decades of life. In this case report, we aimed to share a newborn presenting with a submandibular mass which diagnosed as trichoblastoma.

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Case Report

A 10-day-old male newborn was referred to our clinic because of swelling under the right neck since his birth (Figure 1A). Antenatal and natal history were uneventful. Physical examination revealed a painless, firm, and mobile mass, approximately 3 cm in size on the right submandibular region (Figure 1B). Ultrasound imaging identified a 37x28 mm lesion which is hypervascular on color Doppler ultrasound. A smooth contoured mass with diffuse contrast enhancement on magnetic resonance imaging (Figure 2) was observed, then a tru-cut biopsy was performed. The biopsy results suggested that the tumor was composed of epithelial and myoepithelial components, but the distinction between malignant and benign was not possible.

Total mass excision was performed under general anesthesia when the patient was 4-month-old. The excised material measured approximately 4.5 cm in diameter and had a bilobed appearance.

The patient was discharged on the postoperative day 2 with a clean wound. The final pathological result was a trichoblastoma, showing focal CD10 positivity, predominantly featuring a stromal component with a micronodular pattern (Figure 3).

The 2-year follow-up examination revealed uneventful wound healing (Figure 4).

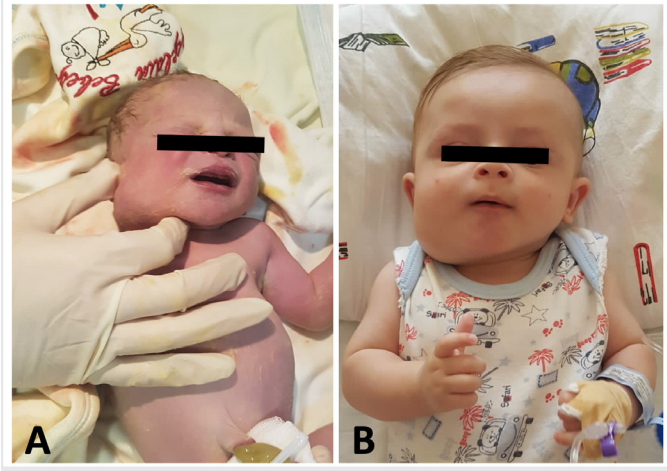

Figure 1. Clinical presentation of the patient. A) Neonatal period, B) Pre-operative period

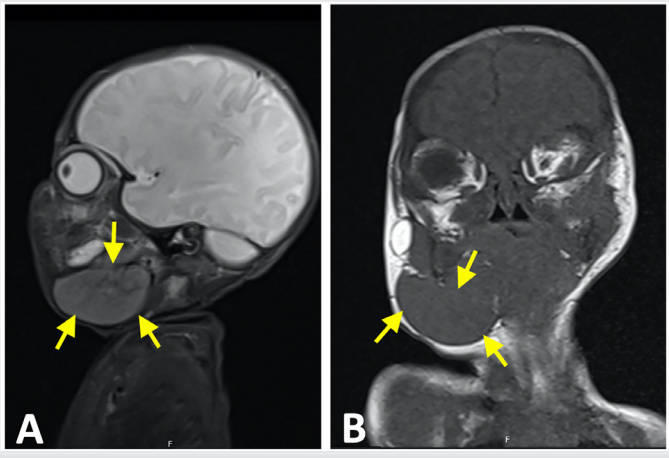

Figure 2. Magnetic resonance images. A) Sagittal T2-weighted image, B) Coronal T1-weighted image (The mass signed with yellow arrows)

Discussion

Trichoblastoma is a benign neoplasm originating from the hair germ, the precursor of the hair follicle (1,2). It is most commonly found as a solitary nodule on the scalp and face, usually less than 2 cm in diameter, and mostly diagnosed in the fourth and fifth decades of life (3). It is extremely rare in childhood. According to the best of our knowledge, we could not find any other case which was reported in the newborn period with trichoblastoma. Moreover, the other distinctive feature of our case was that it had an unusual location such as the neck at birth.

Trichoblastoma can occur sporadically, syndromically or secondarily. In detail, syndromic lesions are found in Curry-Jones and Brooke-Spiegler syndrome (4,5). Secondary lesions are

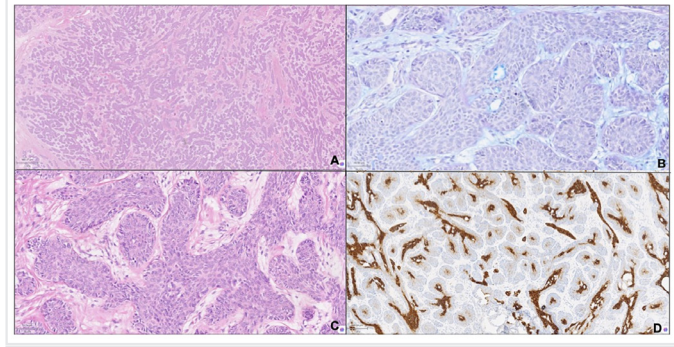

Figure 3. Hematoxylin-eosin stained sections. (A; C); PAS-AB histochemical study, a positive reaction was observed in the luminal secretion of the tumor (B) Luminal staining with CK7 (D)

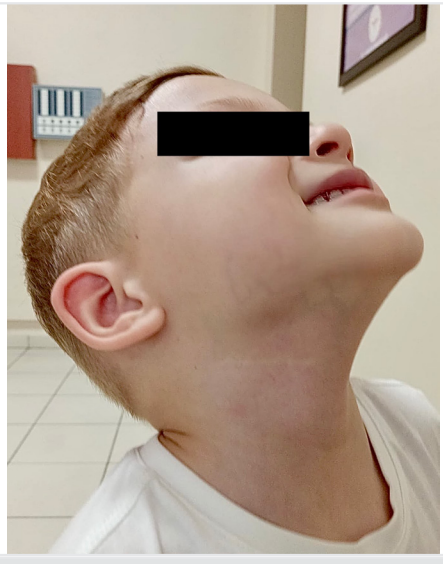

Figure 4. An uneventful wound healing after two years from surgery

mostly seen with nevus sebaceous. An evaluation for underlying familial genetic syndromes may be recommended in cases of multiple lesions (6,7). In the family history of our patient, neither a malign nor benign mass, nor a genetic disorder was identified. Furthermore, given the absence of multiple lesion formations in our patient, there was no need for genetic testing. Nevertheless, according to clinical features of our case, even though drawing conclusions based on a singular case is not advisable, it can be also speculated that congenital etiologies might play a role in the development of trichoblastoma. On the other hand, from a clinical perspective, despite regular antenatal follow-up, the absence of any signs during this period indicated the rapid growth of the mass, particularly in close proximity to delivery.

Pathologically, it can be classified as large nodular, small nodular, adamantine (lymphadenoma), retiform, and racemiform. Conventional and desmoplastic trichoepithelioma have also been referred to as cribriform and columnar trichoblastomas, respectively. The common feature of all these types is the presence of follicular germinative (basaloid) cells (2,3,7-9).

Despite its benign nature, it should be taken into account that it may be confused with basal cell carcinoma and the differential diagnosis between the two may be challenging in some cases (10). The main histopathological differential diagnosis includes basal cell carcinoma and trichoepithelioma. Basal cell carcinoma is a basaloid neoplasm that originates from the epidermis. As opposed to trichoblastoma, it is characterized by mitotic active basaloid nodules with necrosis and, with prominent peripheral palisading and clefting between the neoplasm and surrounding stroma (7,10).

Conclusion

Trichoblastoma shows only peritumoral stromal staining for CD10, whereas basal cell carcinomas typically show intraepithelial staining. The presence of few or many Merkel cells, androjen receptor negativity, PHLDA1 (follicular stem cell marker) positivity, peripheral BCL2 immunostaining in the trichoblastoma differentiates from basal cell carcinoma (7-10). Our case showed no necrosis, the tumor was located in the dermis without connection to the epidermis. It was also observed that the tumor had uniform basaloid cells with narrow cytoplasm, forming follicular papilla-like structures and arranged in layers within the stroma resembling a perifollicular sheath. The absence of peripheral palisading, clefting on the tumor, and immunohistochemical staining features made the distinction.

Trichoblastoma should be taken into account in newborns with neck masses. Additionally, congenital factors may also play a role in the etiology of trichoblastoma.

Ethics

Informed Consent: Written informed consent was obtained from the patient's parents to publish this case report and accompanying images.

Footnotes

Authorship Contributions

Surgical and Medical Practices: M.S.K., M.İ., Concept: M.S.K., C.E.Ç., D.T.T., F.Ö.P., M.İ., Design: M.S.K., C.E.Ç., D.T.T., F.Ö.P., M.İ., Data Collection or Processing: M.S.K., C.E.Ç., D.T.T., F.Ö.P., M.İ., Analysis or Interpretation: M.S.K., C.E.Ç., D.T.T., F.Ö.P., M.İ., Literature Search: M.S.K., C.E.Ç., D.T.T., F.Ö.P., M.İ., Writing: M.S.K., C.E.Ç., D.T.T., F.Ö.P., M.İ.

Conflict of Interest: No conflict of interest was declared by the authors.

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