Extranasopharyngeal Angiofibroma with Lateral Extension to the Buccal Region

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Abstract
A case of extranasopharyngeal fibroma confined to the oral cavity in a 14-year-old boy is reported.

Key words: Angiofibroma, Mouth

Introduction
Juvenile nasopharyngeal angiofibroma (JNA) is an uncommon fibrovascular tumour, which is locally aggressive. It typically occurs in adolescent boys and is associated with nasal obstruction and epistaxis.1 The tumour usually arises from the nasopharynx in the region of the sphenopalatine foramen. This report presents an extranasopharyngeal angiofibroma that was confined to the buccal and subtemporal region.

Case Report
A 14-year-old boy was referred for evaluation and management of a left facial swelling (Figure 1). He had noticed a swelling in the left buccal region 6 months earlier that was firm and non-tender. There was no history of epistaxis or nasal obstruction.

Computed tomography scan showed an intensely enhancing, well-defined mass in the subtemporal and buccal region (Figure 2). The lateral maxillary sinus wall was displaced anteromedially without erosion. Displacement of adjacent tissue planes was noted (Figure 3).

The mass extended from the buccal region to the subtemporal region and further superiorly. Resection was planned and a transoral approach was employed with a contingency to extend the incision to a Weber-Ferguson. The lesion was encapsulated and was resectable through the transoral approach alone. The specimen was oval, soft, and measured $6.0 \times 5.0 \times 4.5$ cm$^3$. Histological examination revealed an encapsulated tumour composed of numerous vessels and dense collagenous stroma. The vessels were thin walled, lacking elastic fibers, had absent or incomplete smooth musculature, and varied in appearance from stel late or staghorn to barely conspicuous due to stromal compression. Stromal cells had plump nuclei and tended to radiate around the vessels. An abundance of mast cells in the stroma and a lack of other inflammatory cells were observed. Localised degeneration was also observed in the stroma.

Figure 1. Swelling in the left buccal region.

Figure 2. Computed tomography scan showing an intensely enhancing, well-defined mass in the subtemporal and buccal region.
The diagnosis was extranasopharyngeal angiofibroma with complete excision.

Physical and computed tomography examination 6 months after operation showed no evidence of recurrence. Further follow-up and monitoring are planned.

Discussion

JNA is a benign non-encapsulated fibrovascular tumour that usually originates from the posterolateral wall of the nasal cavity, where the sphenoidal process of the palatine bone meets the horizontal ala of the vomer and the root of the pterygoid process of the sphenoid. The tumour usually extends medially from the sphenopalatine foramen and pterygopalatine fossa to lie within the nasal cavity. Rarely, the lesion extends laterally into the infratemporal and buccal region. Only a single case of JNA extending into the cheek has been reported earlier. In this case, the lesion extended to the cheek without any common symptoms, such as epistaxis or nasal obstruction. The histogenesis and pathogenesis of JNA are unclear. Popular theories include abnormal growth of embryonic chondrocartilage, testosterone acting on a hamartomatous nidus of inferior turbinate tissue misplaced into the nasopharynx, and tumour growth from normal nasopharyngeal fibrovascular stroma. Other suggested etiologies include trauma, inflammation, infection, allergy, and heredity.

The average age at onset of symptoms is 14 to 18 years, depending on the series quoted. A typical age range is between 7 and 21 years. Patients are almost always male.

The 2 primary therapeutic modalities for JNA have been surgery and radiotherapy over the years. Several adjunctive measures have been tried, including embolisation, hormonal therapy, and chemotherapy. The main concerns with radiotherapy, which is delivered by external beam, are malignancies in the irradiated field and the danger of inhibiting normal facial growth in younger patients. Cummings et al found that, although symptomatic relief is fast, regression of the tumour with radiation is slow. In his study, 50% of patients treated with primary radiotherapy had visible tumour in the nasopharynx or nasal cavity 12 months after treatment. Fifty percent of patients who had visible tumour at 2 years suffered eventual recurrence. Many surgical approaches have been used against juvenile angiofibroma. In this case, we used the intraoral approach and succeeded in complete excision, although we were prepared to extend the incision if called for.

References