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SAPHO Syndrome as a Potential Cause of Chronic Mandibular Bone Inflammation and Temporomandibular Joint Dysfunction – Case Report

Zespół SAPHO jako potencjalna przyczyna przewlekłego zapalenia kości żuchwy oraz zaburzeń funkcji stawów skroniowo-żuchwowych – 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

SAPHO syndrome (synovitis, acne, pustulosis, hyperostosis, osteitis) is a rare disease, with an unknown, probably multifactor etiology, comprising osteoarticular disorders of aseptic inflammatory character, and cutaneous changes of massive acne or palmoplantar pustulosis type. Usually the changes involve the anterior thoracic wall, and sternoclavicular and sacroiliac joints. According to literature reports, the mandible is affected in 10% of cases.

The paper presents a case of SAPHO syndrome in an 18-year-old patient, with the involvement of the right body, angle, and branch of the mandible and the right temporomandibular joint. Considering the results of imaging examinations (computed tomography, magnetic resonance, and scintigraphy), as well as laboratory, histopathological and microbiological investigations, the authors discuss the possible diagnostic problems and the differentiation of SAPHO syndrome with other diseases. The article also presents different treatment methods, including temporomandibular joint reconstruction with the use of an alloplastic prosthesis.

The aim of the paper is to present SAPHO syndrome as a potential cause of primarily chronic mandibular bone inflammation and stomatognathic system dysfunction **(Dent. Med. Probl. 2016, 53, 1, 125–133).**

Key words: SAPHO syndrome, mandibular bone inflammation, hyperostosis, temporomandibular joint inflammation, acne.

Słowa kluczowe: zespół SAPHO, zapalenie kości szczęk, hiperostoza, zapalenie stawów skroniowo-żuchwowych, trądzik.

SAPHO syndrome is a rare disease of joints, bones, and skin. The name, an acronym of terms describing the main signs (synovitis, acne, pustulosis, hyperostosis, osteitis), was introduced to medical literature by Chamot et al. in 1987 [1].

SAPHO syndrome, a rheumatic disease, is included among seronegative spondyloarthropathies by many authors [2, 3]. It can coexist with psoriasis, unspecific enteritis, Crohn's disease, and ulcerative colitis [2, 3].

The diagnosis of SAPHO syndrome can be established if at least one of the following criteria is fulfilled [1, 2]:

1. Osteoarticular signs associated with acne of intense course.
2. Osteoarticular signs in the course of palmoplantar pustulosis (PPP).
3. Hyperostosis with or without cutaneous changes.

4. Chronic recurrent multifocal osteomyelitis (CRMO) affecting the axial or peripheral skeleton.

SAPHO syndrome is considered to be a rare disease. The incidence of not more than 1/10,000 is reported in literature, but this data seems underestimated [4–8].

SAPHO syndrome can occur at any age. Most often it affects children (CRMO), young adults, and middle-aged people [4–9]. It rarely appears later than in the 6th decade [5–7, 9]. Primary investigations on SAPHO syndrome did not point to any dependence on sex [1], although later studies prove slightly more frequent incidence in women [5, 7, 8, 10, 11].

The pathogenesis of SAPHO syndrome remains unknown, and the mechanisms linking cutaneous changes with osteoarticular disorders are unclear. The syndrome has a multifactor etiology; inflammatory, immunological and genetic factors are taken into consideration [7, 8]. According to the inflammatory hypothesis, the cutaneous and osseous changes constitute a response to a chronic infection with a pathogen of low virulence. *Propionibacterium acnes* may be considered an example of such a pathogen; in the course of SAPHO syndrome, it has been isolated from joints and from osseous bioptates [6, 9, 10]. The bacterium, naturally present on skin, has been shown to be able to induce immunologic reaction; owing to its capability of raising the concentration of interleukin IL-1, IL-8 and IL-18, as well as tumor necrosis factor α (TNF-α) in the environment, it can also maintain the inflammatory response [6]. However, it is worth noting that the vast majority of osteitis cases in the course of SAPHO syndrome are aseptic inflammations [5, 6, 9, 10]. It has been proved not only by microbiological investigations, but also by polymerase chain reaction (PCR) tests [9].

According to another hypothesis, SAPHO syndrome is suspected to be an autoimmunologic disease resulting from molecular mimicry of pathogens and normal structures of bones or joints [3, 9]. As a consequence of similarity, the host's immune system, oriented at destroying the microorganism, attacks the osteoarticular structures (the so-called cross reaction), inducing specific changes in them [3, 9].

Familial incidence of SAPHO syndrome points at a probable genetic basis of the disease [12, 13]. A role of the NOD2/CARD15 genetic mutation, related to Crohn's disease, is suspected, leading to excessive activation of nuclear factor κB (NF-κB) [5, 6, 8]. An association of SAPHO syndrome with the T309G allele and the GG genotype of the Mdm2 gene seems also significant [6, 9]. In addition, important implications in the pathogenesis of SAPHO syndrome are ascribed to stress [7, 8]. Stressful situations and excessive emotional tension have been observed to predispose to the disease aggravation.

Clinically, SAPHO syndrome manifests itself with osteoarticular system signs (synovitis, hyperostosis, osteitis) and cutaneous signs (acne, pustulosis) [3, 6, 10].

Acne occurs in 1/4 of patients with SAPHO syndrome, mainly in men [1, 9, 10], and proceeds in the form of acne conglobata, acne fulminans, or suppurative apocrine gland inflammation (acne inversa) [3, 6, 9]. Most often, the acne affects the skin of the face, back, shoulders and chest, and in the case of acne inversa – axillary fossae, groins, and the sex organs region. It is characterized by a severe course and therapy resistance [6, 9].

PPP in SAPHO syndrome more frequently affects women [10]. Yellowish intracutaneous aseptic pustules are observed on hands and soles. The course of the disease is chronic, with phases of aggravations and remissions. PPP was observed in 2/3 of patients with SAPHO syndrome [3, 14].

Other cutaneous signs associated with the syndrome include psoriasis (mainly pustular psoriasis), pyoderma gangrenosum, and Sweet's syndrome [5, 9]. The cutaneous changes in SAPHO syndrome can be accompanied by osteoarticular changes, precede them, or occur later on. Most frequently, the distance between the appearance of these signs does not exceed 2 years; however, a nearly 40-year gap between the manifestation of cutaneous and rheumatic signs has also been described [9, 10]. Lack of cutaneous changes does not exclude SAPHO syndrome. It is estimated that they are not present in as many as 25% of cases [6, 8].

Osteoarticular manifestations are characteristic of and fundamental to SAPHO syndrome.

Synovitis most often involves the joints of the anterior thoracic wall (i.e. sternoclavicular joints and sternocostal synchondroses), as well as the sacroiliac joints [10]. Spinal and peripheral joints are less frequently affected; among them, the least common localizations are knee joints and temporomandibular joints [7, 11, 15–17]. The involvement of joints probably takes place through the expansion of the chronic inflammation present in bones [10]. Synovitis can proceed as a chronic process or as an acute disease imitating a bacterial inflammation [7, 8]. Joint involvement is usually unilateral, and changes can affect one or numerous joints at the same time. The result is articular space narrowing and intra-articular erosion [3]. Clinically, joint involvement can manifest itself by joint mobility disturbances, pain, and edema, but without abscess formation.

The term 'hyperostosis' refers to excessive osteogenesis in both the cortical layer of a bone and the marrow cavity, resulting from excessive subperiosteal and intraperiosteal proliferation. Radiologically, areas of sclerosis with thickened trabeculae, and periosteum thickening are observed. As an effect of these changes, the marrow cavity may be narrowed, and the external bone surface enlarged, thickened and irregular [3, 5].

Osteomyelitis in the course of SAPHO syndrome adopts the form of a primarily chronic inflammation [18]. The process involves both the cortical layer of a bone and the marrow cavity, clinically manifesting itself with bone pain and thickening [3, 18]. The radiographic image initially remains unchanged. At the early stages, osteolytic changes are visible, with or without sclerotic margins, with subperiosteal reaction. The number of osteosclerotic areas rises with the progression of the disease [9]. What is characteristic of SAPHO syndrome is lack of osseous sequesters or abscess formation. Osseous changes heal through osteosclerosis, forming a mixed image of osteolytic and osteosclerotic areas [5].

The most frequent localization of osseous changes in SAPHO syndrome is the anterior thoracic wall; they appear mainly in the region of the sternum bones, ribs, and clavicles [5, 9, 19]. The second most common localization comprises the axial skeleton (initially, the thoracic part, then the lumbar part), with changes affecting chiefly single vertebrae, rarely up to 4 neighboring ones [5, 9, 19]. Changes within the peripheral skeleton or long bones are least frequently observed [5, 9, 10, 19]. The disease called diffuse sclerosing osteomyelitis of the mandible (DSOM) is currently considered to be a manifestation of SAPHO syndrome in the region of the mandibular bone [7, 9, 19]. The mandible is involved in ca. 10% of cases. Usually changes occur unilaterally, mainly in the distal parts of the mandibular body and branch [5, 20]. They are often accompanied with painful edema of the surrounding soft tissues [9]. A change affecting the mandibular bone can be isolated or coexist with osseous manifestations in other parts of the skeleton [4, 7, 11, 16, 20].

The distribution of osseous changes depends on the patient's age. In children, in contrast to adults, CRMO as a manifestation of SAPHO syndrome appears primarily in long bones, and then in the clavicular and spinal region [3, 9, 19].

Many of the osseous changes described above can be observed and evaluated with conventional radiography, which, however, demonstrates low sensitivity in the early stages of SAPHO syndrome [5]. The investigation of choice in evaluating osseous changes is computed tomography (CT). Bone scintigraphy makes it possible to detect areas of increased osseous metabolism and reveals clinically silent changes, which could otherwise remain unnoticed [5, 9]. Magnetic resonance (MR) proves useful in evaluating soft tissue changes and detecting clinically silent changes [5]. Whole-body MR seems especially significant in the context of repeated imaging in the disease course follow-up; its advantage over scintigraphy consists of avoiding ionizing radiation [5, 9].

The results of laboratory investigations in SAPHO syndrome are not distinctive. Typically, elevated values of inflammatory parameters (erythrocyte sedimentation rate [ESR] and acute-phase protein concentration, including C-reactive protein [CRP]) are observed [3, 6, 10]. Blood alkaline phosphatase activity can also be elevated. Neither rheumatoid factor nor anti-nuclear antibodies are present in patients with SAPHO syndrome [6, 10].

Histologically, osseous changes in the course of SAPHO syndrome differ depending on the stage of the disease. Initially, inflammatory infiltrations with predominating neutrophils and strongly expressed subperiosteal reaction are characteristic. With the progression of changes, lymphocytic infiltrations start to predominate, which disappear in later stages. Then, sclerotic trabeculation and marrow cavity fibrosis become visible [5, 9].

The diagnosis seems unproblematic in cases of typical pain localization (e.g. anterior thoracic wall) with corresponding osseous changes visible in radiographic investigation and characteristic cutaneous manifestations. Diagnosing SAPHO syndrome with atypical localization of changes or lack of cutaneous changes is far more challenging. According to Chamot et al. [1] and Benhamou et al. [2], after excluding infectious osteitis, bone neoplasm, and non-inflammatory bone defects, SAPHO syndrome diagnosis requires the fulfillment of one of the criteria enumerated at the beginning of the paper.

It is indispensable to differentiate SAPHO syndrome with infectious osteomyelitis, osteosarcoma, Ewing's sarcoma, other primary and metastatic bone neoplasms, Paget's disease, fibrous dysplasia of bones, eosinophilic granuloma, lymphoma, osteoarticular complications after retinoid group therapy, and also, in children, with juvenile idiopathic arthritis [5, 6, 9].

Apart from clinical examination and history taking, imaging (radiography, CT, MR) and bone scintigraphy are extremely significant in the differential diagnosis. Histopathological investigation does not lead to the diagnosis itself, but it helps to exclude the diseases described above (bone neoplasms, among others). Similarly, microbiological investigation makes it possible to exclude the infectious background of changes.

Because of the rare incidence of SAPHO syndrome, there are no available randomized, controlled clinical studies on the efficacy of particular treatment methods [5, 6, 10]. Therapy is aimed mainly at alleviating the disease symptoms. Nonsteroidal anti-inflammatory drugs (NSAIDs), associated with the smallest complication risk, are administered as the first-line treatment. However, they bring satisfactory results only in some cases [10]. Corticosteroids are recommended mainly in situations when NSAIDs turn out to be insufficient [9, 10]. They lead to a positive response in a considerable group of patients [6, 9, 21], but have the disadvantage of causing significant adverse effects when applied for a long time [9].

In SAPHO syndrome treatment, disease-modifying antirheumatic drugs (DMARDs) are administered, such as methotrexate, sulfasalazine, or azathioprine; nevertheless, their efficacy is questioned by some authors [8–10, 14]. Because of the type of osseous changes occurring in SAPHO syndrome, bisphosphonates (pamidronic acid, zoledronic acid and alendronic acid) seem especially efficient in leading to a long-lasting remission [6, 9, 22–25]. Treatment with TNF-α inhibitors (infliximab, etanercept and adalimumab) should be considered in patients who do not respond to therapy with other medications [5, 8]. In most cases described, administration of TNF-α antagonists turned out to be efficient [9], but in some cases, the TNF-α blockade did not produce the expected results [24]. A patient with Crohn's disease was also described who developed SAPHO syndrome in the course of infliximab therapy [26]. Administering antibiotics seems inefficient and aimless [27], although individual cases have been reported of a positive response to prolonged doxycycline therapy [10, 28].

Surgical treatment, including saucerisation, decortication, or partial or complete resection of the affected bones, is not recommended owing to the low efficiency and high recurrence of changes [9]. Surgical treatment should be considered, however, in cases with severe functional and aesthetic disorders, which become extremely significant when the disease involves the mandibular bone and temporomandibular joints [7, 11, 13, 16, 17]. If ankylosis of the temporomandibular joint is present, condylectomy with the affected bone removal, and joint reconstruction with the use of an alloplastic prosthesis (performed simultaneously or bitemporally) seem to bring the best results [7, 29–31].

Case Report

An 18-year-old patient (S.C.), case record No. 01044/15, was referred to the Department of Maxillofacial Surgery, Wrocław Medical University, Wrocław, Poland, with the initial diagnosis of an intraosseous tumor of the right body and branch of the mandible. For the preceding 2 years, recurrent inflammatory conditions of the right parotid-masseter region had occurred, with coexisting earache, jaw opening difficulty, and face edema and asymmetry (ca. 10 episodes within the previous year). Initially, the patient was treated in a laryngological clinic, and then in a dental clinic; particular episodes of aggravation were treated with antibiotics, with a moderate result. In spite of the administered therapy, progressing facial asymmetry and gradually increasing jaw opening difficulties were observed. Because of the temporomandibular joint dysfunction, the patient had been on a liquid diet for many months. Owing to the disease, the patient had become excluded from ordinary life, as well as had stopped school education and contact with peers. The disease history showed that aggravations occurred mainly after periods of mood depression. As the applied treatment turned out inefficient, the patient was referred to the Maxillofacial Surgery Clinic.

In the center he had stayed at previously, initial imaging diagnostics were carried out. The CT investigation of the facial skeleton and pantomographic radiography revealed considerable thickening and enlargement of the right body and branch of the mandible, as well as mandibular bone remodeling, consisting of the presence of sclerotic areas with numerous osteolytic regions. The changes affected the right half of the mandible, comprising the left part to the area of teeth 33/34. After the initial imaging diagnostics, the patient was referred to the Department of Maxillofacial Surgery, Wroclaw Medical University, Wrocław, Poland, with the above-mentioned diagnosis.

In the clinical examination, facial asymmetry was observed (Fig. 1). Extensive bone distension of the right body, angle, and branch of the mandible could be palpated. There was edema and considerable tenderness on palpation present in the right temporomandibular joint area, with restricted abduction of the mandible (the distance between the crown edges of the incisors equaled 10 mm). No purulent cutaneous fistulas were revealed. Intense acne changes of the face skin were noticed, accompanied by extensive acne scars. Similar changes could be observed on the skin of the back (Fig. 2) and shoulders (Fig. 3). According to the data received from the patient, the persistent massive acne had demonstrated significant resistance to treatment for the previous few years.

The patient did not report dysesthesias referring to the right inferior alveolar nerve, and the exit sites of the trigeminal nerve branches were not painful on palpation.

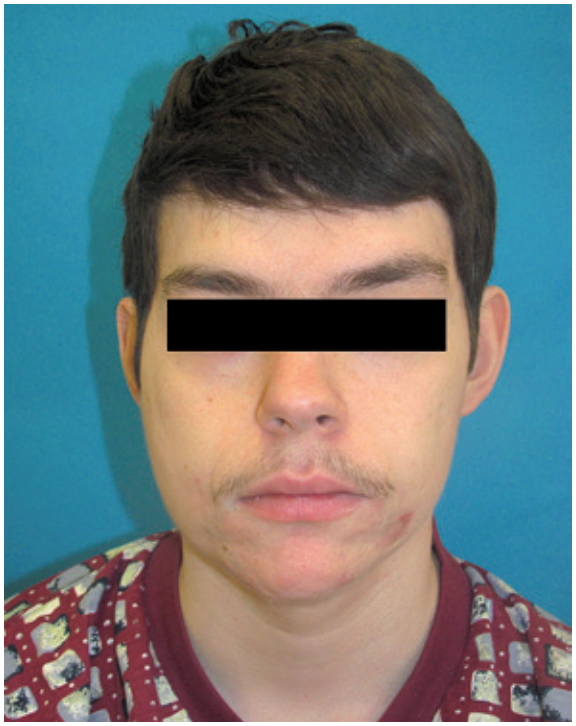

Fig. 1. Patient S.C., aged 18, visible facial asymmetry

In the dental examination, carious defects were noted; the history pointed at difficulties in dental treatment resulting from restricted jaw opening. All teeth in the oral cavity presented positive results in the vitality test. Radiologically, no periapical changes were observed; partly retained teeth 28, 38 and 48 were visualized. The absence of teeth 24 and 45 was revealed; the history made it possible to establish that the teeth had been extracted because of considerable carious destruction and periapical changes. The mucous membrane of the oral cavity was humid, shiny and normally colored, without pathological changes or visible fistulas.

Pantomographic investigation revealed enlarged right body and branch of the mandible, with visible areas of bone structure density increase and reduction; the image suggested chronic mandibular bone inflammation of unknown cause (Fig. 4). In the CT investigation of the facial skeleton and neck, the right branch and body of the mandible were massively thickened and enlarged, and their osseous structure remodeled. Nu-

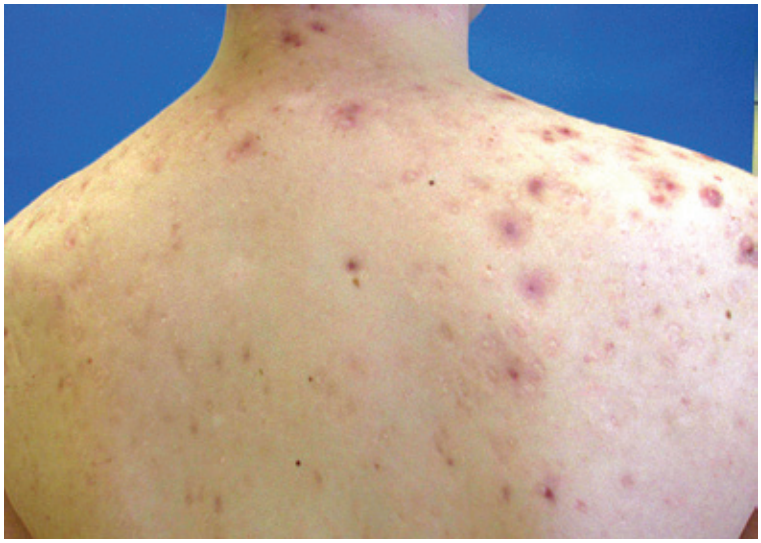

Fig. 2. Intense acne changes of back skin

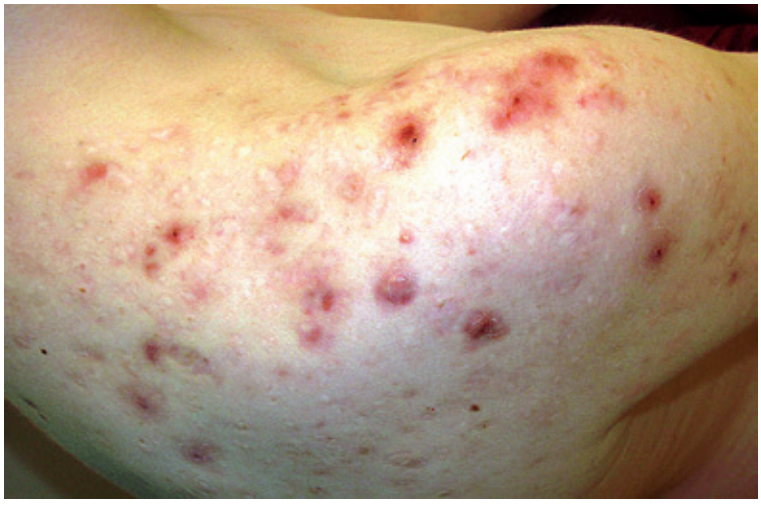

Fig. 3. Intense acne changes of shoulders skin

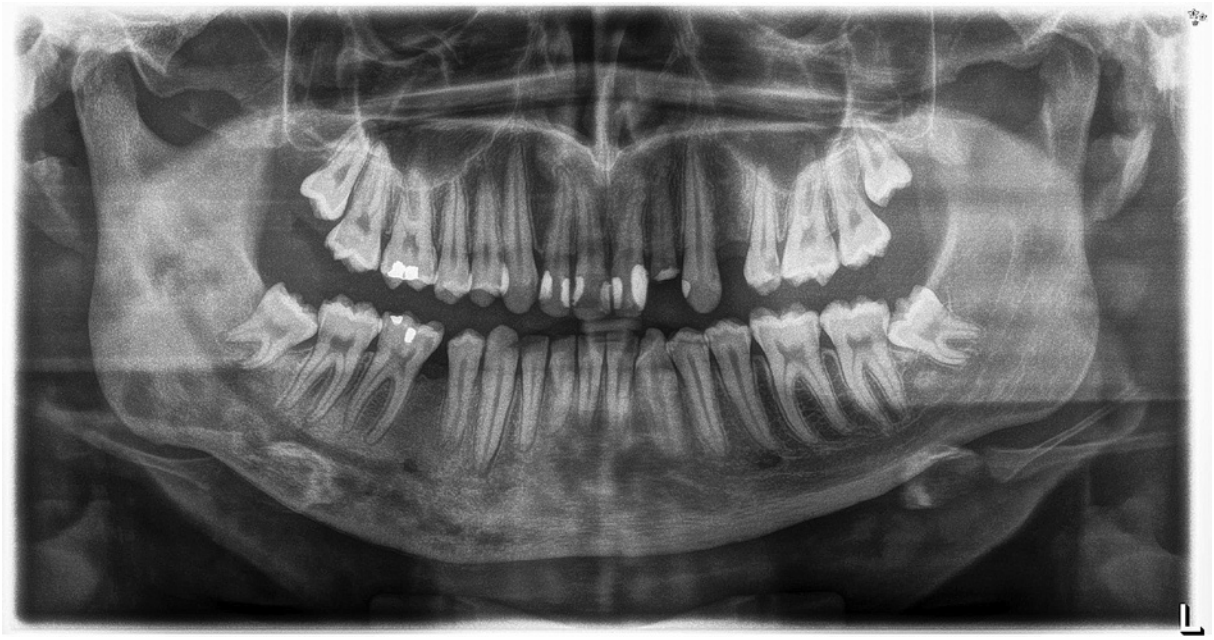

Fig. 4. Pantomographic image with visible areas of bone structure density increase and reduction within the right body, angle, and branch of the mandible

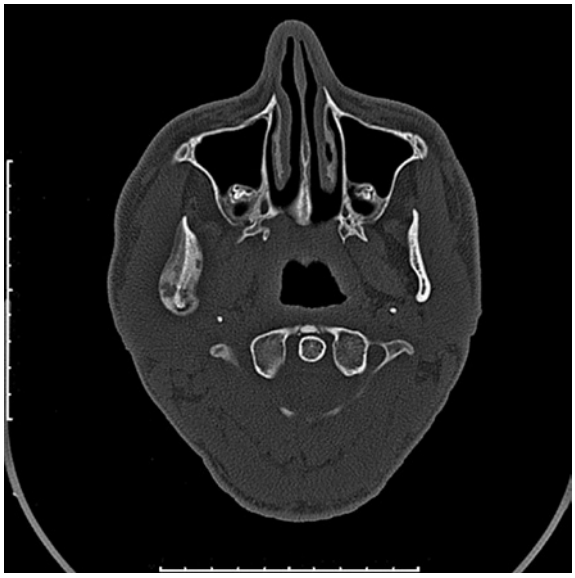

Fig. 5. Computed tomography image. Visible thickening of the right branch of the mandible

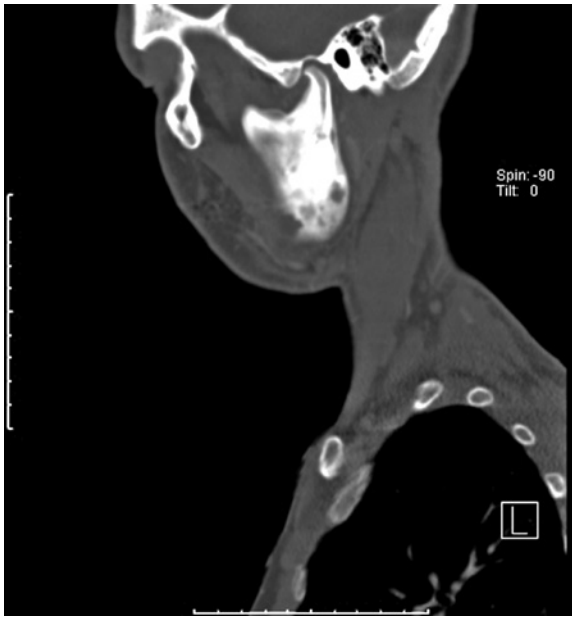

Fig. 6. Computed tomography image of the right body, angle, and branch of the mandible. Visible bone remodeling with mixed areas of osteolysis and osteosclerosis

merous osteosclerotic areas were visualized, with many disseminated osteolytic defects (Fig. 5–7). Moreover, the study showed minor inflammatory osteolytic areas in the right head of the mandible; the space of the temporomandibular joint was preserved. No malignant periosteal reactions were observed. However, edema of the right pterygoid muscles and the masseter muscle, as well as enlarged reactive lymph nodes on the right side of the neck (up to 1 cm in the short axis) were clearly visible. The CT image suggested chronic inflammatory changes of the right mandible part, with the involvement of the right head of the mandible.

In the MR image, similarly as in the CT examination, extensive remodeling of the osseous tissue was visible in the region of the right body and branch of the mandible, which manifested itself as extensive areas of sclerosis of heterogeneous signal, with blurredly delineated minor areas of higher signal in T_2-weighted images and the short tau inversion recovery (STIR) sequence. Also, minor areas of higher signal in T_1-weighted images were present. Extensive bone thickening was observed

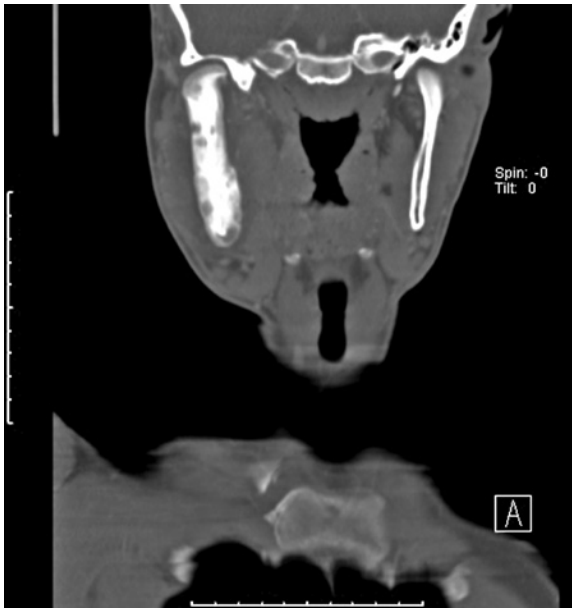

Fig. 7. Computed tomography image of the right body, angle, and branch of the mandible. Visible bone remodeling with mixed areas of osteolysis and osteosclerosis. Note the thickening of the mandibular bone

in the examination, especially in the region of the right angle of the mandible, of maximum thickness of 1.8 cm, as well as extensive infiltrative changes, most likely inflammatory, involving the masseter muscle (with the enlargement of its volume) and the medial pterygoid muscle. Bone scintigraphy (MDP Tc99m) revealed an area of pathologically increased osseous metabolism, comprising the right part of the body and right branch of the mandible. The localization of the increased marker uptake areas corresponded to the changes observed in the CT and MR investigations.

The whole image, i.e. a chronic inflammation of the right half of the mandible, and inflammation of the right temporomandibular joint without a perceptible infectious factor, history of purulent drainage or clear cause of the enumerated complaints, together with the massive, treatment-resistant acne of the face, back and shoulder skin, led the authors to establish the initial diagnosis of SAPHO syndrome.

Treatment with intravenous steroidotherapy (dexamethasone 2 × 4 mg *i.v.*) connected with NSAID (ketoprofen 2 × 100 mg) was applied. This led to a significant local improvement, withdrawal of pain complaints, and lockjaw reduction. Edema of the right temporomandibular joint region and its tenderness on palpation ceased. The patient began to take solid food. Laboratory investigations revealed elevated values of ESR (16.0) and CRP (33.03 mg/l), as well as elevated alkaline phosphatase activity (150 U/l). In order to exclude a malignant neoplasm, it was necessary to take, under local anesthesia, 2 osseous blocks of the right retromolar trigone area for histopathological investigation. Material for the microbiological investigation was collected intraoperatively, which made it possible to exclude an infectious cause of osteitis. During the procedure, the partly-retained tooth 48 was also extracted. The operation itself and the perioperative period proceeded without complications.

The result of the culture for anaerobic bacteria was negative (aseptic), and no microorganisms or leukocytes were observed in the direct specimen. The histopathological investigation of both osseous blocks revealed the presence of marrow cavity fibrosis with moderate chronic inflammatory infiltration. There were no malignant neoplastic lesions in the examined material. The histopathologist confirmed that the histological image of the investigated specimens might correspond with the clinically diagnosed SAPHO syndrome. With the consideration of the whole image, the patient was referred to rheumatologic consultation with the diagnosis of SAPHO syndrome. In accordance with the rheumatologist's recommendation, treatment with sulfasalazine at a dose of 500 mg 2 × 2 tablets, folic acid 5 mg 1 × 1 tablet, and methylprednisolone 16 mg 1 × 1 tablet was administered as an attempt, with a positive result. Currently, the patient remains in the follow-up of a rheumatologic clinic and a university outpatient clinic of maxillofacial surgery. In the 3-month follow-up conducted so far, a considerable reduction of pain complaints and the improvement of the stomatognathic function have been achieved. Since the beginning of the therapy, not a single episode of edema of the right temporomandibular joint region has been observed; also, the range of mandible abduction is constantly increasing (now the distance between the crown edges of the incisors equals 30 mm).

Discussion

In the case discussed in the paper, 2 of Kahn's 4 criteria were fulfilled. Aseptic inflammation of the mandibular bone and the right temporomandibular joint with massive, treatment-resistant acne of the face, back and shoulder skin, associated with hyperostosis in the region of the right body and branch of the mandible, led the authors to the diagnosis of SAPHO syndrome. The imaging investigations revealed signs typical of the later phase of SAPHO syndrome: bone distension and thickening, mixed bone remodeling with areas of osteolysis and osteosclerosis, without osseous sequesters, with histologically confirmed marrow cavity fibrosis. Areas of increased marker uptake

in bone scintigraphy corresponded with the localization of changes visible in the CT, MR, and radiographic examination. The histopathological investigation made it possible to unequivocally exclude neoplastic proliferation of the mandibular bone and fibrous dysplasia, and the microbiological examination confirmed the earlier suspected non-infectious background of changes. The patient's age is characteristic of SAPHO syndrome.

Taking into consideration the whole image of the disease, the established diagnosis seems correct.

The applied treatment with NSAIDs, steroids, and sulfasalazine at the initial stage resulted in significant stomatognathic function improvement, and the reduction of edema and pain complaints.

The aim of further management is to stabilize the signs and stop the progression of changes. What is especially significant is continued evaluation of temporomandibular joint function. The therapy goal is to prevent temporomandibular joint ankylosis, which would necessitate surgical treatment.

In the cases of SAPHO syndrome with the involvement of the temporomandibular joints described so far, various methods of surgical treatment were applied, including partial resection of the articular process of the mandible [13, 17], bilateral high condylectomy [16] and condylectomy with a bone graft [13], as well as condylectomy with temporomandibular joint reconstruction with the use of an alloplastic prosthesis [7, 11]. Currently, the last of the enumerated methods seems to be the management of choice as it brings the most stable and predictable effects [7, 8, 29–31]. This kind of treatment is performed simultaneously or bitemporally [30]. In the latter case, the first operation consists of the ankylotic joint resection. Then, on the basis of the CT investigation data, a 3-dimensional stereolithographic model of the facial part of the cranium is obtained, which makes it possible to prepare an individual alloplastic joint prosthesis and to precisely plan further surgical treatment [7, 30, 31]. The second operation comprises placing the prosthesis in the patient's body [7, 30, 31]. At the same time, fat grafts are applied around the joint, which reduce the risk of reankylosis [7, 8]. In the case of a simultaneous management, the model surgery performed on the 3-dimensional model prepared earlier is reconstructed during the procedure, allowing for prosthesis adaptation [7, 30]. The simultaneous management involves a significant challenge, but it makes it possible reduce the number of surgical interventions under general anesthesia, and eventually lower the treatment costs [29, 30].

In spite of the obvious advantages resulting from treating temporomandibular joint ankylosis with the use of alloplastic prostheses, it should be remembered that there is always a degree of disability associated with the reconstructed joint [29]. Therefore, as long as possible, conservative therapeutic methods should be applied, and the surgical treatment delayed or prevented. In this context, an early diagnosis of SAPHO syndrome is crucial.

The authors emphasize that it is necessary for dentists, including dental and maxillofacial surgeons, to broaden their knowledge of SAPHO syndrome as a possible cause of primarily chronic mandibular bone inflammation and stomatognathic system dysfunction associated with the involvement of temporomandibular joints. The greater awareness of physicians in this area will allow earlier diagnosis in many cases. This, in turn, can make it possible to avoid long-lasting, inefficient therapy with antibiotics, repeated invasive diagnostics, and unnecessary surgical interventions.

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