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Case Report

Dentistry Section

An Uncommon Occurrence of Pleomorphic Adenoma of Submandibular Gland in a 14-Year-Old Child: A Case Report

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ABSTRACT

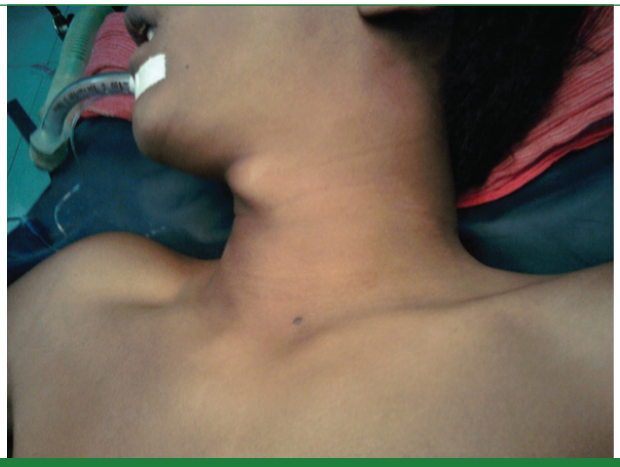
Neoplastic disorders of salivary gland are rare in children and adolescents as compare to salivary gland inflammatory diseases. When salivary gland tumour occur in children, they are approximately 10 times more likely to occur in parotid gland than submandibular gland. Pleomorphic Adenomas (PA) of submandibular glands mostly occurs in third to fifth decade of life but rarely occur in children and adolescents. Patients usually present with a painless mass without any other associated symptoms and with female predominance. Radiologic studies are usually unable to differentiate benign from malignant neoplasm in most cases. Surgical removal of the submandibular gland along with tumour is the best treatment option for PA in children and adolescents. PA consists of epithelial and myoepithelial cells which forms various patterns along with different types of stromal formation and sometimes play challenges for diagnosis. Recurrences are rare in submandibular gland PA and the prognosis is excellent. Here, a case of a 14-year-old male patient who presented with a painless left submandibular mass that developed over the period of four months and was not associated with fever, erythema, oedema has been reported. Preoperative imaging revealed a submandibular mass contiguous with the left submandibular gland. The mass and left submandibular gland were completely excised. The histopathology confirmed the diagnosis of PA arising from left submandibular gland without any malignant changes. The postoperative period was uneventful. Patient follow-up was done till nine months and no recurrence was noted. Early diagnosis and strict follow-up pivots the management of PA in children.

Keywords: Benign neoplasm, Children, Salivary gland

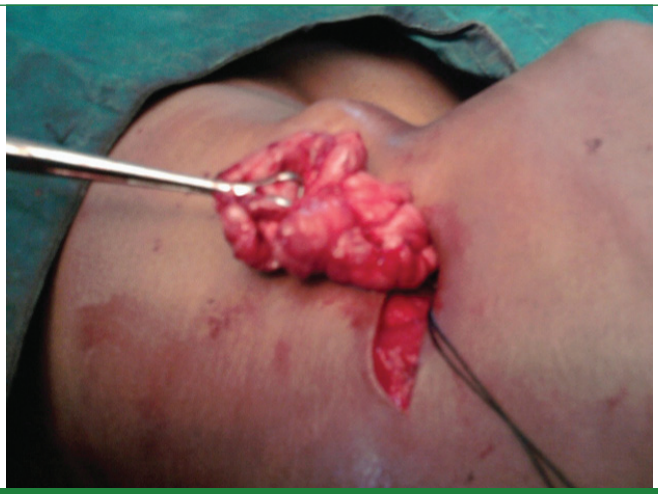
CASE REPORT

A 14-year-old male came to the Surgery department with a four months history of a slowly progressing lump in left submandibular region. On examination, a painless, firm, partially mobile, slow growing lump was present in left submandibular area with ill-defined margins and normal overlying skin [Table/Fig-1]. There was no history of fever, cough, redness, weight loss and anorexia. It was not associated with any relevant past history of medical, dental and surgical illness and family history also being insignificant. On general physical examination, patient was thin built, adequately nourished, well oriented to time, place and person with vitals being normal and all biochemical reports and chest radiograph were in normal limits. On palpation, mass was non tender and firm and no cervical lymphadenopathy was evident. The Ultrasonography (USG) neck showed a hypoechoic lesion of size 3.0x2.6x2.2 cm with well defined margins involving left submandibular gland. There was no obvious involvement or inflammatory changes of the surrounding tissues. Clinical differential diagnosis of sialadenitis and benign and malignant salivary gland tumour was made and provisional diagnosis of tumour of left submandibular gland was made based on clinical and radiological findings. Patient was posted for surgery. The surgery was proceeded with standard submandibular incision and complete excision of mass along with submandibular gland done under general anaesthesia. Closure was done after adequate hemostasis and specimen was sent for histopathologic examination [Table/Fig-2].

On gross examination, the specimen was 3.2x2.8x2.4 cm in size with tumour mass within the submandibular gland. Postoperative course was uneventful. The histopathologic examination revealed a partially encapsulated tissue comprising of both epithelial and mesenchymal components. Epithelial cells were arranged in solid sheets, tubules, acini and rods along with fibromyxoid and chondromyxoid stroma. Most of the tubular structures were composed of inner ductal epithelial and outer myoepithelial cell layer [Table/Fig-3-6]. No areas of malignant transformation were visualised. PA, myoepithelioma

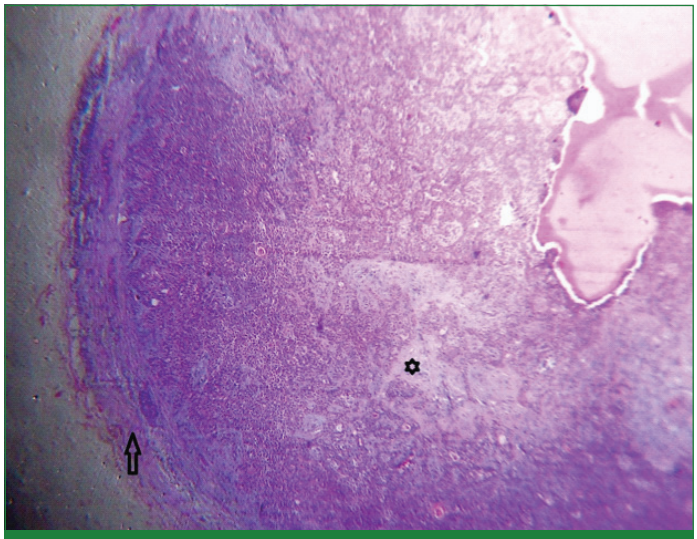


[Table/Fig-1]: Clinical picture having lump in left submandibular area.

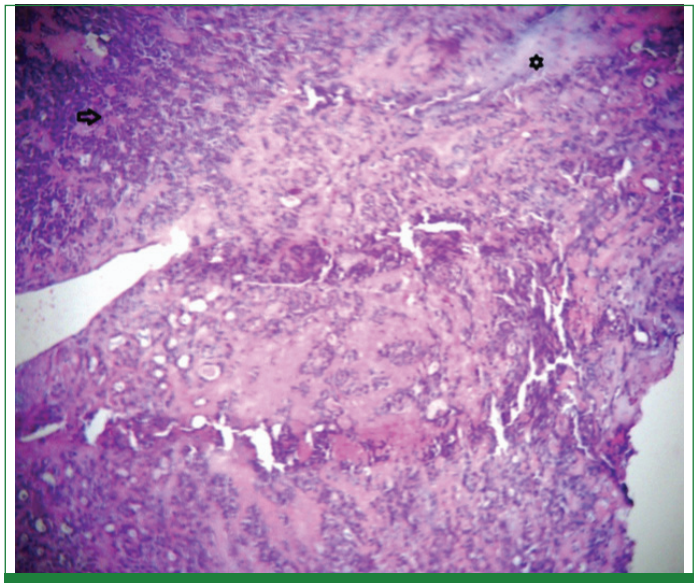


[Table/Fig-2]: Per operative image of the lesion.

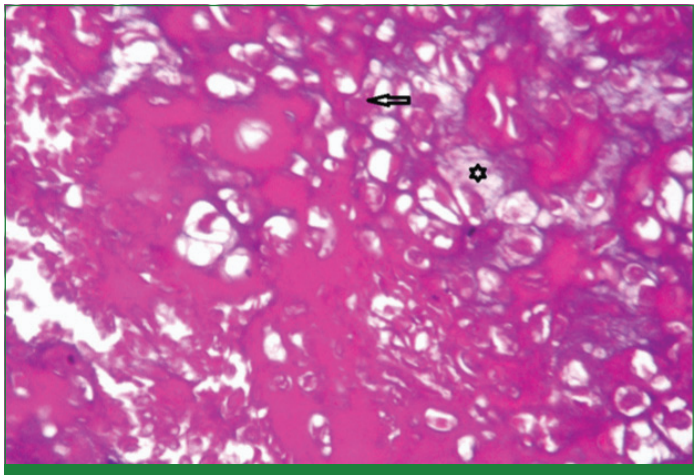
and adenoid cystic carcinoma were included in histopathologic differential diagnosis. The presence of myxochondroid stroma and ductal differentiation ruled out myoepithelioma. Absence of infiltrative growth and perineural invasion supported the diagnosis of PA and excluded adenoid cystic carcinoma. The final diagnosis of PA of left submandibular gland was made. Patient was discharged on second day after surgery without any complication and no recurrence was noted till nine months of follow-up.



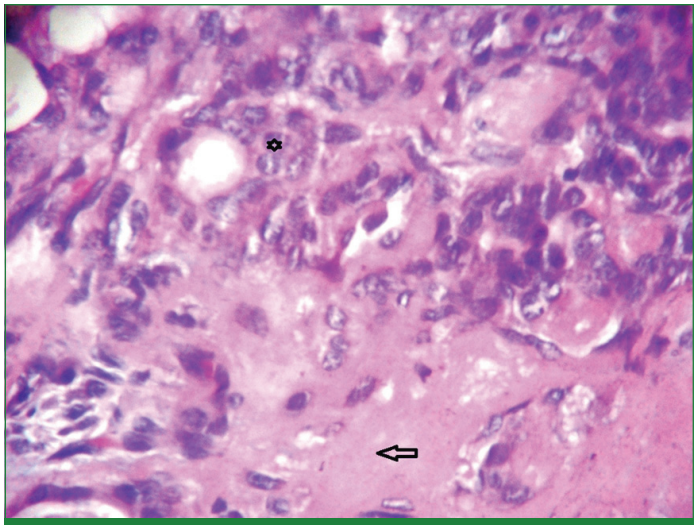
[Table/Fig-3]: Histopathological image showing cell rich proliferation of both epithelial and myoepithelial cells with myxomatous stroma (*) and surrounding capsule (→) (H&E, X40).



[Table/Fig-4]: Histopathological image showing cell rich components with ductal areas (→) along with hyalinised and chondromyxoid area (*) (H&E, X100).



[Table/Fig-5]: Histopahological image showing chondroid area (*) along with Pleomorphic cells (→). (H&E, X400).



[Table/Fig-6]: Histopathological image showing ducts surrounded by myoepithelial cell (*) with hyalinised stroma (→) (H&E, X400).

DISCUSSION

Salivary gland tumours are uncommon in young people under 19 years, representing only 3% of cases of all salivary gland tumours [1]. As such, the aetiology of PA is unknown but the increased incidence of PA has been noted in 15 to 20 years after exposure to radiation. According to one study, association of simian virus 40 was found in genesis of PA [2]. These evidences though explain tumour development in adult but could not justify development of PA in childhood and adolescents. No large series of PA is available to justify rarity of incidence in childhood and adolescents in comparison to adults. A study of 41 patients with PA in head and neck in children revealed that only seven were in submandibular gland [3]. PA in submandibular gland is extremely rare in children and adolescents. A largest reported series of 9,993 salivary gland lesions revealed that 430 cases occurred in children and of these only 10 cases of PA were in the submandibular gland [4]. When salivary gland tumours occur in children and adolescents they are approximately 7-10 times more likely to occur in parotid than in submandibular glands [4-7]. According to a study by Guzzo M et al., 87% PA was in parotid gland and only 10% cases in submandibular gland [7]. A study on 52 juvenile and adolescent patients shows PA in 60% of cases and only 7.5% of those in submandibular gland. The highest incidence was found in group of 14-16 years of age [5]. In a series of 640 cases of primary epithelial salivary gland neoplasm, only 2.9% were diagnosed in children and adolescents with female to male ratio of 1.7: 1 and only one case was the submandibular gland PA [6]. According to a review of the Japanese literature of PA of submandibular gland in 22 cases in children shows mean age of 12 years and female to male ratio 3:1 [8]. The symptoms were mainly painless slow growing mass and the mean duration of symptoms of evolution was 12.8 months. The clinical presentations of PAs are almost similar in children and in adult patients except the duration of symptoms which is relatively short compared with adults, that can be because of attention to lesion by parents [9]. In the present case, patient was 14-year-old male with painless slow growing mass since four months in submandibular area.

Imaging is useful in determining the size and extent of lesion such as ultrasound scanning, computed tomography scans and magnetic resonance imaging. The important first preoperative diagnostic setup in young patients is sonographic examination [5]. It is widely available, relatively cheap and safe for patient and easily replicable [9]. Radiographic examination by computed tomography and magnetic resonance imaging rarely offer more information on aetiology of submandibular lesion and also fails to differentiate between malignant and benign lesion. Fine Needle Aspiration Cytology (FNAC) is widely used to achieve the preoperative diagnosis of salivary gland tumours in adults; however its diagnostic role is disputed in children due to

compliance and difficulties in interpretation. So, there might be no definite preoperative examination to detect the malignant change in PA of submandibular gland [8,10,11].

Best surgical treatment option for submandibular gland PA includes complete resection of tumour along with associated submandibular gland [9]. When tumour enucleation is performed, recurrences occur in 50% of benign salivary gland neoplasm in children [7]. Recurrence is rare in submandibular gland PAs when compared with parotid tumours [9]. In the present case also complete submandibular gland excision was done along with the tumour.

Pleomorphic Adenoma usually present a remarkable degree of morphological diversity. The predominant components are epithelial and myoepithelial cells along with mesenchymal and stromal elements with varied capsule thickness [9]. PAs can be divided into four histological subtypes by Seifert G and Donath K, type 1 is the classic type with a stroma content of 30-50%, type 2 is the stroma rich with stroma content of 80%; and type 3 and type 4 are the stroma poor with stroma content of 20-30% or less [12]. Classic subtype is the most common histopathologic subtype in children and adolescent's overall and stroma rich type is the least common type but in submandibular gland PA, stroma poor type (cell rich) is the more common than classic type. Among 28 cases of submandibular gland PA, 13 cell rich type, 4 stroma rich type and 11 were of classic subtype [9]. Stroma poor type of PA reported in a study, was present in in the present case.

CONCLUSION(S)

The PAs of submandibular gland are unusual in children and adolescents and sometimes clinically indistinguishable from other masses of submandibular triangle, causing diagnostic uncertainity. The gold standard surgical treatment for PA of submandibular gland is complete resection of both, the tumour and the involved gland. Postoperative complications and malignant change are extremely rare and prognosis of submandibular gland PA is thought to be good in children.

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