Received: 18 August 2023 | Revised: 1 September 2023 | Accepted: 12 September 2023

DOI: 10.1002/ccr3.7941

CASE REPORT

Clinical Case Reports Open Access WILEY

Partially involuting congenital hemangioma with pyogenic granuloma-induced rapid progression following incisional biopsy in infancy: A case report

Yuki Sasaki | **Kosuke Ishikawa** | **Takahiro Miura** | **Emi Funayama** | **Yuhei Yamamoto** | **Taku Maeda**

Department of Plastic and Reconstructive Surgery, Faculty of Medicine and Graduate School of Medicine, Hokkaido University, Sapporo, Japan

Correspondence
Taku Maeda, Department of Plastic and Reconstructive Surgery, Faculty of Medicine and Graduate School of Medicine, Hokkaido University, Kita 15, Nishi 7, Kita-ku, Sapporo, Hokkaido 060-8638, Japan.
Email: takumaeda1105@yellow.plala.or.jp

Key Clinical Message

We present a case of partially involuting congenital hemangioma (PICH) that showed rapid growth with ulceration after incisional biopsy. PICH does not typically grow after birth. However, if growth occurs due to stimuli like biopsy, it is essential to consider the possibility of pyogenic granuloma complication.

KEYWORDS

dermatology, surgery, vascular surgery

1 | INTRODUCTION

Congenital hemangiomas are rare tumors that typically reach their maximal growth during the embryonic period. They are classified into three types: rapidly involuting congenital hemangioma (RICH), which rapidly regresses after birth[1]; noninvoluting congenital hemangioma (NICH), which does not regress[2]; and partially involuting congenital hemangioma (PICH), which exhibits an intermediate pattern of involution.[3] PICH demonstrates partial involution tendencies, but does not regress completely.[3] None of these three types of congenital hemangiomas usually grow after birth.[4] On the other hand, pyogenic granuloma is a benign lesion characterized by inflammatory cell infiltration and capillary proliferation, and it can arise in response to factors such as trauma or infection and may rapidly enlarge.[5] In this report, we present a case of PICH that showed rapid growth with ulceration after incisional biopsy.

2 | CASE REPORT

A male infant presented with a purple mass lesion located on the right upper arm since birth. The parents sought medical attention for him at 2 months of age due to lesion growth (Figure 1A). Magnetic resonance imaging at 5 months of age showed a relatively homogeneous 3 cm lesion within the subcutaneous fat layer, with low signal intensity on T1-weighted images (Figure 2A), high signal intensity on fat-suppressed T2-weighted images (Figure 2B), and contrast enhancement at the lesion margins (Figure 2C). No contrast enhancement was evident in the deltoid muscle fascia adjacent to the tumor, but dilated vessels were apparent surrounding the lesion (Figure 2C). Histopathological examination of the biopsy specimen revealed numerous dilated blood vessels in the dermis, and immunohistochemical staining was negative for both glucose transporter 1 (GLUT1) and D2-40, leading to a diagnosis of capillary malformation (Figure 3A).

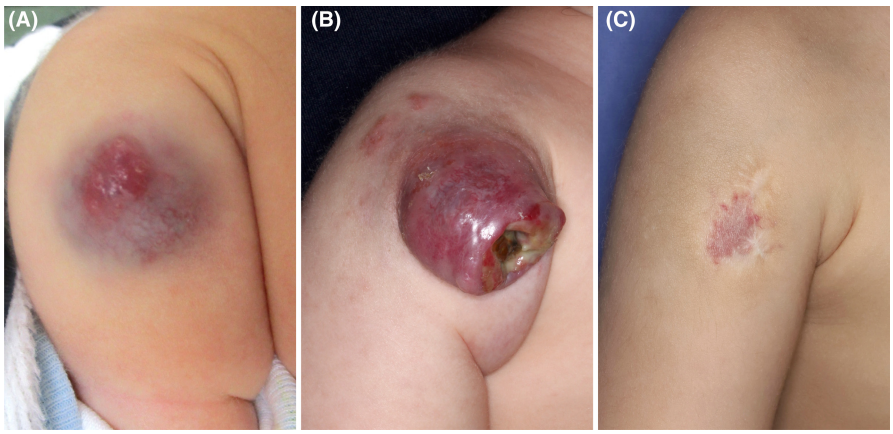

FIGURE 1 (A) A 2-month-old boy with congenital hemangioma. (B) Rapid tumor growth after the biopsy upon referral to us at 8 months of age. (C) At the age of 3 years, the lesions had regressed, resulting in the presence of erythema and scarring of the biopsy site.

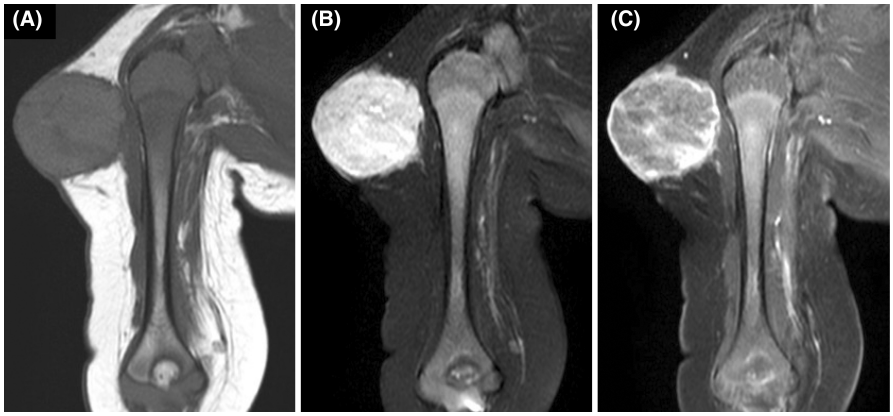

FIGURE 2 Magnetic resonance images at 5 months of age: (A) T1-weighted image, (B) fat-suppressed T2-weighted image, and (C) fat-suppressed gadolinium contrast image.

Subsequently, the lesion grew rapidly and exhibited ulceration and necrosis after the biopsy, and the patient was referred to our department at the age of 8 months (Figure 1B). Laboratory investigations revealed no evidence of thrombocytopenia that would suggest the presence of Kasabach-Merritt phenomenon. To rule out malignancy, an additional incisional biopsy was conducted. The histopathologic examination revealed a pale fibrous interstitium intervening from dermis to subcutis, proliferation of dilated vessels with severe congestion, and partial necrosis (Figure 2B). A microvascular component with a tendency to proliferate in some areas and thick-walled vessels was also observed (Figure 2C). Immunohistochemistry revealed positive anti-CD31 staining (Figure 2D), negative anti-D2-40 staining (Figure 2E), and negative anti-GLUT1 staining (Figure 2F) in the vascular endothelium. Based on these findings, congenital hemangioma with dilated capillaries secondary to changes caused by congestion was diagnosed. Thereafter, the mass regressed to small erythema at the end of 1 year. Consequently, a clinical diagnosis of PICH was made. At the age of 3 years, there was no regrowth of the lesion (Figure 1C).

3 | DISCUSSION

PICH typically presents as a mass characterized by surface telangiectasia and a surrounding pale halo,[4] and it is recognized to exhibit partial regression by 12 months of age, with further regression attainable by 30 months of age.[3] Histopathological examination suggests that PICH involves the dermis to subcutis with a background of fibrosis, capillary lobules, and extra-lobular abnormal veins.[6] The present case exhibited regional capillary hyperplasia with an intervening pale fibrotic interstitium, consistent with PICH. PICH is histopathologically indistinguishable from RICH and NICH and is diagnosed clinically.[6] Unlike infantile hemangioma, PICH exhibits negative expression of GLUT1.[6]

In general, congenital hemangioma does not exhibit growth after birth.[3] However, in the present case, there was a rapid growth with ulceration and necrosis following the initial biopsy. This may have been caused by pyogenic granuloma, a complication resulting from infection at the biopsy site. Symptoms of pyogenic granuloma include friable granulation tissue-like changes and hemorrhage, and the histopathological findings reveal granulomatous microvascular proliferation, which is consistent with the present case.[7] To date, only two cases of pyogenic granuloma complicating congenital hemangioma have been reported.[7,8] In one case, friable granulation tissue-like changes with bleeding spontaneously developed within an infant's congenital hemangioma located on the mandible.[7] Successful treatment was accomplished through the complete excision of the

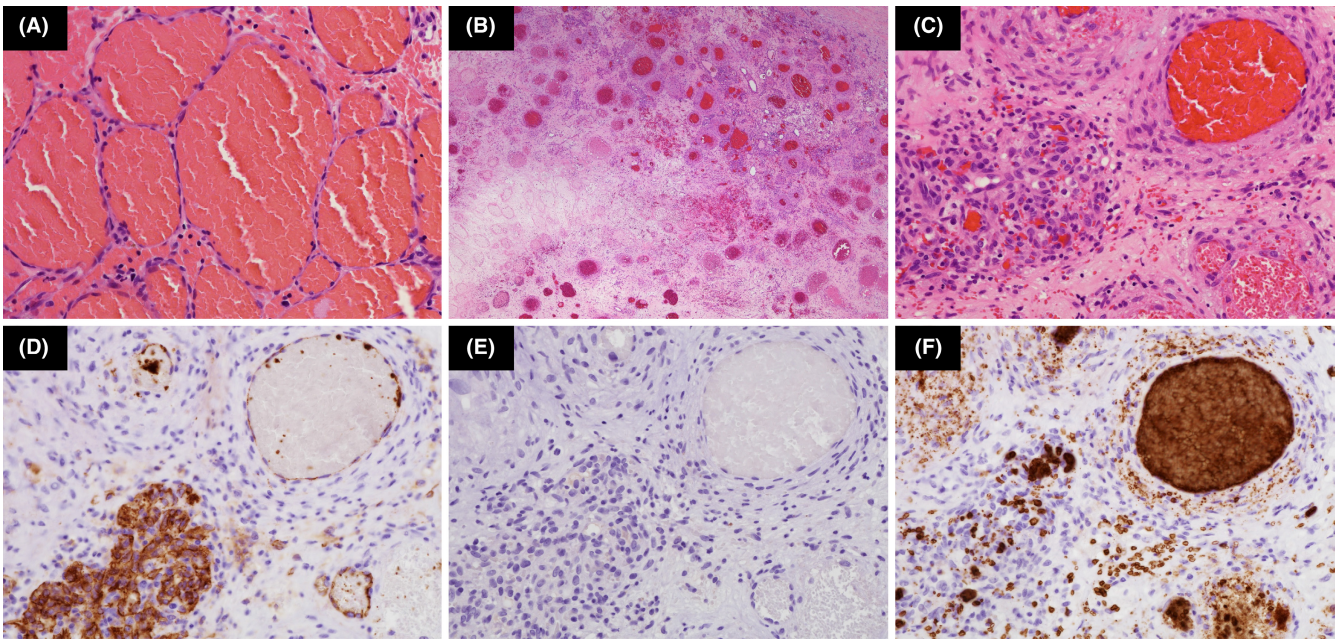

FIGURE 3 (A) Hyperplasia of thin vessels with dilated walls in the dermis of the initial biopsy specimen (Hematoxylin and eosin, ×200). (B) An additional biopsy specimen showing a regional distribution of dilated vessels and necrosis with severe congestion from dermis to subcutis (Hematoxylin–eosin stain, ×20), and (C) a microvascular component with a tendency to proliferate in some areas and thick-walled vessels, leading to a diagnosis of congenital hemangioma with dilated capillaries secondary to changes caused by congestion (Hematoxylin–eosin stain, ×200). The vascular endothelial cells exhibit (D) positive immunostaining for anti-CD31 (×200), (E) negative immunostaining for anti-D2-40 (×200), and (F) negative immunostaining for anti-glucose transporter 1 (GLUT1) (×200). It should be noted that erythrocytes demonstrate positivity for GLUT1.

lesion.[7] Histopathologically, the presence of pyogenic granuloma was confirmed.[7] In the other case, an erythematous nodular mass with bleeding was observed in an adult's NICH located on the penis.[8] Following an incisional biopsy, a concurrent diagnosis of NICH and pyogenic granuloma was established.[8] In this instance, treatment involved complete excision of the lesion and skin grafting.[8] Congenital hemangiomas may be surgically excised for curative purposes; however, performing resection while the hemangioma is enlarged can lead to more significant scarring and increased invasiveness.[6] If rapid growth is due to pyogenic granuloma complication, a conservative observational approach might enable the lesion's natural regression, potentially reducing the need for unnecessary interventions.

4 | CONCLUSION

We present a case of PICH complicated by pyogenic granuloma. To our knowledge, this is the first report of pyogenic granuloma after a biopsy of a congenital hemangioma in infancy. The continued investigation will offer insights into the etiology and management of congenital hemangiomas and the proliferative lesions associated with this condition.

AUTHOR CONTRIBUTIONS

Yuki Sasaki: Writing – original draft; writing – review and editing. **Kosuke Ishikawa:** Writing – review and editing. **Takahiro Miura:** Data curation. **Emi Funayama:** Conceptualization. **Yuhei Yamamoto:** Project administration. **Taku Maeda:** Project administration; writing – review and editing.

FUNDING INFORMATION

No fund was available for this study.

CONFLICT OF INTEREST STATEMENT

The authors declare no conflict of interest.

DATA AVAILABILITY STATEMENT

The data that support the findings of this study are available from the corresponding author upon reasonable request.

ETHICS STATEMENT

The patient has provided written informed consent for the publication of this case report.

CONSENT

Written informed consent was obtained from the patient to publish this report in accordance with the journal's patient consent policy.

ORCID

Yuki Sasaki https://orcid.org/0000-0002-1953-4366
Kosuke Ishikawa https://orcid.org/0000-0002-6068-7276
Takahiro Miura https://orcid.org/0000-0003-4180-6556
Taku Maeda https://orcid.org/0000-0003-3612-8201

REFERENCES

1. Boon LM, Enjolras O, Mulliken JB. Congenital hemangioma: evidence of accelerated involution. *J Pediatr*. 1996;128:329-335.
2. Enjolras O, Mulliken JB, Boon LM, Wassef M, Kozakewich HP, Burrows PE. Noninvoluting congenital hemangioma: a rare cutaneous vascular anomaly. *Plast Reconstr Surg*. 2001;107:1647-1654.
3. Nasseri E, Piram M, McCuaig CC, Kokta V, Dubois J, Powell J. Partially involuting congenital hemangiomas: a report of 8 cases and review of the literature. *J Am Acad Dermatol*. 2014;70:75-79.
4. Braun V, Prey S, Gurioli C, et al. Congenital haemangiomas: a single-Centre retrospective review. *BMJ Paediatr Open*. 2020;4:e000816.
5. Lin RL, Janniger CK. Pyogenic granuloma. *Cutis*. 2004;74:229-233.
6. El Zein S, Boccara O, Soupre V, et al. The histopathology of congenital haemangioma and its clinical correlations: a long-term follow-up study of 55 cases. *Histopathology*. 2020;77:275-283.
7. Barrick B, Lehman J, Tollefson M. Agminated pyogenic granuloma–like growth arising in a congenital hemangioma. *JAMA Dermatol*. 2014;150:781-783.
8. Movassaghi M, Wu J, Carpenter CP. Pediatric penile non-involuting congenital hemangioma with an associated pyogenic granuloma: surgical management of a rare vascular anomaly. *Urology*. 2021;158:197-199.

How to cite this article: Sasaki Y, Ishikawa K, Miura T, Funayama E, Yamamoto Y, Maeda T. Partially involuting congenital hemangioma with pyogenic granuloma-induced rapid progression following incisional biopsy in infancy: A case report. *Clin Case Rep*. 2023;11:e7941. doi:10.1002/ccr3.7941