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Delayed Diagnosis of Sarcoma of the Oral Cavity – Case Report

Późne rozpoznanie mięsaka jamy ustnej – opis przypadku

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A – research concept and design; B – collection and/or assembly of data; C – data analysis and interpretation;
D – writing the article; E – critical revision of the article; F – final approval of article

Abstract

The paper describes the case of a 16-year-old boy (R.G.) with misdiagnosed pain in teeth 12 and 11 and tumor of the right cheek. The patient was treated in a private dental surgery for over four months without proper diagnosis made. As no effect was gained by the endodontic treatment of two permanent upper incisors, the extraction of one of them (tooth number 12) and three courses of antibiotic therapy, the patient was referred to the Department of Pediatric Dentistry, Medical University of Łódź. During an initial investigation, cancer was suspected. In the Department of Pediatrics, Oncology, Hematology and Diabetology, Medical University of Łódź, the diagnosis of sarcoma (Rhabdomyosarcoma alveolar) in the suborbital region was confirmed by histopathological investigation. Oncological treatment was begun and the root canal therapy of tooth 11 had to be continued, despite it being located inside the malignant tumor. However, despite intensive anticancer therapy, the patient died 5 years after diagnosis.

On the basis of the described case, a dentist should be aware that atypical symptoms and the failure of typical dental treatment can be signs of a general disease, including cancer of the oral cavity. Early detection of neoplasm always improves the patient's chances of full recovery (**Dent. Med. Probl. 2015, 52, 3, 345–350**).

Key words: child, sarcoma, toothache.

Słowa kluczowe: dziecko, mięsak, ból zęba.

Sarcomas comprise 3–5% of all cancers in childhood, and rhabdomyosarcoma (RMS) make up 50–70% of all cases of sarcomas in children [1]. These tumors are most often found in the head and neck regions (40% of all RMS) [2]. The incidence of the disease peaks in early childhood, between 2 and 4 years, and then during the second decade of life – in children over 12 years of age. Prognostic factors worsening the outcome of the disease are the diameter of the tumor > 5 cm, the presence of distant metastases and a parameningeal location. Prognosis is also worse in children over the age of 10, in the case of a non-radical tumor resection or if the histopathological type of the tumor is an unfavorable, *alveolar* type [3–5].

Case report

In February 2009, a 16-year-old boy (R.G.) was referred to the Department of Pediatric Dentistry, Medical University of Łódź, from a private dental surgery for constant “swelling of the right cheek” (Fig. 1 a, b).

In the interview in the Department of Pediatric Dentistry, it was revealed that the patient had been dentally treated for over 4 months because of persistent pain in the region of teeth 11 and 12, swelling in the region and constant elevation of body temperature (37–37.5 degrees Celsius). Endodontic treatment had been started in teeth 11 and 12. However, as no improvement was observed, the

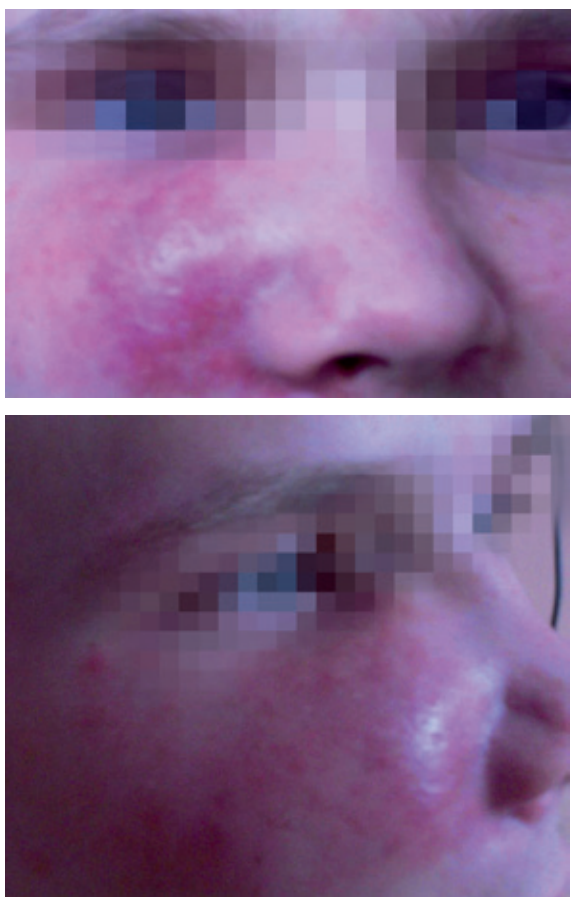


Fig. 1 a, b. Patient R.G., aged 16, at arrival

decision was made to extract tooth 12. The patient was concomitantly treated with antibiotic therapy. After the extraction, despite antibiotic cover, the swelling of the right cheek enlarged. The patient was transferred to the university outpatient clinic one week after the extraction. In the meantime, he experienced a few episodes of epistaxis.

On admission, a tumor measuring 5×3 cm was found in the right suborbital region. The tumor was painful on palpation, tough and unmovable, and the skin over the tumor was reddened. The edema was also noticeable in the right part of the oral vestibule and slightly crossed the midline of the body. The submandibular lymph nodes of group C were found to be grossly enlarged, solid, painless and unmovable. Dental examination revealed tooth 12 had already been extracted, while root canal therapy of tooth 11 had been started but not finished.

Oncological Treatment

The patient was transferred to the Department of Pediatrics, Oncology, Hematology and Diabetology, Medical University of Lodz with the primary diagnosis of cancer. After CT scans of the head (Fig. 2) the child was qualified for open surgical

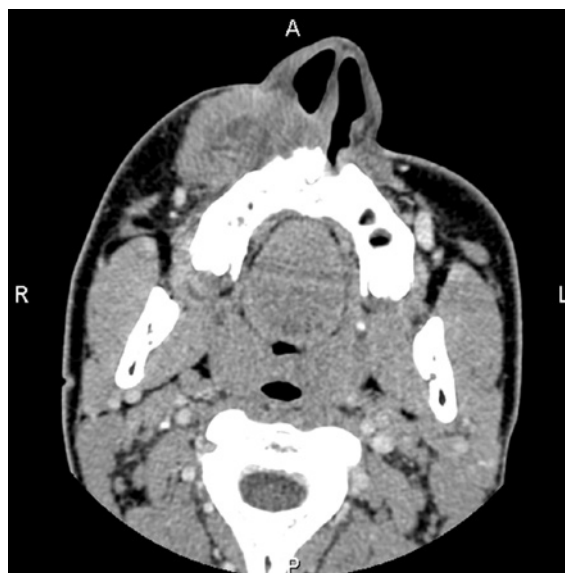


Fig. 2. CT scan of patient's head, tumor located on the right side

biopsy of the tumor. On the basis of histopathological examination, the diagnosis of *Rhabdomyosarcoma alveolar* was confirmed. Histopathological examination also revealed cancer infiltration in regional lymph nodes (right submandibular and right neck region). No distant metastases were identified. The patient was qualified for pre-operative chemotherapy within the very high-risk group according to the CWS 2006 protocol (chemotherapy VAIA III with ifosfamide, vincristine, dactinomycin and doxorubicin). Chemotherapy caused regression of the tumor. In June 2009, total removal of the tumor was performed (resection R1). Local treatment was continued with radiotherapy of the tumor and affected lymph nodes, to a total dose of 42–49 Gy. Post-operative chemotherapy was finished in October 2009 according to the protocol of treatment.

In August 2010, a local relapse of the disease was revealed. The child was re-operated and second-line chemotherapy (ifosfamide, carboplatin, etoposide) was continued to January 2011. Unfortunately, three local relapses were diagnosed during the following two years. The patient was treated with third-line chemotherapy (docetaxel, gemcitabine) and individual therapy (PACE protocol, Temodal + irinotecan). In December 2013, dissemination to the central nervous system was diagnosed. The patient died in March 2014 due to the progression of the cancer.

Dental Treatment

Together with the beginning of oncological treatment, root canal therapy of tooth 11 was continued: the pulp chamber had been left open



Fig. 3. X-ray of tooth 11, at arrival



Fig. 4. X-ray of tooth 11, 18 months after root canal therapy

by the previous dentist. Necrosis of the pulp and periodontal exudating inflammation of periapical tissues were found (Fig. 3). The pulpal remnants were removed and root canal was prepared by rinsing the root canal with 0.02% chlorhexidine solution, EDTA and saline. Non-invasive solutions were used to avoid irritating the surrounding tissues. A temporary dressing of calcium hydroxide was applied in the root canal for three weeks. During root canal therapy, there were problems with establishing a root length with aid of an electronic locator, which were probably caused by neoplasm tissues around the tooth apex. During the third visit, a control x-ray with a gutta-percha point placed in a canal was taken.

During the final preparation with the step-back method, the working length was shortened as the patient felt strong pains. The root canal was prepared to a length of 22 mm, MAF = 50, FF = 70. It was obturated with gutta-percha points sealed with AH+ paste using the cold lateral condensation method. However, on an x-ray, a small under-fill of about 1 mm of the canal obturation was found. After 18 months, a control radiogram was made of tooth 11 and no inflammatory changes or clinical symptoms were observed (Fig. 4).

During anticancer therapy, the patient also felt pain in tooth 46. An x-ray revealed the presence of periapical inflammation around the mesial root (Fig. 5). After gaining written consent from the oncologist, it was decided to extract tooth 46 under an antibiotic cover.

During the first remission of the cancer, irreversible inflammation of the pulp of tooth 25 occurred. The extirpation of pulp under local anesthesia was performed, and two root canals were prepared and obturated with gutta-percha points sealed with AH+ paste (Fig. 6).



Fig. 5. X-ray of tooth 46, inflammatory changes around mesial root



Fig. 6. X-ray of tooth 25, after pulp extirpation and obturation of roots

Discussion

According to data from the Polish Central Statistical Office, about 7 million children and adolescents aged between 0 and 17 years live in Poland [6]. Annually, about 1100–1200 new cases of

neoplasms are diagnosed in these children. On a positive note, the incidence rate of cancer in the Polish child population is slightly below the EU average (134.1 compared to 138.5). However, the incidence rate of Rhabdomyosarcoma (RMS) is found to be higher: 10.6 for Poland compared to 9.1 for the EU [7].

Great progress has been made in oncological treatment in Poland during the last 50 years, which has led to increasing numbers of long-term remission and complete recovery, as well as a higher survival rate [8]. However, despite rapid growth in diagnostic and therapeutic aids, early recognition of the first cancer symptoms is still a key part of the process of diagnosis. It should be stressed that early recognition is the responsibility of general practitioners, pediatricians and dentists, who are in everyday contact with their patients. Any lack of knowledge and skills in the field can lead to a delay of anticancer treatment, which grossly worsens the prognosis for full recovery. Unfortunately, in Poland, about 70% of all childhood cancers are diagnosed in the 3rd or 4th stage of the disease, when the probability of complete recovery is much lower than in the 1st or 2nd stage [9]. In a systemic review of the relationship between delays in diagnosis and advanced cancer stages, Gómez et al. [10] reported the strongest association to be in cases of oral cancers and when the delay in diagnosis was longer than 1 month.

However, early detection of oral cancer can be easily performed by both medical and dental practitioners by a simple investigation of the state of the oral mucosa. A survey by Carter and Ogden [11] of 238 medical practitioners (GMP) and 133 dental practitioners (GDPs) on knowledge of oral cancer detection and prevention revealed that routine examinations of the oral mucosa were performed by 95% of GDPs and by only 20% of GMPs. Even in cases of patients at high risk of cancer development, more than 65% of GMPs did not examine their patients' oral mucosa.

However, there might be little purpose to regular investigations if dentists are unaware of the symptoms of cancer. Janas et al. [12] analyzed 71 cases of patients with non-diagnosed tumors of the oral cavity who were referred to the Department of Dental Surgery, Medical University of Łódź from private practices, between the years 1998 to 2005. They found that 38 patients were treated without a proper diagnosis for more than 2 months, and in a further 30 cases, dentists planned an extraction of the moving teeth. Two patients were referred because of rapid growth of a tumor after extraction of teeth. When the patients were actually diagnosed, most of them suf-

fered from highly advanced cancers. The authors concluded that more stress should be placed on education of the prevention and early detection of neoplasms. Aware and responsible dentists should refer their patients with suspect changes in the oral cavity to a specialist early enough to allow complete recovery. According to McGurk and Scott [13], attention should be elicited during the investigation if one of these symptoms are found: non-healing ulcers, persistent discomfort or pain, persistent red or white patches in mucosa, a lump or thickening. The symptoms of cancer may also include difficulty in chewing and swallowing, unusual bleeding or numbness of teeth, loose teeth for no apparent reason, difficulty in moving the jaw, speech problems, sore throat or earache in the same side, or lymphadenopathy. Also special attention should be paid to patients at high risk of cancer development. The main risk factors of cancer are tobacco smoking or chewing, alcohol consumption, UV light exposure, viral factors, chronic infections and nutritional problems such as a diet low in iron, vitamin A or C, or a high fat diet. In dental practice, special attention should be paid to patients with chronic irritation from jagged teeth and with poor oral hygiene and dental state.

In the case described above, in spite of the presence of several cancer symptoms, the start of anticancer therapy was delayed by four months. Dental treatment had been started without proper diagnosis, then continued despite no results gained. The dentist administered several courses of antibiotic without strong indications and then extracted one of the upper permanent incisors directly from a tumor. However, the misdiagnosis of a malignant tumor mimicking dental abscess or other dental problems has been already described in contemporary papers [14–17]. A similar case was also reported by Soares et al. [18], who describe a case of osteosarcoma of the mandible. The tumor had initially resembled a lesion of the dental periapex and a premolar had been subjected to endodontic treatment before a proper diagnosis was made.

Our patient also had to undergo endodontic therapy of the upper central incisor, which continued despite medical problems. Special effort was made not to irritate the neoplasm tissues around the root during dental treatment to avoid any stimulation of the growth of the tumor. During further therapy, periapical inflammation of tooth 46 and inflammation of the pulp of tooth 25 were also noted. In the first case, it was decided that tooth 46 should be extracted, while an extirpation of the pulp of tooth 25 was performed later during disease remission. The methods of dental treatment

were chosen in accordance with the medical condition of the patient.

The dental management of children during anticancer therapy requires special thoughtfulness that has been widely described by other authors [19, 20]. During the course of anticancer therapy and later, patients suffer from several health and dental complications. Childhood cancer treatment negatively affects the immune system, increasing the risk for bacteremia and septicemia. That led to the conclusion that all teeth with non-vital pulp in active cancer patients should be prophylactically removed [21]. However some authors denied the necessity of such an acute approach. Halperson et al. [22] performed an investigation of cancer patients who had dental pulpotomy treatment in 41 milk teeth and compared them with such records in 41 healthy children.

They found no statistically significant difference of pulpotomy success rates among the groups.

In our patient, the incisors should not have been removed as they were placed directly in the middle of the tumor. As has been already described by Janas et al. [12], such surgical intervention may lead to the stimulation of tumor growth.

However, despite all the later efforts made by dentists and oncologists, our patient was lost.

On the basis of the described case, a dentist should be aware that tumors of the head and neck can also appear in patients without risk factors, including children and young adults. Atypical symptoms and the failure of typical treatment should alert the dentist to the possibility of other diseases, including cancer. Such an approach allows delayed diagnoses to be avoided and improves the chances of full recovery.

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