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Implication of embolization in residual disease in lateral extension of juvenile nasopharyngeal angiofibroma

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ABSTRACT

A few studies have reported incomplete resection of juvenile nasopharyngeal angiofibroma (JNA) following embolization. The likely effects of embolization, viz. shrinkage/reduced tensile strength may have increased the fragility of tumour, leaving remnants postoperatively particularly across hour-glass constriction of sphenopalatine foramina. This paper describes the likelihood of residual disease being left following embolization and hence the importance of extended surgery for complete excision.

The pure endoscopic resection of juvenile nasopharyngeal angiofibroma (JNA) following preoperative selective arterial embolization (PSAE) continues to be the treatment of choice, but some debate has existed regarding its indication for advanced disease.[1] There are several reports[2,3] regarding the risk of incomplete resection following PSAE. Furthermore some authors[4,5] have also suggested a suboptimal degree of tumour excision following embolization, particularly for lesions that exhibit deep sphenoid invasion, potentially leading to increased recurrence. With insufficient evidence the consensus against the advantages of PSAE has not found much gravity whereas the benefits of embolization clearly outweigh the small risk. It cannot be denied that a higher recurrence (based on some other factor such as embolization) in extensive tumours is potentially confounded by the fact that large tumours have a higher recurrence rate because of their size. Although this case seems to suggest a guarded role of PSAE for open surgery of JNA in selected cases, the report is intended to highlight the importance of an 'extended' excision in these selected instances where PSAE may create a situation that may predispose to a residual disease. This may suggest an explanation for the controversial reports against PSAE.

1. Case report

A 9 yr male was admitted with a pinkish nasal mass where imaging suggested JNA. The sphenopalatine foramen (SPFr) and sphenopalatine fossa (SPFs) were completely involved and tumour extended laterally into infratemporal fossa (ITF) through pterygomaxillary fissure (PMF). In addition the cancellous bone of pterygoid-base was eroded (Figs. 1 and 2) with involvement of vidian canal, posterior ethmoid/sphenoid sinuses.

The angiography revealed a tumour blush more towards lateral than posterior-superior extension (Fig. 3). Furthermore the inset of Fig. 3 reveals a lesser blush superiorly (cancellous bone of pterygoid base) than more inferior location.

An endoscopic assisted open excision was undertaken within 24 h following PSAE. The medial/posterior wall of the maxillary sinus was removed and stalk of tumour in PMF was exposed. The posterosuperior attachment of tumour was dissected and periosteum of the orbital floor was exposed. Some difficulty was encountered while delivering the lateral-most attachment from ITF. A small transpalatal incision was simultaneously undertaken to facilitate the final delivery of entire tumour. The total bleeding was < 60 ml. Subsequent endoscopy excluded any residual disease. The histopathology confirmed JNA.

A recurrence 5 weeks after surgery was seen in lateral aspect of nasal cavity around SPFr. The patient being reluctant for immediate surgery unfortunately returned after another 1.5 months with additional snoring/proptosis. The imaging revealed recurrence on right side involving the SPFs, PMF, ITF, orbit, posterior paranasal sinuses, basisphenoid including the right pterygoid base (Fig. 1 inset). The angiography revealed prominence of right IMA with no feeders from internal carotid system.

The surgery was further delayed for 6 weeks during which palatal bulge became evident. Accordingly a combined anteroinferior approach was undertaken under hypotensive anaesthesia with transpalatal incision extending across retromolar area, sublabial region, further splitting the lip, across the ala and extending superiorly as lateral rhinotomy and then laterally as Weber Furgeuson extension. Anterolateral wall of maxillary sinus was drilled and pterygoid muscles excised to completely approach the tumour (Fig. 4). The tumour was excised in 2 parts, the

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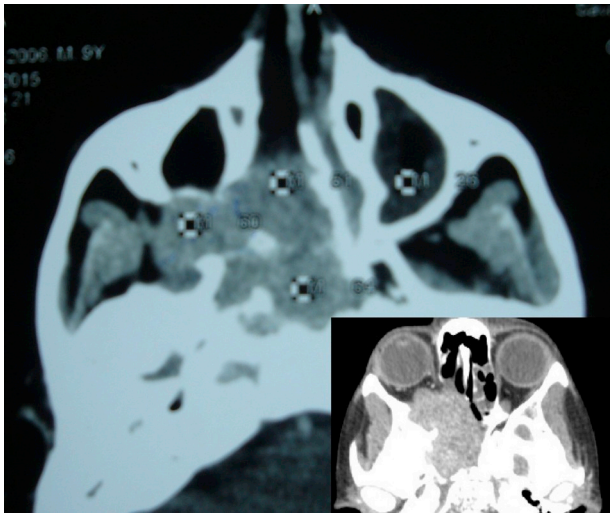

Fig. 1. CT Scan (axial cut) showing widened sphenopalatine foramen, complete involvement of sphenopalatine fossa and the tumour extending laterally into the infratemporal fossa through pterygomaxillary fissure. The cancellous bone of basisphenoid including base of pterygoid is also eroded. The inset CT Scan (contrast enhanced, axial cut) showing a large recurrence on the right side involving the nasal cavity, posterior ethmoids, sphenoid sinus, orbit and basisphenoid including pterygoid base on the right side.

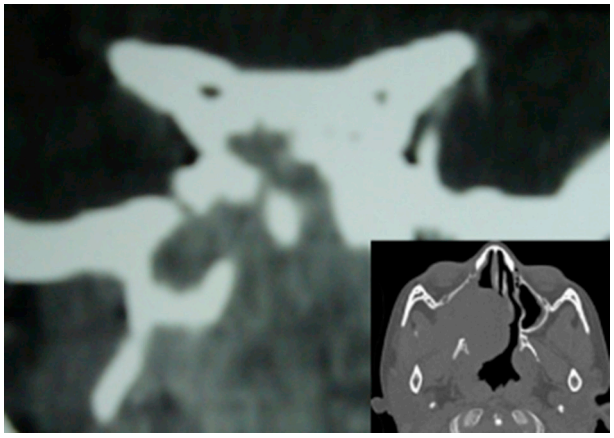

Fig. 2. CT scan coronal and axial (inset) cuts (bone window) showing erosion of cancellous bone of pterygoid-base with involvement of vidian canal, posterior ethmoid/sphenoid sinuses and lateral extension as per Fig. 1.

first being nasal/nasopharyngeal component while the second lateral part from SPFs, ITF and orbit under endoscopic assistance. Subsequent immediate endoscopy revealed residual mass in superior oblique fissure that was carefully extracted and the pterygoid-base was drilled to remove the bleeding extensions. The tumour volume was 50 cc while blood loss < 650 ml. An enhanced CT after 2 weeks and endoscopy after 2.5 m revealed no disease.

2. Discussion

This case revealed extensions into ITF and the pterygoid-base. An endoscopically assisted open transpalatal excision following PSAE was sincerely attempted but unfortunately resulted in large recurrence within 5 weeks. The PSAE may have resulted in shrinkage of tumour with reduced tensile strength. This may have increased the fragility of tumour, leaving some remnants postoperatively particularly across hour-glass constriction of SPFr. A wide open approach without effective embolization in the second sitting achieved complete resection. It seems that PSAE despite reducing blood loss and facilitating intraoperative

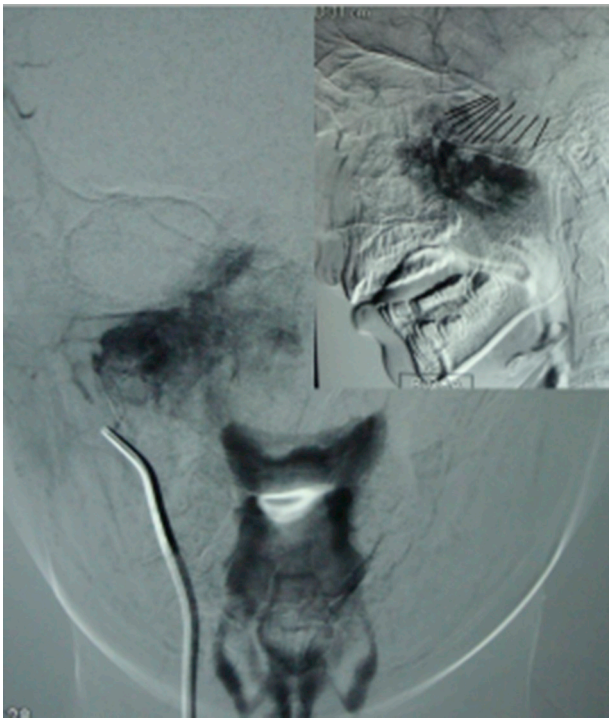

Fig. 3. The angiography showing a tumour blush more towards the lateral aspect than the posterior-superior extension. The inset angiogram showing a lesser tumour blush superiorly across the cancellous bone of the basiphenoid including the pterygoid base as compared to more inferior location. The inferior edges of the marker lines denote the lower boarder of basisphenoid which does not show any significant blush as compared to more inferior component.

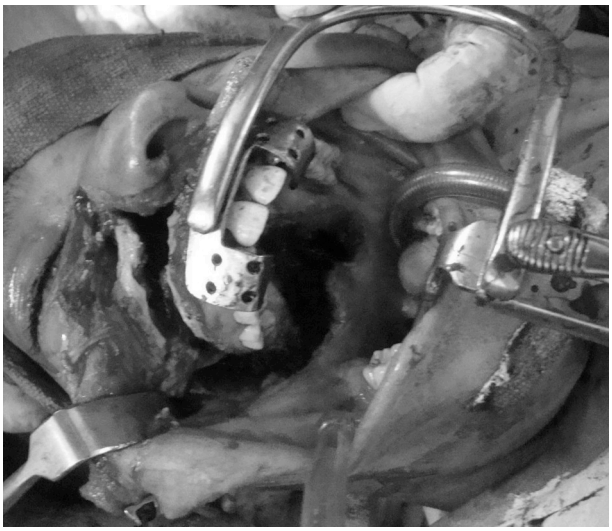

Fig. 4. A wide surgical exposure through transpalatal incision extending across retromolar area, sublabial region, further splitting the lip, across the ala and extending superiorly as lateral rhinotomy and then laterally as Weber Furgeuson extension. Anterolateral wall of maxillary sinus was drilled and pterygoid muscles excised to completely approach the tumour.

visualization may have also predisposed to incomplete resection. It may not be absolutely incorrect to hypothesize that marked lateral extension can have reduced recurrence without PSAE. However with the current evidence it seems more appropriate to emphasize that while excising the extreme lateral component following PSAE, a more complete/extended dissection is needed.

The largest systematic review[6] till date suggests a decreased

recurrence rate for early stage JNA (with pure endoscopic techniques following PSAE) than for larger and more complex tumours (with endoscopic assistance). Many authors have also questioned the usefulness of PSAE[7,8] in terms of significant reduction in blood loss, but we strongly disagree. Our facility has witnessed a reduced intraoperative blood loss following embolization and also demonstrated[9] even better results in terms of recurrence with hypotensive anaesthesia. Recently a low recurrence rate with transnasal endoscopy of stage IIB and lower stage lesions has been reported without embolization.[3] Overall the growing evidence[10] supports the equivalence of endoscopic and open surgical approaches in terms of JNA recurrence.

This was a large stage [IIC (Radkowski); IIIA (Mishra)] tumour invading pterygoid base and extending into ITF. Despite PSAE, endoscopic manipulation and open resection, an unexpected early recurrence was seen. In situations like this it is possible that PSAE can reduce the vascularity and shrink the tumour, thereby providing advantage in terms of identification of feeding vessels and residual disease by endoscopy. However this shrinkage may increase the fragility particularly of finger like projections of tumour across cancellous bone that may break during traction. A deep invasion of cancellous bone (around vidian canal) is an accepted site of incomplete surgery.[2] Similarly we feel that the hour-glass constriction across SPFr may also weaken, resulting in tear in that bottle-neck area. The residual devascularized masses after detachment may either retract or become too inconspicuous to be recognised as a residue. JNA is also hypothesized to arise as an expression of incomplete regression of first brachial artery, or from paraganglionic cells of terminal maxillary artery (IMA). Hence detachment of fragile tumour may leave some residue at the site of origin (terminal end of IMA). Such shrunken residues along with occluded artery may be camouflaged by the surrounding structures. Moreover residual tissue may be devoid of collateral vascularity initially and hence does not bleed to be recognised during surgery. Hence embolization on one hand may definitely reduce intraoperative bleeding and facilitate dissection but on the other hand may also contribute to incomplete resection. In accordance we propose a meticulous extended look for the residual disease in such instances particularly the lateral extension. This may in fact have been the reason for conflicting reports in the literature regarding non-effectivity of PSAE in terms of recurrence.

Involvement of 2 experienced surgeons reduces a possibility of a gross disease being left intentionally but however an early recurrence suggests a probable residual disease. The rapidity of growth reflects both an aggressive biology as well as a sizable residue that was large enough for spontaneous involution or necrosis. It is more likely that ITF would have been the preferred site for this residue rather than pterygoid-base. This is further suggested by enhancement (angiography) more across ITF and some operative struggle in delivering the lateralmost extension. As recurrence was seen first in the lateral wall of nose around the SPFr, it is reasonable to assume IMA (in closest proximity) as main feeder. This was also further evident by subsequent angiography. Hence the lateral–stalk that was laterally retracted in ITF or even lost after detachment is expected to be more aggressive than pterygoid-base component (which presented later). After tumour delivery (second surgery) the endoscopy revealed residual disease around superior oblique fissure (SOF) whereas no ‘shrunken’ residues could be appreciated after the first surgery. A large JNA after open excision following PSAE may not reveal any immediate postoperative bleeding and hence create a false impression of complete resection despite presence of devascularized residues. Therefore the propensity of residual being left in foramen such as SOF, SPFr and PMF just as in the vidian canal need further evaluation particularly after PSAE.

The vascular supply in the initial stages of JNA is more predictable with ipsilateral IMA as the main feeding vessel. As the tumour enlarges, other arteries participate sometimes including internal carotid system with intracranial/superior orbital fissure extensions. Hence the effective embolization of large tumour becomes more difficult and somewhat risky considering the possibility of accidental embolization of intracranial collateral. A similar situation may be expected in recurrence following resection of advanced JNA, as the previously functional collateral channels in the original tumour may get re-canalized early in such residual tumours. Hence with theoretical presence of more collaterals, a recurrent disease may pose more difficulty in embolization as compared to a similar sized upfront disease. Importantly the collateral branches of IMA can be missed during routine carotid angiography, a situation that is likely to arise with ITF extension.

To conclude our experience shows that open approach under hypotension but without PSAE for lateral extensions of JNA has three main advantages: (1) gross appearance of ‘bloody’ tumour is better recognised than a pale residue; (2) better tensile strength of vascular tumour would facilitate traction in comparison to devascularized segment that is likely to be torn due to enhanced fragility; (3) detached component may constantly bleed and hence less likely to be camouflaged by surroundings. In addition to the limited current evidence we strongly feel that an extreme lateral extension of JNA has a high recurrence potential particularly when tackled by pure endoscopic approach following PSAE. Accordingly this site also needs an extended overdoing of the resection just like bone drilling for vidian canal involvement. However while it might be true that visualizing the vascularity of the lesion (versus blanching) could encourage more-complete resection, it is also true that lack of embolization prolongs operative time and increases bleeding and more bleeding can further make complete resection more difficult. Anatomic and intrinsic factors such as local extension and growth rate are clearly very important factors predicting recurrence, and any small series cannot distinguish all these other factors. We fully agree that our experience based on a single such case cannot be generalized and hence more of such studies are required to enumerate the ‘precautions’ needed following PSAE to reduce recurrence and hence resolve the reasons suggested for the demerits of embolization.

Financial disclosure

None.

Conflicts of interest

None to be declared.

Appendix A. Supplementary data

Supplementary data to this article can be found online at https://doi.org/10.1016/j.jobcr.2018.12.001.

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