Recurrent Cirsoid Aneurysm of Face: A Rare Vascular Entity - Case Report

Dr Deepak Chhatbar *

ENT Consultant, Dhoraji City, Rajkot (Gujarat)

Abstract: Cirsoid aneurysms, although rare, represent an intriguing vascular pathology characterized by tortuous, dilated blood vessels with a propensity for arteriovenous shunting. While these lesions can occur in various regions of the body, their manifestation in the face (cheek) is exceptionally uncommon. This article presents case of 44 yr. male patient, suffering from recurrent Cirsoid aneurysm, rarely reported in literature. The main feeder for the aneurysm being Superficial Temporal artery and drainage to Internal Jugular vein. The Aneurysm was operated. Article involves a comprehensive review of cirsoid aneurysms, with a focus on their etiology, clinical presentation, diagnostic modalities, and management strategies. Through the exploration of a clinical case and pertinent literature, the article sheds light on the challenges associated with diagnosing and treating cirsoid aneurysms of the cheek.

Keywords: Cirsoid aneurysm, arteriovenous malformation, vascular anomaly, face, Superficial temporal artery, Internal jugular vein

Introduction:

Cirsoid aneurysms are rare vascular anomalies characterized by a tangle of dilated, tortuous vessels with arteriovenous communications. These lesions, also referred to as racemose hemangiomas, are rare vascular abnormalities distinguished by intricate networks of twisted arteries and veins, without a discernible capillary network in between. First described by Sir Astley Cooper in 1809, these lesions typically occur in the scalp, face, or extremities. However, their presence in the cheek region is exceedingly rare, posing diagnostic and therapeutic challenges for surgeons.[1] This article aims to elucidate the clinical features, diagnostic approach, and management options for cirsoid aneurysms of the cheek through a review of literature and a case illustration.

*Author, Email- deepakdyc@gmail.com

Etiology and Pathogenesis:

The exact etiology of cirsoid aneurysms remains elusive. However, they are thought to arise from congenital vascular malformations or acquired causes such as trauma or inflammation [2]. The pathogenesis involves abnormal vascular development leading to the formation of arteriovenous fistulas within the lesion, resulting in the characteristic arteriovenous shunting and subsequent vessel dilatation. Genetic predisposition and hormonal influences have also been proposed as contributing factors.

Histology- Tissue contain blood vessels. Intermingled with thick walled blood vessels are thin walled dilated blood vessels. Thick walled blood vessels resemble arteries, but they lack a well formed elastic internal membrane, and represent ectatic veins.[3]

Clinical Presentation:

Very few cases of cirsoid aneurysms have been reported worldwide. The aneurysms of the cheek typically present as a painless, pulsatile mass with a bluish hue, reflecting the underlying vascular nature of the lesion. Patients may report cosmetic concerns or discomfort due to the visible and palpable nature of the swelling. On appearance they are commonly seen as angleworm like tortuous, pulsating vessels. [4]

Rarely, symptoms such as localized warmth, tenderness, or audible bruits may be observed. Differential diagnoses include vascular tumours, arteriovenous malformations, or pseudoaneurysms, highlighting the importance of thorough clinical evaluation and diagnostic workup.

Diagnostic Modalities:

Imaging plays a crucial role in confirming the diagnosis and assessing the extent of cirsoid aneurysms. Doppler ultrasonography provides valuable information regarding the vascularity and flow characteristics of the lesion. Computed tomography (CT) angiography and magnetic resonance imaging (MRI) with angiography offer detailed anatomical visualization and aid in surgical planning. Digital subtraction angiography (DSA) remains the gold standard for delineating the arterial feeders, venous drainage, and precise angioarchitecture of the lesion [5].

Management Strategies:

The management options of cirsoid aneurysms of the cheek involves surgical excision, embolization, or a combination thereof. Surgical resection aims to remove the abnormal vascular network while preserving facial aesthetics and function. Embolization techniques, such as coil embolization or sclerotherapy, offer minimally invasive alternatives for reducing blood flow to the lesion, particularly in cases unsuitable for surgical intervention [6].

Case Illustration:

A 44-year-old male presented with a slowly progressively enlarging pulsatile mass on his right cheek, which had been present since long time. He is being operated once for same complaints, elsewhere many years back, for which no details were available. Clinical examination revealed a bluish, non-tender swelling with palpable thrill and audible bruit. CT angiography confirmed the presence of a cirsoid aneurysm involving branches of the facial artery. The main feeder for the aneurysm being Superficial Temporal artery and drainage to Internal Jugular vein. Due to financial constraints and confirmation on CT angiogram, further investigations related to lesion were not performed. The patient underwent successful surgical resection with meticulous dissection preserving facial nerve, resulting in resolution of symptoms and cosmetic improvement. The skin involved was also excised with the lesion, primary closure of wound was done in the end. Patient was discharged the next day. Histopathologic report suggests- Section reveals lining epidermis with presence of a lesion in the underlying stroma composed of aggregated thick and thin walled vascular structures separated by fibrous tissue. Findings consistent with Arteriovenous hemangioma.

Fig 1 Pre-operative image of lesion

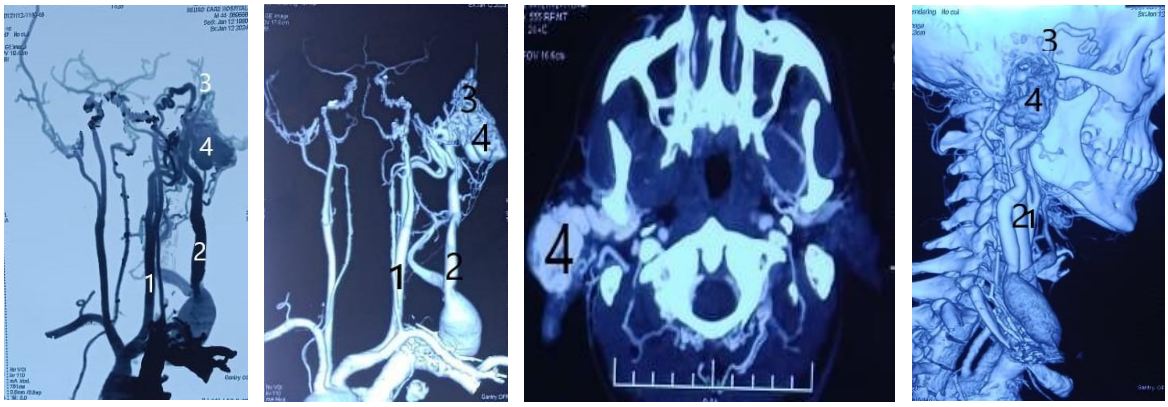

Fig 2 CT Angiography image of lesion (1. Right Common carotid Artery, 2. Internal Jugular Vein, 3. Superficial Temporal Artery, 4. Lesion)

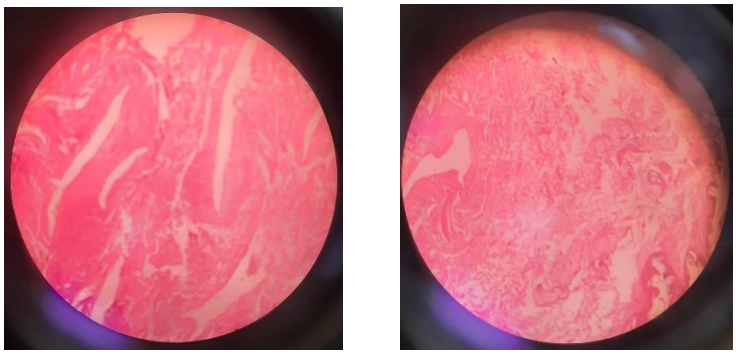

Fig 3 Histopathologic image (magnification 10x) (Microscopic finding- underlying stroma composed of aggregated thick and thin walled vascular structures separated by fibrous tissue. Dilated and fragmented vessel wall seen by Hematoxylin and eosin (H&E) staining)

Conclusion:

A case report of Cirsoid aneurysms of the cheek which are rare vascular anomalies characterized by tortuous, dilated vessels with arteriovenous shunting. The disease is recurrent in the present case report. Despite their rarity, prompt recognition and appropriate management are essential to prevent complications and optimize patient outcomes to prevent further recurrence. A thorough understanding of the clinical features, diagnostic modalities, and treatment options is imperative for clinicians encountering such cases.

References:

1. Chopra, S., Verma, S. K., & Kaur, G. (2018). Cirsoid aneurysm of the scalp: a review. Neurosurgical Reviews, 41(4), 1097-1103.
2. Lexer, E., Bevan, A. D., Lewis, D. D. (1908). General Surgery: A Presentation of the Scientific Principles Upon which the Practice of Modern Surgery is Based. United Kingdom: Appleton.
3. LeBoit, P. E. (2006). Pathology and Genetics of Skin Tumours. Switzerland: IARC Press.
4. Lim, Y. C., Kim, H. J., & Lee, J. W. (2016). Cirsoid aneurysm of the buccal region. Journal of Craniofacial Surgery, 27(4), e337-e338.
5. Zhao, L., Wu, H., & Xiong, H. (2019). Clinical and radiologic features of cirsoid aneurysms: a report of six cases. Interventional Neuroradiology, 25(6), 631-640.
6. Lasjaunias, P., & Berenstein, A. (2004). Surgical Neuroangiography: Clinical and Endovascular Treatment Aspects in Adults. Berlin: Springer.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form, the patient has given his consent for his images and other clinical information to be reported in the journal. The patient understand that name and initials will not be published and due efforts will be made to conceal identity, but anonymity cannot be guaranteed.

Financial support and sponsorship

Nil.

Conflicts of interest

There are no conflicts of interest.