Avoiding Orbital Blindness in managing epistaxis for a child with Duane’s Syndrome

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Learning Points:

1. In Duane’s Syndrome, the Ophthalmic Artery may not have any contributions from the internal carotid artery (ICA), and may be solely supplied by branches of the Sphenopalatine Artery (SPA) and or internal maxillary artery (IMA).

2. In this condition, ligation or embolisation of the IMA or SPA to manage epistaxis may compromise vision and potentially cause blindness.

3. This is the first case, in our knowledge, of angiographic findings confirmed in Duane’s syndrome, and further studies to confirm the association and/or aetiology may be warranted.

This case describes a 15-year-old girl who sustained a traumatic nasal deformity during childhood, and presented with nasal obstruction. Apart from Duane’s syndrome, an ophthalmological congenital condition that manifests clinically as strabismus, she was otherwise well.

She underwent an external approach septoplasty, where moderate intraoperative bleeding was encountered, but settled with cautery and application of internal and external splints.

After removal of splints at five days, she experienced multiple episodes of large volume bleeds, which necessitated resuscitation, packing and eventual exploration under anaesthesia. Prior to proceeding to sphenopalatine artery (SPA) ligation, the on call surgeon decided to request an angiogram to consider embolisation, given the severity of the epistaxis.

Fig 1. Angiography revealed an anatomical variation where her ophthalmic artery and resultant retinal blush was supplied by branches of the facial artery and via posterior septal branches off the SPA and branches off the middle meningeal artery (MM). There were no contributions from her internal carotid artery (ICA) on either side (right picture), which meant embolisation or ligation of the maxillary artery or contributing branches of the SPA would potentially compromise her vision directly. (STA - Superficial Temporal artery)(Int Max – Internal maxillary artery)

To control the epistaxis, tamponade of the unusual plexus of vessels around the nasal dorsum was performed using splints applied internally and externally. This was left for a period of 10 days before removal. Her nasal obstruction and lateral profile has improved to the patients satisfaction.

This confirms the association with Duane’s syndrome, where the Abducens nerve is classically absent or hypoplastic. Theories postulate a disruption occurring during embryogenesis as a contributory factor but, as far as we know, there have not been any described angiographic findings reporting associated vascular anomalies.

Apart from the clinical implications for nasal surgery and epistaxis management in patients with Duane’s syndrome, this may be important in understanding this condition’s aetiology.

It is important to consider preoperative angiography whilst planning surgery where vascular supply is required to be selectively altered.

References


Ongoing interests in laryngeal cleft and obesity, embryology of adenoids, multilevel airway surgery for OSA in obese children.