Trismus in a 6 year old child: a manifestation of leukemia?

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Trismus is a firm closing of the jaw due to tonic spasm of the muscles of mastication from disease or the motor branch of the trigeminal nerve. Trismus may be produced by a variety of reasons such as dental abscess, trauma, following mandibular block with local anesthesia, as a result of radiation to the facial muscles, and patients after chemotherapy. A case of a referral of a six-year-old boy to a dentist from an ENT due to severe limitation in jaw opening is presented. Intraoral examination and panoramic radiograph demonstrated no signs of infection and/or other pathology. After a diagnosis of trismus was made, due to his icteric appearance, the general fatigue and loss of appetite in the last few days, palpated and sensitive lymph nodes in the submandibular and cervical regions, the child was referred for a complete blood count and sedimentation rate. The laboratory and clinical findings resulted in the diagnosis of acute lymphoblastic leukemia (ALL). Dental and oral manifestations of ALL are discussed, and the trismus may be explained by an intensive infiltration of leukemic cells into the deep portion of the contracting muscles of the face. This case emphasizes the importance of physical examination and independent judgement made by dentists, even when patients are referred to them by other members of the medical communities.
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INTRODUCTION

Trismus is a firm closing of the jaw due to tonic spasm of the muscles of mastication from disease or the motor branch of the trigeminal nerve, usually associated with and due to general tetanus.[1]

Trismus may be produced by painful lesions in the neighborhood of the jaw such as dental abscess, trauma, or it may follow mandibular block with local anesthesia. The presence of the causative lesion and the localized character of the spasm enable these cases to be distinguished from tetanus.[2]

Trismus may also occur in-patients, who are treated with phenothiazine drugs, in-patients with muscle infections and scleroderma. Muscle contractures of the face, and trismus often occur as a result of radiation to the facial muscles, and in patients after chemotherapy.[3,4]

Lesser degrees may be associated with disease of the pharynx and temporomandibular joint, myofacial pain dysfunction (MPD), and osteoarthritis, or may be a manifestation of encephalitis.[5]

Trismus is symptomatic of tetanus, an intoxication of the nervous system with the exotoxin of *Tetanus bacillus*. It is characterized by the progressive development of muscular rigidity, which is subject to paroxysmal exacerbations. However conditions causing trismus may be confusing with tetanus.

Hysteria may cause either trismus or general rigidity. Hysterical trismus is relatively common, often after minor injury.[2]

Theoretically any strong triggering of the contractor muscles of the jaw may induce trismus including leukemic cell infiltrate. However, to the best of the authors' knowledge, no report of a trismus as a sign of acute lymphoblastic leukemia (ALL) has been published.

A case of a referral of a child to a dentist due to a severe limitation in jaw opening, which resulted in the diagnosis of ALL is presented.

Case report

A 6 yew old boy was referred to the dental clinic specializing in oral medicine, by his in ear, nose and throat (ENT) doctor due to severe limitation in his opening of

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Figure 1. A panoramic radiograph demonstrating no signs of infection and/or other pathology.

the mouth. The ENT physician could not diagnose any disease associated with this symptom.

On examination, the child looked icteric, his mother mentioned general fatigue and loss of appetite in the last few days. Clinical extraoral examination revealed that the lymph nodes in the submandibular and cervical regions were palpated and sensitive bilaterally. His jaw opening was limited to 20 millimeters. The masseter and temporalis muscles were both tender bilaterally on palpitation. Intraoral examination was difficult because of the limitation in the jaw opening, but the soft tissues appeared normal, and no carious teeth were noted.

Panoramic radiograph demonstrated no signs of infection and/or other pathology (Figure 1). A diagnosis of trismus was made.

The child was further referred for a complete blood count and sedimentation rate due to his general clinical appearance. Blood count revealed the following: white blood cells were $1.5x10^3$/micl, red blood cells were $3.08x10^6$/micl, Hgb was 8 g/dcl, hematocrit was 22.9%, platelets were $45x10^3$/micl, neutrophils were 7.2%, lymphocytes were 87.4%, monocytes were 3.8%, eosinophils were 1% and basophils were 0.1%. Microscopic examination revealed lymphocytes with irregular appearance: cells without cytoplasm, partly with nucleuses, and partly with beaded nucleuses. A diagnosis of acute lymphoblastic leukemia was made, and the child was immediately referred to pediatric hematology for treatment.

DISCUSSION

Trismus is a firm closing of the jaw due to tonic spasm of the muscles of mastication from disease or the motor branch of the trigeminal nerve, usually associated with and due to general tetanus.[1] However, a variety of other factors such as oral infections, muscle stress, temporal mandibular joint dysfunction and direct injection into the muscle were associated with this phenomenon.

Leukemia is cancer of the white blood cells. It is characterized by the progressive over proliferation of white blood cells, which usually appear in the circulating blood in an immature form.[6] Any of the white blood cells may be involved by this disorder. Thus, the disease can involve proliferation of myeloid or lymphoid cells. It can occur in both an acute and a chronic form.

About 50% of all leukemia is in the acute form. Acute lymphoblastic leukemia (ALL) is the type most commonly found in children.[7] ALL accounts for about 50% of all neoplasms in children. It is found most often in 2-to 4-year-old children. Seventy-five percent of the cases are in this age group, and 25% are in teenagers and adults. ALL is classified based on the type and size of the lymphocytes found. Over 90% of the leukemia lymphoblasts in ALL contain a nuclear enzyme, terminal deoxynucleotidyl transferase (Tdt), which serves as a marker for this disease. The cause for leukemia is unknown however, an increased risk is found to be associated with large doses of ionizing radiation, certain chemicals, and a few viruses.[7]

General signs and symptoms of leukemia include: dyspnea, palpitations, fever, weakness, recurrent infections, weight loss, body pains, and pallor.[6,7]

Oral signs and symptoms are common in the clinical course of leukemia. Therefore, the dentist may be the first to suspect the disease. Head and neck signs result from leukemic infiltrates or marrow failure. These include cervical lymphadenopathy, oral bleeding, gingival infiltrates and enlargement, oral infections, pulpal abscesses, oral ulcers, and loose teeth.[7] Thrombocytopenia and anemia caused by the marrow suppression from disease and chemotherapy result in pallor of the mucosa, petechiae, and ecchymoses, as well as gingival bleeding. Spontaneous gingival bleeding is common when the platelet falls below 20,000/mm.[3,6] Oral mucosal ulcers are caused by the direct effect of chemotherapeutic drugs on the oral mucosal cells. Bacterial invasion secondary to the disease is very common.[8]

In the present case, the decision-making analysis as to the differential diagnosis of the condition included the following parameters:

Intra oral examination, as well as panoramic radiograph, did not reveal any obvious local cause for the trismus. There was no abscess, nor did the child receive any dental treatment prior to the visit to the dentist. MPD and osteoarthritis are very rare in children. The child had tetanus vaccination, and no muscle progressive rigidity was observed, therefore tetanus was not considered firstly. The fatigue behavior did not confirm any aspect of hysteria. We could not, at this stage, establish a reasonable cause for the presence of trismus since this phenomenon is quite rare in this age group.[9,10]

The general appearance, icteric, increased fatigue and loss of appetite in the last days, and the sensitive lymph nodes suggested a general disease, and justified blood tests. No intraoral pathology was noted, obviously

none of the signs of leukemia, and the only sign that could be associated with the general condition was the trismus. The dental literature does not suggest an association between trismus and ALL. The diagnosis of ALL was made after the blood tests and in accordance with the clinical presentation of the patient.

The trismus that occurred in the present case, may be explained by an intensive infiltration of leukemic cells into the deep portion of the contracting muscles of the face.[11,12] Lymphatic cells are known to infiltrate oral tissues such as marginal gingivae, peripheral blood vessels and intrinsic visceral, organs,[13,14] the possibility therefore, that infiltrating leukemic cells are responsible for the trismus is quite appealing. The case emphasizes the importance of physical examination and independent judgement made by dentists, even when patients are referred to them by other members of the medical communities.

REFERENCES

1. Steadman's medical dictionary. 25th edition, Williams and Wilkins, Baltimore, 1990.
2. Walton IN. Brain's diseases of the nervous system. Oxford: Oxford University Press, pp 819-20, 1977.
3. Jaffe N, Toth BB, Hoar RE, Ried HL, Sullivan MP, McNeese MD. Dental and maxillofacial abnormalities in long-term survivors of childhood cancer: effects of treatment with chemotherapy and radiation to the head and neck. Pediatrics 73: 816-23, 1984.
4. Dahllof G, Krekmanova L, Kopp S, Borgstrom B, Forsberg CM, Ringden O. Craniomandibular dysfunction in children treated with total-body irradiation and bone marrow transplantation. Acta Odontol Scand 52: 99-105, 1994.
5. Victor M, Adams RD. Diseases of cranial nerves. In G Thorn, RD Adams, E Braunwald, KJ Isselbacher, RG Petersdorf (Edst), Harrison's Principles of Internal Medicine, 8th ed., Tokyo, McGraw-Hill Kogakusha Ltd., p 1820, 1977.
6. Lynch MA, Brightman VJ, Greenberg MS. Burket's Oral Medicine. Diagnosis and Treatment, 9th ed., Philadelphia: Lippincott-Raven, pp 521-3, 1997.
7. Little JW, Falace DA. Dental management of the medically compromised patient. 4th ed., St. Louis, Mosby, pp 445-9, 1993.
8. Wahlin YB, Matsson L. Oral mucosal lesions in patients with acute leukemia and related disorders during cytotoxic therapy. Scand J Dent Res 96: 128-36, 1988.
9. Cousin GC. Oral manifestations of leukaemia. Dent Update 24: 67-70, 1997.
10. Fleming P. Dental management of the pediatric oncology patient. Current Opin Dent 1: 577-82, 1991.
11. Okamoto K, Hiral S, Honma M. Extensive leukemic cell infiltration into skeletal muscles. Muscle Nerve 19: 1052-4, 1996.
12. Fukuzawa J, Akaishi T, Tanaka H, Nakanishi K, Okada M, Haneda T, Kikuchi K. Adult T-cell leukemia with invasion by malignant cells of both parotid glands diagnosed by an Ga-67 imaging: a case report. Clin Nucl Med 22: 61-3, 1997.
13. Fulp SR, Nestok BR, Powell DL, Evans LK, Geisinger KR, Gilliam JH. Leukernic infiltration of the esophagus. Cancer 71: 112-6, 1993.
14. Anil S, Smaranayake LP, Nair RG, Beena VT. Gingival enlargement as a diagnostic indicator in leukaemia. Case report. Aust Dent J 41: 235-7, 1996.