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Major salivary gland tumors in children – diagnostic and therapeutic difficulties

Case Report

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Abstract: Major salivary gland tumors are very rare in the developmental period. Confirming tumor changes of the salivary gland requires precise diagnostic imaging involving an ultrasonography scan, computed tomography and magnetic resonance. A needle aspiration biopsy (NAB) of a tumor is of high importance. Excision of the tumor is the main treatment method in the case of parotid gland tumors. Statistical data concerning tumors suggest choosing less invasive methods, which seems very logical in children. The operational methods used in the tumor treatment are: extracapsular excision of a tumor, partial parotidectomy, total parotidectomy, sometimes proceeded with lymphatic nodes operations. Extracapsular excision of a tumor is a noninvasive method chosen because of simplicity and lesser risk of serious complications. This method is reserved only for the cases of benign tumors of the gland. Most authors, however, consider a partial parotidectomy as a method of choice in benign tumor cases and a total parotidectomy in cases of carcinomas of the parotid gland. Submandibular gland tumors need total gland excision. The clinical cases presented in this paper show the difficulties in diagnosis and treatment choices in cases of major salivary gland tumors in children.

Keywords: *Tumor • Salivary gland • Children*

1. Introduction

Salivary gland tumor changes in children are rare. They constitute about 10% of all child tumors in the head and neck area [1]. In the youngest children, the most common are hemangiomas and in pre-school and school children – inflammatory tumors [2,3]. Other tumors of the glands are very rare. The most common benign tumor of major salivary glands in this group is pleomorphic adenoma [4]. The most common location of this tumor, just as in adults, concerns the parotid gland [5]. The frequency of these neoplasms in children rises when they are over ten but about 70% of them appear between the ages of 16-20 [6]. The most common malignant salivary gland tumor in children is mucoepidermoid carcinoma. It occurs mainly in older children and young people but it is also a problem in children up to 10 years old [7]. Other salivary gland malignant tumors are as follows: adenoid cystic carcinoma, undifferentiated carcinoma, cancer originating from alveolar cells and sarcoma [8]. The examples of diagnostic and therapeutic difficulties in the cases of major salivary gland tumors in children are two patients hospitalized in the Department of Pediatric Otorhinolaryngology, at the Medical University in Lodz.

2. Case reports

The boy M.K, aged 15, was admitted to the clinic because of a left preauricular area tumor. The lesion drew attention for about 10 months. Previously, the case was consulted on by a surgeon and a laryngologist who recommended needle aspiration biopsy (NAB) of the lesion. The results after the second histopathological examination showed that it had the texture of a pleomorphic adenoma.

On the day of admission, the physical examination confirmed a tuberous lesion located in the left parotid gland projection, moving, about 2x2cm in dimension, with no colour or morphology skin change above the lesion. The examination did not show any features of

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the peripheral paresis or paralysis of the facial nerve. The patient's general condition was good and laboratory tests did not show any significant anomalies. Because of the size of the lesion and after the consultation with the oncological surgeon, a decision to excise the tumor by using the extracapsular method was made. During the operation the left parotid gland tumor of the size 1,7x1,5 cm was enucleated. However, in one pole of the lesion, infiltration with inflammatory reaction was noticeable. The whole lesion with the enlarged lymph node located between the parotid gland lobes was removed and sent for histopathological examination. The patient's general and local condition after the procedure was good and wound healing went well.

The histopathological examination of the tuberous lesion showed diametrically textured low grade mucoepidermoid carcinoma, which proved to have high histopathological differentiation. The lymph node demonstrated features of inflammation.

A quick decision about re-operation, with total parotidectomy on the left side on the seventh day after the surgery, was caused by the result of the histopathological examination. The trunk and branch of the facial nerve were preserved. The patient's general and local condition after the operation was good. Histopathological examination showed radicalness of surgery. The boy has been under outpatient clinic care for 12 months. Clinical trials and control ultrasonography did not show any alarming abnormalities. But in the fourth month of follow-up examinations, the patient was diagnosed with diabetes, which resulted in establishing diabetes treatment.

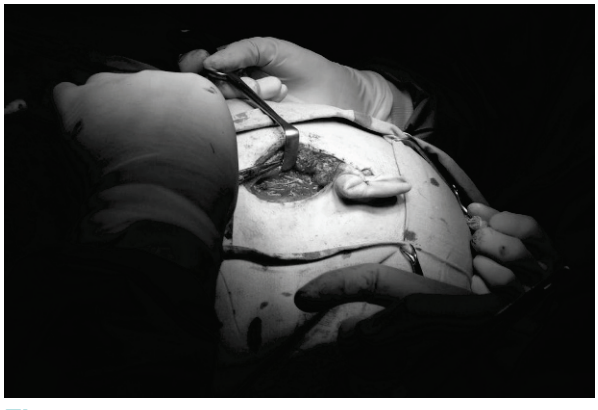

Figure 1. Total parotidectomy. Facial nerve visible beside the superficial lobe of the gland.

An example of a patient with submandibular gland tumor is the boy M.S., aged 12, admitted to the Clinic because of the left submandibular area tumor, visible and enlarging for about 1,5 months. The patient was treated pharmacologically by a general practitioner because of the "left side submandibular lymphadenopathy" for over a month. Ambulatory neck ultrasonography showed a solid focus 26x20x27 mm in the left submandibular gland in the area of the palpable lesion and submandibular lymph nodes of 8 mm, 12 mm and 15 mm in mandible angle area. Bilaterally single lymph nodes along the neck vessels were 9 mm in size.

In boy's case, an ambulatory NAB of the lesion was made. The results of NAB histopathological examination were as follows: cytological picture suggests adenoma pleomorphicum. On the day of admission to the Clinic, the clinic trial confirmed a tumor in the left submandibular area, 3 cm in diameter, painless and mobile. The skin above the lesion was not changed with regard to morphology and color. Computed neck tomography showed a ring-enhancing focus in the left submandibular gland less reactive to contrast medium than gland parenchyma of 24x26x30mm. The lesion did not infiltrate the neck vessels. In the lesion area there was a 14 mm lymph node and a few smaller ones. In the left neck area there were few lymph nodes 7-11 mm in size.

On the basis of clinical examination and supplementary tests, the boy qualified for the operation including excision of the submandibular gland on the left side. During the surgical procedure, a tumor 3 cm in diameter, lying on the outside of the submandibular gland in one block with the gland connected to it and enlarged submandibular lymph nodes, was removed. It was necessary to cut and ligate facial vessels going above the salivary gland and through the submandibular gland. The post operative period was without complications. Histopathological examination confirmed polymorphic adenoma and radicalness of surgery. Lymph nodes removed during the operation showed features of inflammation. The boy has been under outpatient clinic care for 6 months. Clinical trials do not show any signs of recurrence.

3. Discussion

Salivary gland tumors in children often show problems connected with the preoperative diagnostic workup of lesions in this area. The tumors are rare and special care should be provided by general practitioners in the cases of suspicious masses of the glands as it shortens the time between first sight of the disease and the operational treatment. The second case is an example of the situation. Confirming tumor changes of the salivary gland requires precise diagnostic imaging involving an ultrasonography scan and sometimes computed tomography and magnetic resonance. In the scans of computed tomography, these tumors are defined as

clearly separated and moderately saturated formations. In magnetic resonance, the description suggests the presence of the tumor whose saturation is bigger than salivary gland tissues and smaller than those present in lymph node areas and is usually well separated from the area [9]. In histopathological preoperative diagnostic needle aspiration biopsy (NAB) of a tumor is of high importance and, if possible, under ultrasonographic control. Specificity of fine-needle aspiration cytology for diagnosing malignant salivary gland tumors is 96% [10]. Many authors stress the necessity of careful histopathological evaluation of the preoperative NAB. What is more, they suggest taking repeated samples to confirm a preoperative diagnosis [11]. Despite established principles of diagnostics, clinical evaluation, the results of imaging studies, and a pathomorphological NAB do not always eliminate malignant processes [12]. The first case is an example of such a preoperational misdiagnosis. The principles of operational treatment of major salivary gland epithelial tumors in children do not differ from those in adults.

Benign parotid gland tumors in most cases require a superficial lobe excision only. But when NAB gives evidence of a malignant tumor, the operation covers total parotidectomy. In cases of a proved malignant lesion the method of radical excision of the parotid gland is used. The essential problem concerning total excision is the high probability of injuring the facial nerve and the necessity of the nerve fragment excision in case of infiltration [13]. Many authors believe that in cases of the most common benign tumor – pleomoprhic adenoma -superficial - partial parotidectomy should be used. In their view, it remarkably decreases recurrence of the tumor but at the same time it may often damage the facial nerve. Extracapsular excision of the tumor prevails in sparing of the facial nerve which decreases the number of complications among which Frey syndrome is the most common [14]. This method was used in the first case. It was proved that this method does not make it difficult to use complete parotidectomy after recognizing a malignant tumor from the samples removed during the first surgery.

In cases of submandibular gland tumors, total gland excision is needed. An operation in the submandibular gland area often requires cutting and ligating facial vessels, sparing the hypoglossal and lingual nerve as well as the facial nerve marginal branch of the mandible [15]. Sometimes it is possible to omit facial vessels during the operation. However, it was impossible in the case of our patient because of topography and tumor dimension [16]. Submandibular gland tumors are very rare in the developmental period [17]. A tumor lying beyond the gland is described in about 20% of cases of submandibular gland tumors in children. Ultrasonography helps to recognize a submandibular gland tumor, to differentiate it from other pathologies, and to recommend successful treatment [4]. As the ultrasound of the submandibular region can be ordered even by the general practitioner, it seems to be a basic method in the ambulatory differential diagnosis of the submandibular pathologies.

In cases of malignant tumors, when there is a suspicion that there are metastases to the lymph nodes, their excision is necessary [18]. Sometimes surgical excision is followed by radiotherapy, yet most authors consider surgical treatment to be the only therapeutic management, especially in children, because of the negative consequences of radiation therapy on their development. Total parotidectomy proved to be a successful treatment in the case of the boy with mucoepidermoid carcinoma of the parotid gland and augmentation therapy was not necessary. Chemotherapy is not used in the cases of major malignant salivary gland tumors in children [19].

Conflict of interest

All authors disclose any financial and personal relationships with other people or organisations that could inappropriately influence (bias) their work.

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