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CASE REPORT

Palatal schwannoma in a young female patient: report of a case and review of the literature

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Abstract

Background A few cases of oral schwannomas in paediatric patients have appeared in the literature so far; however, there are no studies focusing solely on paediatric oral schwannomas. The aim of this study is to report a case of palatal schwannoma in a young female patient and review the pertinent literature on oral schwannomas in paediatric patients.

Case report A 12-year-old girl presented with a growing swelling of 2 weeks duration on the hard palate. With a provisional diagnosis of a salivary gland neoplasm an incisional biopsy was performed under local anesthesia and the histologic examination disclosed an Antony A type schwannoma.

Treatment Complete surgical resection of the lesion was performed under local anaesthesia through a palatal mucosa incision and paraffin gauze was sutured on the surgical area for protection during secondary healing. The bone underlying the tumor was normal. The postoperative period was uneventful and 2 weeks after excision the gauze were removed.

Follow-up There have been no signs of recurrence during a 18-month follow up period.

Conclusion A palatal swelling in a child or adolescent may represent a neoplasm, such as a schwannoma and requires careful clinical and radiographic evaluation of the dentition.

Keywords Mouth neoplasms · Palatal neoplasms · Nerve sheath neoplasms · Neurilemmoma · Schwannoma · Child · Adolescent

Background

Schwannoma or neurilemmoma is a benign nerve sheath tumour, originating from the Schwann cells of peripheral nerves (Butler et al. 2016). Although 25–48% of schwannomas manifest in the head and neck area (do Nascimento et al. 2011), they are extremely rare in the oral cavity, representing 9.3% of head and neck schwannomas (Butler et al. 2016) and 0.04% (do Nascimento et al. 2011) to 0.1% (Jones and Franklin 2006a) of oral biopsies.

Oral schwannomas are usually encountered in adults, mostly women [62.5% (Butler et al. 2016) − 65% (Jones and Franklin 2006a)] in the second and third decades of life (Hatziotis and Asprides 1967), with a mean age ranging from 21.6 (Butler et al. 2016) to 48.6 (Jones and Franklin 2006a) years. They may be peripheral or less frequently intraosseous (Buric et al. 2009). Peripheral schwannomas usually involve the tongue (55.7% of cases) followed, in descending order, by the palate, floor of mouth and buccal mucosa (Hatziotis and Asprides 1967). They manifest as slow-growing solitary masses covered by normal mucosa that measure a few millimeters to many centimeters in diameter (Sanchis et al. 2013). Most cases are asymptomatic, but pain is reported in approximately 1/3 of the cases (Butler et al. 2016). Tongue lesions may interfere with speech or swallowing, and less frequently breathing (Hatziotis and Asprides 1967). The lack of symptoms may delay diagnosis (Hatziotis and Asprides 1967; Sanchis et al. 2013). Intraosseous schwannomas usually arise in the posterior mandible and present as unilocular or multilocular radiolucencies with or without well-defined borders. They cause swelling in more than half of the cases, whereas pain or paresthesia are rare (Buric et al. 2009).

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Histopathologically, conventional schwannomas are usually encapsulated tumors (Butler et al. 2016) that show a proliferation of S-100 protein positive Schwann cells that are organized in two different architectural tissue patterns (Bouquot et al. 2009): the most common Antoni A, characterized by fascicles of spindle-shaped cells streaming around acellular eosinophilic areas known as Verocay bodies, and the less cellular Antoni B, where spindle cells are randomly arranged in a loose myxomatous stroma (Bouquot et al. 2009). Histologic variants include ancient schwannoma, in which degenerative changes in long-standing tumors, such as haemorrhage, nuclear atypia, haemosiderin deposits, inflammation and fibrosis, may lead to a misdiagnosis of a malignant neural tumor or sarcoma (Humber et al. 2011); plexiform schwannoma, i.e. schwannoma with a multinodular, plexiform pattern, occasionally associated with neurofibromatosis type II (NF2) or schwannomatosis (Berg et al. 2008); epithelioid or melanotic schwannoma; schwannomas with glandular elements; and neuroblastoma-like schwannomas (Sedassari et al. 2014).

Surgical excision is the treatment of choice both for peripheral and intraosseous schwannoma and the rate of recurrence or malignant transformation is low (Hatziotis and Asprides 1967; Buric et al. 2009).

The aim of the present study is to report a rare case of palatal schwannoma in a 12-year-old girl and review the pertinent literature on oral schwannoma in the paediatric population.

Case report

A 12-year-old girl was referred to the Department of Oral Medicine and Pathology for evaluation of a painless swelling on the palate. The patient reported that the swelling appeared abruptly 2 weeks before presentation. There was no history of local trauma or infection, and no altered sensation in the area. Her medical and family history were unremarkable, as well as the results of a recent review of systems and a physical examination.

Clinical examination showed a tumor measuring 2.5 × 2 cm on the left side of the hard palate in close proximity to the apices of the adjacent second premolar to the second molar teeth (Fig. 1). The tumor was soft and painless on palpation, covered by normal mucosa with a slightly erythematous hue in the center and adherent to the overlying mucosa. The remaining tissue of the oral mucosa was within normal limits. The patient had a healthy permanent dentition and good oral hygiene and all adjacent teeth showed a normal response to pulp tests. A panoramic radiograph did not show any bone abnormality (Fig. 2) and a maxillary cone beam CT revealed thinning of the cortical palatal bone underlying the tumor, attributed to pressure (Fig. 3). With a provisional diagnosis of a salivary gland neoplasm, the patient was referred to the Department of Oral and Maxillofacial Surgery and an incisional biopsy was performed under local analgesia.

Microscopic examination of 5 μm-thick, formalin-fixed, paraffin-embedded tissue sections stained with hematoxylin-eosin revealed a well encapsulated tumor exhibiting areas with spindle or oval shaped cells on a fibrous connective tissue stroma (Fig. 4). The cells showed palisading arrangement around eosinophilic areas, consistent with Verocay bodies and reacted immunohistochemically with S-100 protein. The diagnosis of Antoni type A schwannoma was

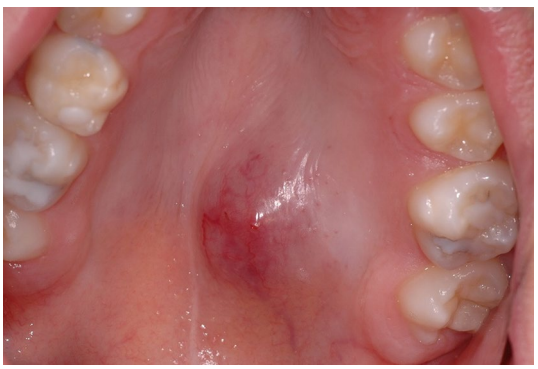

Fig. 1 Swelling on the left side of the hard palate in close proximity to the apices of the second premolar to second molar teeth

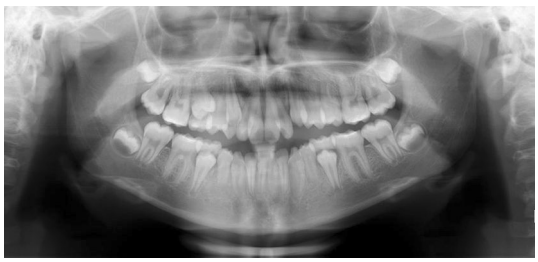

Fig. 2 Panoramic radiograph. There is no bone abnormality or obvious pathology of odontogenic origin

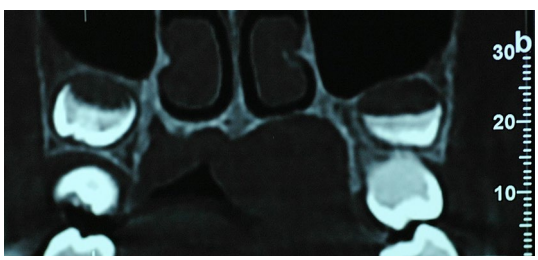

Fig. 3 CBCT-coronal view. Thinning of the cortical palatal bone underlying the tumor

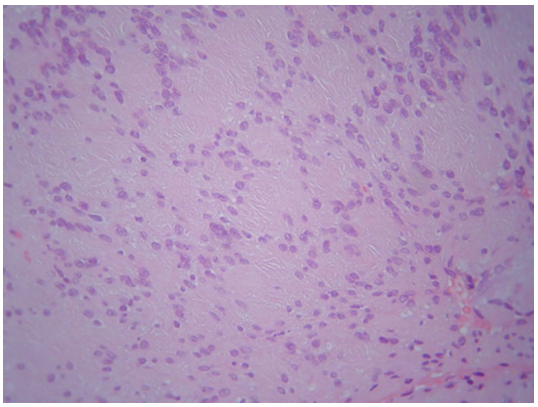

Fig. 4 Spindle or oval shaped cells in a fibrous connective tissue stroma (hematoxylin-eosin stain, original magnification ×100)

rendered. The patient did not fulfill the diagnostic criteria for either NF2 or schwannomatosis.

Treatment

Complete surgical resection of the lesion was performed under local anaesthesia through a palatal mucosa incision and paraffin gauze was sutured on the surgical area for protection during secondary healing. The bone underlying the tumor was normal. The postoperative period was uneventful and 2 weeks after excision the gauze were removed.

Follow-up

There have been no signs of recurrence during a 18-month follow up period.

Discussion

Oral schwannomas are rare in children and adolescents. Shah et al. (2009) reported 3 cases (0.05%) among 5.457 oral biopsies in patients 0- to 16-year-old; Sklavounou-Andrikopoulou et al. (2005) 2 cases (0.19%) among 1.040 soft tissue biopsies over a 32-year period in patients up to 18-year-old; Dhanuthai et al. (2007) 3 cases (0.24%) over a 15-year period in patients up to 16-year-old; and Jones and Franklin (2006b) 5 cases (0.11%) over a 30-year period amongst 4.406 oral biopsies in patients up to 16-year-old.

A review of the pertinent English literature revealed 86 previously published case reports. These are listed in the Reference List Appendix.

The main clinical features of those cases and the present one are summarised in Table 1. There is no gender predilection, as 42 patients (50%) were males and 42 (50%) females (male to female ratio 1:1). In contrast, oral schwannomas as a whole are more common in females (Jones and Franklin 2006a; Butler et al. 2016). The youngest patient was 6-year-old and the mean age of the patients was 14 ± 3.4 years (median 14 years). Approximately 1/3 (34.5%) of the patients were in the 6–12 years age group and 2/3 (65.5%) in the 13–18 years old age group. There was no significant difference in the mean age between males (14.2 ± 3.4 years, median 15 years) and females (13.9 ± 3.5 years, median 14 years).

The most common site of occurrence was the tongue in 57.5% of the cases, followed by the lips (10.3%), the hard palate (8%) and the buccal mucosa (6.9%). The tongue is the most common site involved, regardless of age (Hatziotis and Asprides 1967).

Clinically, they usually presented as solitary asymptomatic tumors of normal colour (86.3%) and surface (78.8%) that were elastic-firm on palpation (78.6%). Maximum diameter ranged from 0.5 to 6.5 cm (mean 2.1 ± 1.1 cm, median 2 cm). In 47 cases (73.4%) no symptoms were reported, 9 cases (14.1%) were described as painful, while in another 8 cases (12.5%) difficulty in swallowing, speaking or breathing, bleeding, macroglossia, or loss of sensation and taste were reported. Similar symptoms have been reported in non-paediatric oral schwannomas (Hatziotis and Asprides 1967; Cohen and Wang 2009; Sanchis et al. 2013). The lack of symptoms justifies the long duration of 23.1 ± 38.2 months (range 10 days to 16 years, median 10 months) before diagnosis.

The differential diagnosis of a paediatric oral schwannoma may include other benign lesions that are common in this age group, such as an irritation fibroma, pyogenic granuloma, neurofibroma or vascular tumors (Iatrou et al. 2013; Pinto et al. 2014). For palatal tumors as in the case presented herein, an odontogenic abscess should be considered and therefore pulp tests and radiographic examination should be performed (Dhanuthai et al. 2009). Other lesions included are salivary gland tumors, such as pleomorphic adenoma (Dhanuthai et al. 2009) and mucoepidermoid carcinoma (Baumgardt et al. 2014) that are less common in children compared to adults; soft tissue tumors, such as neurofibroma (Leduc et al. 2017), myofibroma (Vered et al. 2007) and leiomyoma (Brooks et al. 2002); and non-Hodgkin lymphoma (Epstein et al. 2001).

Microscopic examination is the only accurate and reliable mean to establish a final diagnosis. Histologically, six cases (6.9%) of oral paediatric schwannoma were described as plexiform (Heifetz et al. 1991; Di Giovanni et al. 2006; Berg et al. 2008; Lobo et al. 2009; Santos et al. 2010) and two cases (2.3%) as ancient (Subhashraj et al. 2009; Amirchaghm et al. 2010) with the remaining of the cases (90.8%) being conventional schwannomas. Immunohistochemistry

Table 1 Clinical and demographic data of 86 previously published cases of oral schwannoma in paediatric patients plus the case of the present study

- Reported cases: 87
  - Gender (data from 84 cases)
    - Male: 42 (50%)
    - Female: 42 (50%)
  - Male:female ratio
    - 1:1
  - Age (data from 87 cases)
    - Range: 6–18 years
    - Mean: 14 ± 3.4 years
    - Median: 14 years
    - Age related to gender
      - Male patients
        - Range: 7–18 years
        - Mean: 14.2 ± 3.7 years
        - Median: 15 years
      - Female patients
        - Range: 6–18 years
        - Mean: 13.9 ± 3.5 years
        - Median: 14 years
    - Age categories and gender
      - Infant (1–23 months): 0
      - Pre-school child (2–5 years): 0
      - Child (6–12 years): 15 male/14 female (34.5%)
      - Adolescent (13–18 years): 27 male/28 female (65.5%)
  - Site (data from 87 cases)
    - Tongue: 50 cases (57.5%)
      - Lateral tongue: 10 cases
      - Dorsal tongue: 6 cases
      - Posterior tongue: 5 cases
      - Ventral tongue: 5 cases
      - Anterior tongue: 4 cases
      - Base of tongue: 3 cases
      - Tip of tongue: 2 cases
      - Not specified: 15 cases
    - Lip: 9 cases (10.3%)
      - Upper lip: 4 cases
      - Lower lip: 4 cases
      - Not specified: 1 case
    - Hard palate: 7 cases (8%)
    - Buccal mucosa: 6 cases (6.9%)
    - Floor of mouth: 4 cases (4.6%)
    - Vestibule: 4 cases (4.6%)
      - Mandibular vestibule: 3 cases
      - Not specified: 1 cases
    - Soft palate: 3 cases (3.4%)
    - Hard and soft palate: 2 cases (2.3%)
    - Gingiva: 2 cases (2.3%)
      - Upper anterior gingiva: 1 case
      - Lower anterior gingiva: 1 case
  - Clinical presentation (data from 60 cases)
    - Mass: 34 cases (56.7%)
    - Swelling: 14 cases (23.3%)
    - Nodule: 8 cases (13.3%)
    - Lobulated mass: 3 cases (5%)
    - Multiple masses: 1 cases (1.7%)
  - Colour (data from 22 cases)
    - Normal: 18 cases (81.8%)
    - Yellow: 2 cases (9.1%)
    - Purple: 1 case (4.5%)
    - Normal-bluish: 1 case (4.5%)
  - Consistency (data from 28 cases)
    - Elastic: 12 cases (42.9%)
    - Firm: 10 cases (35.7%)
    - Semielastic: 2 cases (7.1%)
    - Fluctuant: 1 case (3.6%)
    - Hard: 1 case (3.6%)
    - Normal: 1 case (3.6%)
    - Soft and friable: 1 case (3.6%)
  - Surface (data from 33 cases)
    - Normal: 14 cases (42.4%)
    - Smooth: 12 cases (36.4%)
    - Ulcerated: 4 cases (12.1%)
    - Papillary: 3 cases (9.1%)
  - Size (largest diameter in centimeters) (data from 62 cases)
    - Range: 0.5–6.5 cm
    - Mean: 2.1 ± 1.1 cm
    - Median: 2 cm
  - Symptoms (sata from 64 cases)
    - No: 47 cases (73.4%)
    - Yes (with or without other symptoms (difficulty in shallowing, speaking)): 9 cases (14.1%)
    - Other symptoms (difficulty in shallowing, speaking or breathing, bleeding, macroglossia, loss of sensation and taste): 8 cases (12.5%)
  - Duration (data from 59 cases)
    - Range: 10–16 years
    - Mean: 23.1 ± 38.2 months
    - Median: 10 months
  - Post-operative course
    - Follow-up (data from 34 cases)
      - Range: 3 months to 14 years
      - Mean: 2.5 ± 3.2 years
      - Median: 1 year
    - Recurrence (data from 44 cases)
      - No: 41 cases (93.2%)
      - Yes: 3 cases (6.8%)
      - Mean time of recurrence: 17 ± 13.5 months
      - Median time of recurrence: 9 months

is not necessary for final diagnosis. The tumours were positive for S-100 protein and vimentin, showed variable positivity for glial fibrillary acidic protein (GFAP), epithelial membrane antigen (EMA) and Leu-7, and were negative for HHF-35 and alpha smooth muscle actin (a-SMA).

The histopathologic differential diagnosis of conventional schwannoma includes neurofibroma, palisaded encapsulated neuroma, traumatic neuroma, and leiomyoma. Neurofibroma in not encapsulated, Schwann cells are arranged in hypocellular sheets, Verocay bodies are not present, and usually has mucoid stroma and many mast cells (Koutlas and Scheithauer 2010). Palisaded encapsulated neuroma shows Schwann cells organized in cellular microfascicles coursing in various directions, Verocay bodies are uncommon and the stroma is collagenous with occasional myxoid areas in the periphery (Koutlas and Scheithauer 2010). In addition, the cells do not react for GFAP (Koutlas and Scheithauer 2010). Traumatic neuroma is not encapsulated and consists of numerous intertwining or haphazardly arranged nerve fibers in a mucoid to fibrocollagenous matrix (Bouquot et al. 2009; Koutlas and Scheithauer 2010). Leiomyoma may demonstrate Verocay-like formation, but is S-100 negative and SMA positive (Brooks et al. 2002). Finally, approximately, 28% of head and neck schwannomas, may contain foci resembling neurofibroma, leading to an erroneous diagnosis of malignant degeneration (Butler et al. 2016).

Two hereditary syndromes that are related to schwannoma are NF2 and schwannomatosis (Berg et al. 2008). NF2 is an autosomal dominant disorder caused by mutation of the NF2 gene on chromosome 22q12.2 (Ruggieri et al. 2015). It is characterized by the development of vestibular schwannomas; other cranial, spinal or cutaneous nerve schwannomas; cranial and spinal meningiomas or other central nervous system tumors; ocular abnormalities (early onset cataracts, optic nerve sheath meningiomas, retinal or pigment epithelial hamartomas or both and epithelial retinal membranes); skin abnormalities (subcutaneous schwannomas, and café au lait pigmentation) (Ruggieri et al. 2015). Diagnosis is based on the modified criteria of the Manchester Group (Smith et al. 2017). Schwannomatosis is a rare disorder characterized by predisposition to develop multiple schwannomas that commonly affect peripheral nerves and the spine, and less commonly meningiomas.

The most common symptom is chronic pain, either diffuse or local (Kehrer-Sawatzki et al. 2017). The majority of schwannomatosis cases are sporadic with only 13–25% being familial (Kehrer-Sawatzki et al. 2017). Two predisposition genes for schwannomatosis have been identified, SMARCB1 on chromosome 22q11.23 and LZTR1 on chromosome 22q11.21 (Kehrer-Sawatzki et al. 2017).

Diagnosis can be set with the combination of both molecular and clinical testing or be only clinical (Kehrer-Sawatzki et al. 2017). A hybrid tumor with microscopic characteristics of both neurofibroma and schwannoma or an abundant myxoid stroma should raise the suspicion of NF2 or schwannomatosis (Plotkin et al. 2013), while paediatric plexiform schwannoma may also associate with either of the two syndromes (Berg et al. 2008). Of the 87 cases of the present review, only one patient (1.1%) was diagnosed with NF2 (Berg et al. 2008), while none of the patients had schwannomatosis.

Surgical excision is the treatment of choice for paediatric oral schwannoma. In 44 cases with follow-up, recurrence was reported in only 3 cases (6.8%), which were microscopically characterized as multinodular (Krolls et al. 1994; Hashiba et al. 2007) or plexiform (Di Giovanni et al. 2006). No case of malignant transformation was described.

Conclusion

A palatal swelling in children or adolescents is usually a dental abscess, but neoplasms, although rare, may have a similar clinical presentation, requiring careful clinical evaluation of the dentition and radiographic examination.

Compliance with ethical standards

Conflict of interest Author Tamiolakis Paris declares that he has no conflict of interest. Author Kalyvas Demos declares that he has no conflict of interest. Author Arvanitidou Ioanna declares that she has no conflict of interest. Author Vlachaki Adamantia declares that she has no conflict of interest. Author Tosios Konstantinos I declares that he has no conflict of interest. Author Sklavounou-Andrikopoulou Alexandra declares that she has no conflict of interest.

Research involving human participants and/or animals This article does not contain any studies with human participants or animals performed by any of the authors.

Informed consent For this type of study informed consent is not required.

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