

Giant Perineal Hemangioma in Adulthood: Clinical Insights from a Case Report

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Abstract

Benign tumors like hemangiomas can develop anywhere on the body. It is quite uncommon for genital hemangiomas to involve the scrotum, penis, or perineum. Rarely, they may spread to the perianal area, in which case preoperative imaging is vital to the course of therapy. The medical literature has very few case reports. A review of the literature is included here along with a rare case of massive hemangioma involving the entire perineum, spreading into the scrotum superiorly and into the perianal and pararectal area inferiorly in a 45-year-old man. We want to put a hypothesis that unexplained long-term sequelae of post-fistulectomy repair can present as acquired perineal hemangioma secondary to surgical trauma, posing a diagnostic dilemma to clinicians.

Key-words: Genital hemangioma, Perineum, Scrotum, Acquired hemangioma, Fistula in ano repair,

Introduction

The musculoskeletal system, liver, and spleen are the most prevalent sites for hemangiomas, which are the most commonly seen in children. Less than 1% of our scrotal and penile (genital) hemangiomas are uncommon abnormalities that can spread to nearby regions of the anterior abdominal wall, thigh, or perineum.[1]

The hemangioma could result from trauma or neoplasia, or it could be congenital. Rapid growth, spontaneous regression, and non-recurrence are the hallmarks of hemangiomas [2-4]. Most hemangiomas appear during the first two decades of life and may grow larger as the kid grows. Hemangiomas can involve both the skin and the subcutaneous layer, or they can be deep (subcutaneous) [2].

Clinical suspicion should be raised if the lesion had a history of fast growth in early childhood. During our review of the literature, we came across a few examples of perineal, scrotal, and penile hemangiomas. To our knowledge, though, this is the first and uncommon instance of a perineal hemangioma in a patient who is over 40, the etiology of this lesion was unclear. We show an adult giant perineal hemangioma, discuss its probable causes, and talk about the challenges in diagnosis and treatment.

Case Report

A 45-year-old male with no medical comorbidity presented with chief complaints of swelling over the perineal region, which was insidious in onset and progressive in nature over the past two years. The patient experienced discomfort due to the swelling. He had no complaints of pain, fever, urinary symptoms, or perianal discharge. He had undergone a fistulectomy for fistula in Ano 4 years back. On examination approximately 6 x 5 cm, soft, non-tender, non-

pulsatile, irreducible, lobulated swelling with normal overlying perineal skin was noted, extending superiorly into the scrotum and inferiorly into the perianal area (**Figure 1A**). Penis and bilateral testis were normal. The scar from the previous fistulectomy was noted at 5 o'clock. Digital rectal examination was normal.

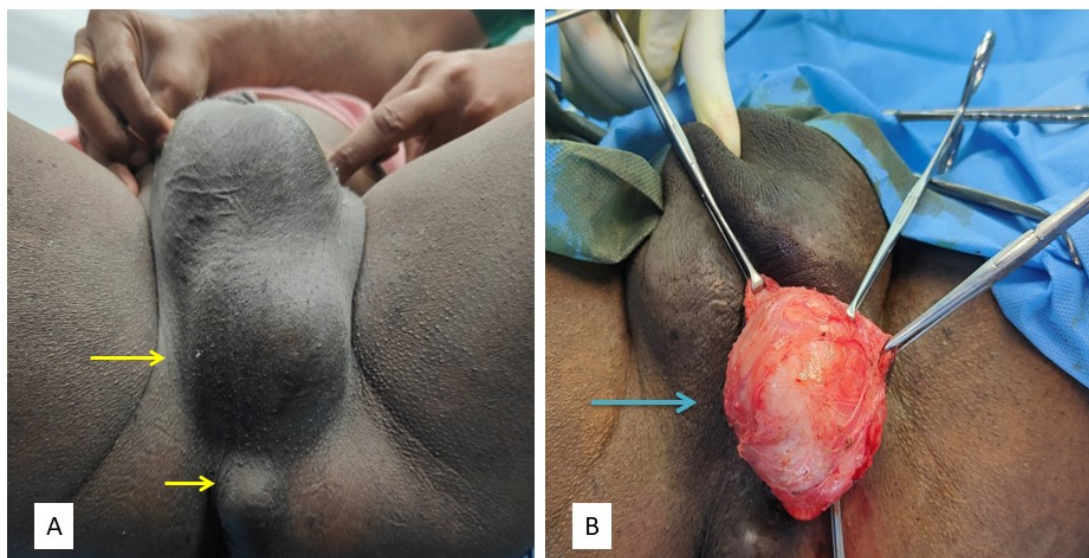


Figure 1: Clinical appearance and intraoperative findings.

Fig1A shows the pre-operative appearance of the hemangioma in the perineum with its extensions (Yellow arrow). Fig 1B illustrates intraoperative presentation of hemangioma (Blue arrow).

All baseline hematological and biochemical investigations were normal during the evaluation. On further evaluation, magnetic resonance imaging (MRI) of the perineum disclosed an elongated, smoothly margined, well-circumscribed, polypoidal lesion in the left perianal region, extending from the inter-sphincteric plane adjoining the site of focal thinning in the left posterior lateral infra levator anal canal at 4 to 6 o'clock position and anteriorly up to 1 o'clock position. This lesion is seen extending into the left hemi-scrotum, where it was seen to lie

posterior to the testis and epididymis (**Figure 2**). The lesion was 14 cm in length, with maximum cross-sectional dimensions of 5.5 x 5.4 cm. Few small blood vessels are seen within the lesion.

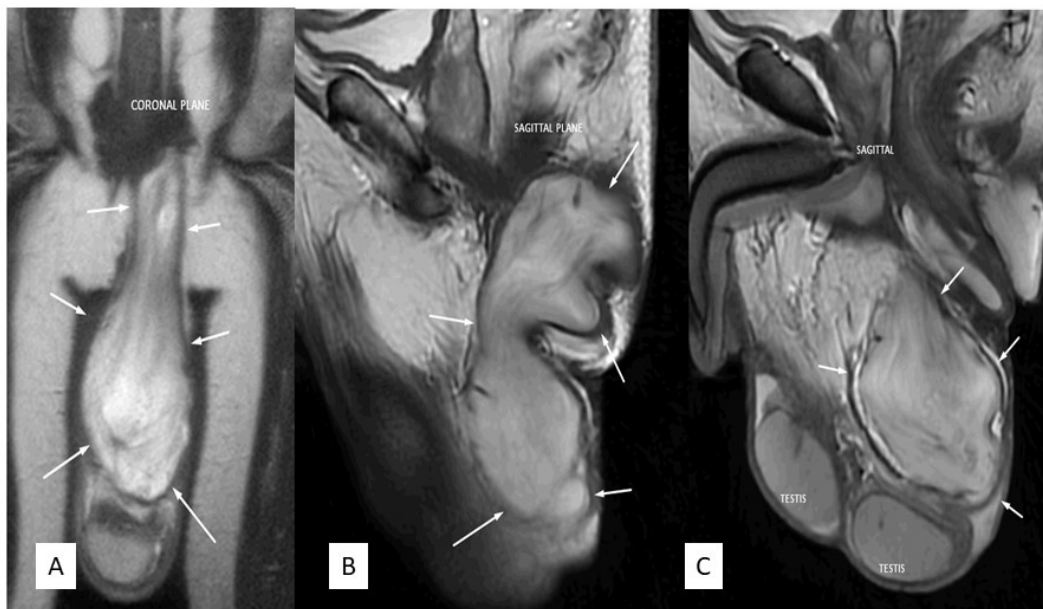


Figure 2: Magnetic resonance imaging (MRI) of perineum

Fig 2A illustrates MRI perineum revealed an elongated, smoothly margined, well-circumscribed, polypoidal lesion in the left perianal region in the coronal plane (white arrows denote extent). Fig 2B and 2C show lesion extends into the left hemi-scrotum, which was seen to lie posterior to the testis and epididymis in the sagittal plane (white arrows denote extent).

Based on the above radiological features, a provisional diagnosis was made as hemangioma. The patient underwent a cystoscopy with perineal mass excision. The urethra and bladder were normal on cystoscopic examination. An incision was made at the base of the scrotum and the mass was delineated by sharp and blunt dissection (**Figure 1B**). The mass was found to be grey-brown gelatinous measuring 15 x 8 x 4 cm in dimensions (**Figure 3A**), adherent to lateral pararectal space and underlying perineal muscles but not attached to the bilateral testis, root of

the penis, and urethra. Following complete tumor dissection, the perineum, and perianal region's undamaged tissue were approximated and the closure of the wound was done. The histopathologic examination report stated it was a hemangioma (**Figure 3B & 3C**). The patient recovered well with an uneventful postoperative period.

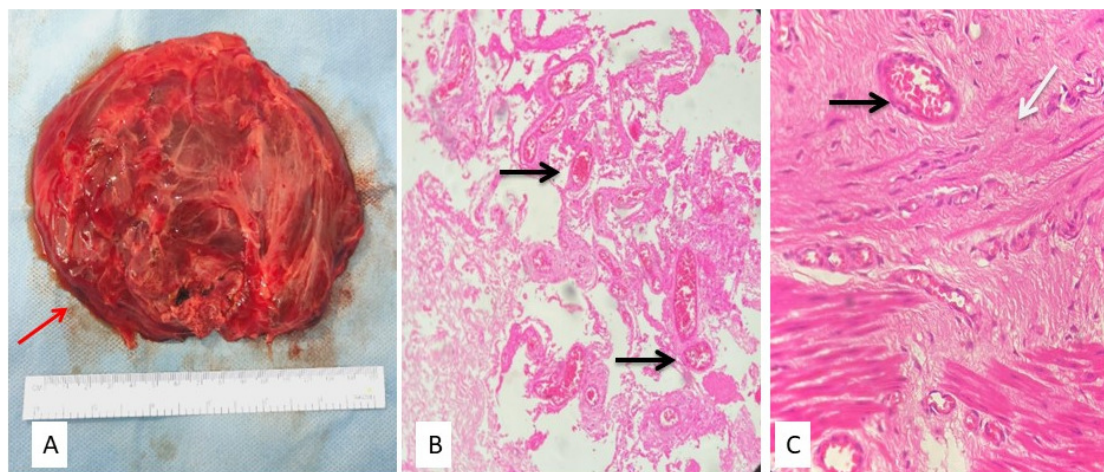


Figure 3: shows the removed specimen in both a gross and microphotograph.

Fig3A illustrates the Gross specimen- a grey-brown and gelatinous mass (Red arrow). Fig 3B Shows the histopathological appearance of hemangioma- blood vessels of varying size (Black arrow). 10X H & E Stain. Fig 3C Shows blood vessels (Black arrow) and fibro-collagenous tissue (White arrow).

Discussion

Hemangiomas typically involve the head and neck area and are superficial lesions. They are regarded by many researchers as benign tumors and congenital vascular abnormalities [5]. Adult cases of genital hemangiomas are extremely uncommon and have primarily been described in the childhood age range [6, 7]. During the first year of life, proliferation continues, and modest involution follows. Hemangiomas resolve in 30% of cases in 3 years, half in 5 years, and 70% to 90% in 7 years [2]. Adult genital hemangiomas involving the scrotum, penis, and perineum are uncommon. Osman Ergün et al. described a case of massive scrotal

hemangiomas in an adult patient [8]. However, our case report presents a massive *adult perineal hemangioma*. Our patient, a 45-year-old man, is an intriguing case due to his age and the location of the lesion.

Even though hemangiomas commonly are congenital, they could result from trauma or neoplasia. As per the chronological events in clinical history, the patient in our case developed this lesion after the surgical repair of the fistula in Ano and the patient denies any history of lesions in childhood period, we would like to put a hypothesis that it could be an acquired hemangioma secondary to trauma post-fistulectomy repair. Even though to date no report of hemangioma as a complication of fistula in ano repair is mentioned in literature. We like to attribute it to unexplained long-term sequelae of trauma due to fistulectomy. To understand the causal relationship between fistulectomy and the occurrence of perineal hemangioma further research is needed.

Imaging can be used to determine the hemangioma's extent and find any related abnormalities. A soft-tissue mass or prominence containing phleboliths, or tiny calcifications, is the most common radiography finding. Phleboliths strongly signal the presence of cavernous hemangioma [9, 10]. Cavernous hemangiomas can appear either hyperechoic or hypoechoic in sonographic pictures, depending on what kind of lesion is present (blood-containing units and septa, for example). Blood flow within these lesions may be visible on a Colour Doppler, but the lack of flow does not exclude the existence of these lesions [11]. Simple, non-invasive methods for diagnosing and assessing the size of these lesions, as well as identifying how they relate to nearby structures, are offered by computed tomography (CT) and magnetic resonance imaging (MRI). As such, these imaging modalities are the preferred ones for this condition and are required to be performed before any surgical treatments [10].

Eliminating the lesion should be advised after a diagnosis has been made. A sclerosing agent (such as alcohol), surgery, or laser therapy are available as treatments. In addition to having greater risks of complications (such as scarring, deformity, intraoperative bleeding, or recurrence), non-surgical therapy techniques are not always effective and produce results that are comparable to or less efficient than surgery [8]. In one trial, laser treatment had a 92.8% success rate and a low rate of acute complications (around 3.57%), with little deformity and scarring. There were no observed long-term side effects from laser therapy. According to the study's findings, treating these lesions with lasers yields positive outcomes with a very low rate of problems. In certain situations, laser treatment might be the best option [12, 13].

Because of the hemangioma's huge size and degree of extent, we chose to operate on the patient, and the surgery resulted in a great surgical outcome. Therefore, for big and numerous lesions, surgical excision is still a good choice, and with proper surgical planning and technique, the results are generally satisfactory.

Conclusion

1. Hemangioma that involves the perineum in adults is extremely rare, imaging studies like magnetic resonance imaging (MRI) help in planning therapy in these diagnostic difficulties, and surgical excision is preferable for large and multiple lesions with satisfactory outcomes.
2. Giant perineal hemangioma in an adult theoretically could be acquired due to unexplained long-term sequelae of trauma due to repair of fistula in Ano, but to understand their causal relationship further research is needed.

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