

Case Report

Pediatric Traumatic Neuroma of the Upper Lip - A Case Report

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ABSTRACT:

Traumatic neuroma (TN), represents a reactive proliferation of neural tissue following damage to an adjacent nerve. Clinically, patients usually present with inimitable pain, oral lesions appearing as nodule of size less than 2 cm with normal or greyish white smooth surface mostly involving the mental foramen, tongue or the lower lip. We report a rare case of an upper lip traumatic neuroma, in an uncommon age group and uncommon location, which was clinically thought to be hemangioma. Histopathologically, excision biopsy specimen showed haphazard proliferation of fascicles of nerve fibres. Immunohistochemical analysis revealed strong cytoplasmic positivity for S100 and was negative for SMA and CD34, affirming the neural origin of the lesion. TN could be considered as a differential in cases of soft tissue swelling from uncommon location in the oral region especially in the pediatric age group, where a clear trauma history cannot always be illustrated.

Key words: Traumatic neuroma, upper lip

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Background-

Traumatic neuroma is a hyperplastic lesion and are not true neoplasms. It represents a reactive proliferation of neural tissue following damage to an adjacent nerve [1]. It may occur in any part of the body, including the head, neck, gallbladder and thigh [2]. In the oral region, though being a rare disorder, the lesion appears clinically as a nodule of size less than 2 cm with normal or greyish white smooth surface and has a predilection for the mental foramen region, tongue or lower lip and intra-osseous areas. It is extremely rare in the upper lip. Though TN can occur in a wide age range, they are typically diagnosed in middle-aged women who mainly present with pain, tenderness over the lesion and paraesthesia (1-4). The recommended treatment for a traumatic neuroma is simple excision rather than nerve resection or alcohol blocks (5). Here, we report a case of an extremely rare traumatic neuroma in an uncommon age group and uncommon location, that was clinically thought to be haemangioma

Case presentation

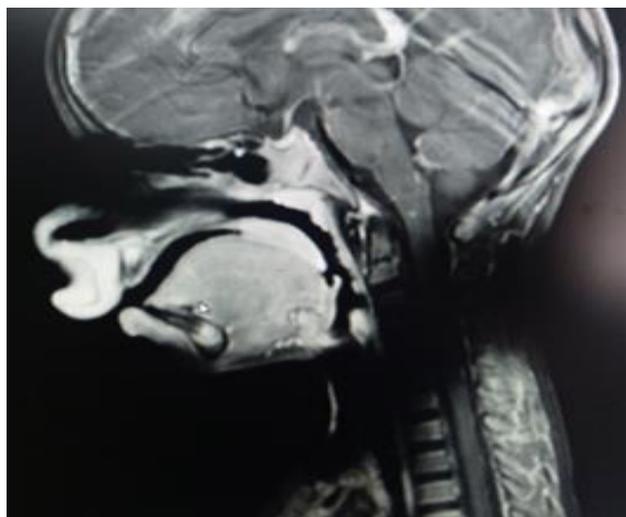
A 7-year-old boy was brought by his parents with complaints of an unesthetic looking swelling in the upper lip, which had developed incidentally and had been present for approximately 2 years, the swelling did not show any increase in size and was occasionally associated with pain. No history of any noticeable trauma or inflammation involving the head and neck region could be elicited. Past medical and dental history were insignificant. On examination, a swelling of size 2x1 cm was noted in the mucosal aspect of the upper lip. It was tender on palpation and was not associated with any pulsations or bruit.

Investigations

When worked up further, magnetic resonance imaging of face revealed a homogenous enhancing hypointense lesion in T1 weighted images and the swelling exhibited increased signal intensity in T2-weighted magnetic resonance images (Fig 1), suggesting a likely benign lesion, and the radiological differentials included haemangioma, Schwannoma and Epitheloid hemangioendothelioma. In view of absence of any

feeder vessels into the lesion, the possibility of haemangioma was remote.

Fig 1: MRI Lateral view of face showing hypointense lesion in the upper lip.

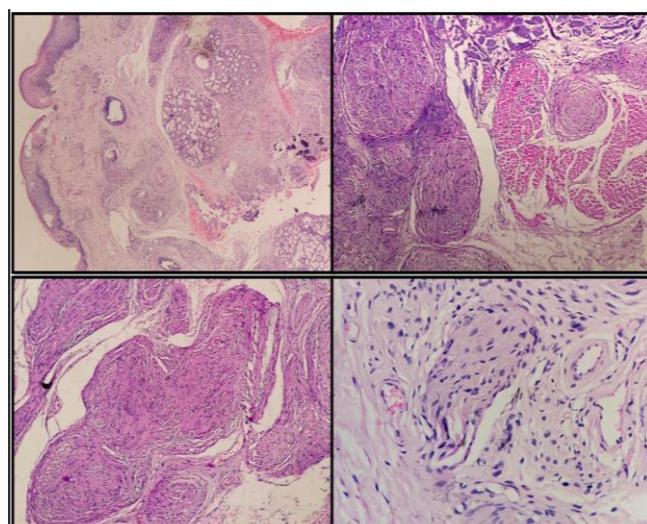


Based on the clinical and radiographic examinations, the initial diagnosis was a soft tissue tumour probably a haemangioma, patient was taken up for excision biopsy. The patient underwent complete resection of the tumour with an additional clearance of margin. Resected specimen was then sent for histopathological examination.

Grossly, it was a grey brown soft tissue mass measuring 2x0.8x0.5 cm with cut surface showing grey white to grey brown areas. All of the tissue was embedded and processed. Microscopically,

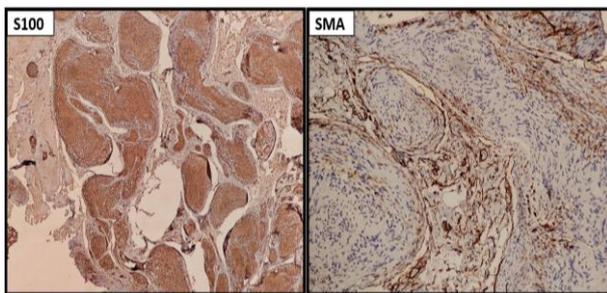
the sections revealed fragments of tissue lined by stratified squamous epithelium with the underlying stroma showing mucous glands (Fig - 2A), surrounded by haphazard proliferation of nerve fascicles including axons, Schwann cells and fibroblasts embedded in a collagenous stroma (Fig-2B,C,D). Focal areas of haemorrhage along with few congested vessels were also seen. No evidence of atypia or malignancy noted.

Figure 2 A- H& E, 4X, stroma showing mucous glands and haphazard proliferation of nerve fibres. Figure 2 B and C- H& E, 10X, haphazard proliferation of nerve fibres in fascicles along with Schwann cells and fibroblast. Figure 2 D – H&E,40X, High power view of nerve fibre arranged in fascicle.



Further immunohistochemical analysis revealed strong cytoplasmic positivity for the neural marker S-100(Fig-3A) and was negative for SMA (Fig-3B) and CD34, which affirmed the neural origin of the lesion. Based on these findings, the final diagnosis was given as traumatic neuroma.

Figure 3 A - IHC,4X, Positive for S100, Figure 3 B -IHC,10X, Negative for SMA



DISCUSSION

Proliferative lesions of the peripheral nerves are grouped as non-neoplastic (traumatic neuroma), benign lesions (schwannoma, neurofibromas and perineuriomas) and malignant lesions collectively designated as malignant peripheral nerve sheath tumours. (8,9)

Among the neuromas large majority follow trauma and termed as traumatic neuromas. They occur after transection or damage to a nerve

bundle. After a nerve has been crushed or transected due to trauma or surgery, the proximal portion attempts to regenerate and re-establish innervation to the distal segment which underwent Wallerian degeneration. This happens via the growth of axons through tubes of proliferating Schwann cells. Aberrant repair occurs when the regenerative tissues encounter a scar or cannot re-establish innervation (1-3, 8-9). Thus, a tumour-like mass may develop at the site of injury.

On an average, 25–33% of oral TNs, present with intermittent or constant pain and the nature of pain mostly reported as burning to severe radiating pain, with or without tenderness at the localised site. Inimitable pain, which is a characteristic feature of this lesion is mainly due to the constriction of nerves by stromal fibrosis (3). As per Sist's research oral TN located at the mental foramen were the most painful of all other sites (11). Predominantly asymptomatic patients as in this case, can be explained by the absence of inflammation, as TN with inflammation are most likely to be painful than those without significant inflammation.

Traumatic neuromas occur at any age but most commonly seen in middle aged women. Bimodal occurrence in young and older age group individuals is noted. Trauma is the major attributing factor in young patients with oral region TN, However, in our case, there was no possible history of trauma or surgery incurred by the patient according to his attenders. Upper lip can easily get injured by minimal repetitive mechanical trauma during eating and also by trivial falls during play time especially in the paediatric age group. Oral habits in children like persistent lip sucking are often overlooked and can also lead to injuries of the lips (1,5). Likewise, the elderly are prone to chronic irritation, mechanical destruction due to ill fitted dentures and procedures like dental extractions, surgeries like parotidectomy.

According to study by Jones and Franklin, the frequency of traumatic neuromas in the oral region is 0.34% (8). The most common sites of involvement in the head and neck are the inferior alveolar nerve, lingual nerve and greater auricular nerve. In our case, anterior superior alveolar nerve

(branch of infra orbital branch nerve) involvement is speculated and this is substantiated by upper lip region being specifically innervated by this nerve. Through an extensive review of literature, though a small number of oral region TN, predominantly involving the palate and the lower lip have been documented there has been only one case of upper lip TN involving the anterior superior alveolar nerve recorded till date (3).

Traumatic neuromas are characteristically smooth surfaced, nonulcerated nodules and are generally less than 2 cm in diameter as in the present case. The lesions, especially in the head and neck region, must be differentiated histologically from neurofibroma and schwannoma. Neurofibromas, may be associated with von Recklinghausen disease, usually lacks a capsule, contains mucopolysaccharide ground substance and has fewer axons with myelin sheaths. Neurofibromas show strong positivity for S100 and SOX10, while CD34 shows fingerprint-like positivity in these tumours. Schwannoma usually is present in a subcutaneous location, contains hypercellular (Antoni A) and hypocellular (Antoni B) areas with

Verocay bodies and lacks axons. Likewise, Schwannomas show strong positivity for S100, SOX10 and variable positivity for CD34. Whereas, traumatic neuromas usually contain irregular, haphazard proliferation of nerve fascicles which contain axonal and Schwann cells along with the addition of scarring and inflammatory cells and stain positive for the neural immunohistochemical marker S100.

Patients can be generally reassured that excision is curative, with less chance of recurrence, and there is no indication for further investigations for malignancy. Although the follow-up for this patient was relatively short to monitor for neuroma recurrence, a close long term follow up is always advised.

Learning points

Traumatic neuroma could be considered as a differential diagnosis for soft tissue swelling from uncommon location in the oral region, especially in paediatric population, where a defined cause cannot always be illustrated.

The treatment of choice for such lesions is surgical excision which has a good outcome with very less recurrence rates.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms.

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Conflicts of interest

There are no conflicts of interest

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