

Diagnostic Delay in Sternocostoclavicular Hyperostosis: Impact on Various Aspects of Quality of Life

WILLEM A. VAN DER KLOOT,¹ SADHNA A. CHOTKAN,² AD A. KAPTEIN,² AND NEVEEN A. T. HAMDY²

Objective. Sternocostoclavicular hyperostosis (SCCH) is a rare, debilitating, chronic inflammatory disorder of the anterior chest wall due to a chronic sterile osteomyelitis of unknown origin, often associated with characteristic skin lesions of palms and soles: pustulosis palmoplantaris. SCCH goes often unrecognized for years before the diagnosis is established and treatment instituted. The objective of this study was to trace the diagnostic paths of patients with SCCH and to investigate the consequences associated with diagnostic delay.

Methods. Data were collected through structured interviews of 52 patients with a clinically, scintigraphically, and radiologically established diagnosis of SCCH.

Results. The majority of patients presented with swelling and/or pain in the sternocostoclavicular region and/or limited movement of the shoulder girdle. Pustulosis palmoplantaris was present in ~30% of patients. The disease went unrecognized for a median of 3.5 years. Patients were often seen by at least 3 members of the medical profession before the diagnosis was suspected and eventually established. Lack of recognition of the clinical manifestations of the disease and delay in diagnosis were associated with important physical, psychological, and socioeconomic consequences affecting quality of life.

Conclusion. SCCH remains an ill-recognized disease despite its characteristic clinical features. A low level of awareness of the disorder leads to a delay in diagnosis, which has a significant impact on various aspects of quality of life. Awareness should be raised for this disorder, enabling timely diagnosis and initiation of treatment to prevent the irreversible physical and psychological sequelae associated with the protracted untreated state.

INTRODUCTION

Sternocostoclavicular hyperostosis (SCCH) is a chronic, inflammatory disease of the axial skeleton, characterized by a chronic sterile osteomyelitis of the sternum, the medial end of the clavulae, and the upper ribs. Clinical manifestations include local inflammatory changes in the form of redness and swelling in the sternocostoclavicular region (Figures 1A and B) often associated with restricted mobility of the adjacent shoulder joint, which may also be

the sole presenting symptom. The characteristic lesions of pustulosis palmoplantaris (PPP), a chronic, sterile inflammation of the palms and soles (Figure 1C), may develop at any stage in the natural history of the disease in approximately 50% of patients. The spine and mandible may also be affected, leading to the additional clinical manifestations of back pain, limitation of movement of the spine, and pain and limitation of movement of the jaw (1). The disease has a variable natural course, characterized by periods of exacerbation and remission. Despite what may be perceived as mild nonspecific manifestations, the untreated disease is often associated with significant morbidity in the long term due to progressive local sclerosis and hyperostosis and secondary degenerative changes of the sternocostoclavicular joints, with loss of productive years and decreased quality of life.

The diagnosis of SCCH is established on the basis of its characteristic clinical, scintigraphic, and radiologic features (Figure 2) (2–4). The diagnosis is suggested by a focal increase in uptake of the radiopharmaceutical on a technetium bone scintigraphy. The bull head sign (Figure 2A) is characteristic of SCCH, but is not present in all patients, particularly in the early stages of the disease, when hyper-

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¹Willem A. van der Kloot, PhD: Leiden University, Leiden, The Netherlands; ²Sadhna A. Chotkan, MD, Ad A. Kaptein, PhD, Neveen A. T. Hamdy, MD, PhD: Leiden University Medical Center, Leiden, The Netherlands.

Dr. van der Kloot is the chairman of the Dutch Sternocostoclavicular Hyperostosis Patients Association.

Address correspondence to Willem A. van der Kloot, PhD, Institute of Psychology, Leiden University, PO Box 9555, 2300 RB Leiden, The Netherlands. E-mail: w.a.van.der.kloot@umail.leidenuniv.nl.

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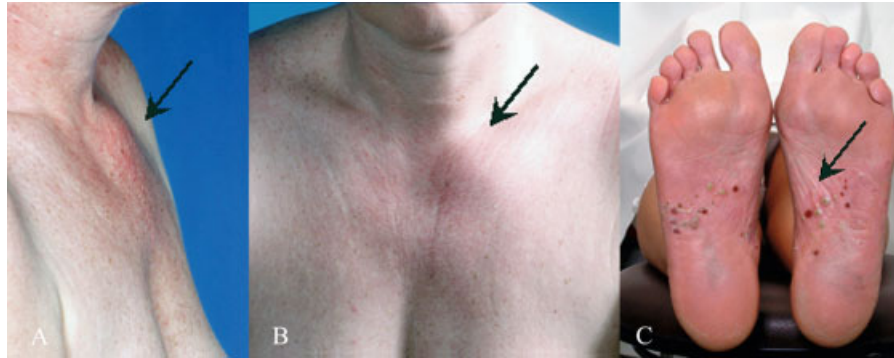


Figure 1. Clinical signs of sternocostoclavicular hyperostosis (SCCH): swelling and redness of the left sternocostoclavicular region (A and B), and the associated skin lesions of pustulosis plantaris found in some 50% of patients with SCCH (C).

ostosis is not prominent. The diagnosis is confirmed radiologically by pathognomonic hyperostosis and sclerosis of the sternum and/or medial ends of the clavulae and first ribs on a computed tomography (CT) scan of the sternocostoclavicular region (Figure 2B). Laboratory mark-

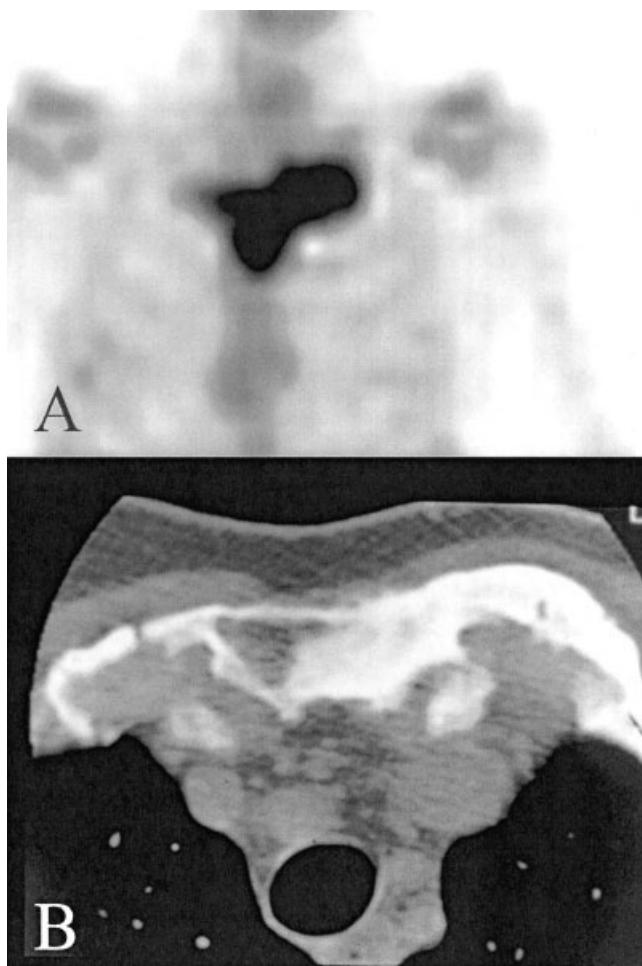


Figure 2. A, Skeletal scintigraphy and B, computed tomography (CT) images of the sternocostoclavicular hyperostosis (SCCH) patient shown in Figure 1. Note the characteristic (half) bull head appearance of the scintigram due to increased uptake in the sternum and the medial end of the left clavicle, with corresponding sclerosis and hyperostosis on the CT image.

ers of inflammation or parameters of bone turnover are rarely abnormal except in the most severe cases. Differential diagnosis includes instability of the joint following trauma, degenerative or inflammatory arthritides, and infection (5). Treatment is aimed at alleviating pain and decreasing the local inflammatory changes, thus preventing changes in bone structure that would lead to abnormal joint apposition and increase the risk of developing secondary degenerative changes. Were SCCH to be left untreated, these secondary degenerative changes represent a significant cause of morbidity in the form of chronic pain and limitation of movement of the shoulder girdle.

The aim of treatment of SCCH is to control the local inflammatory changes, thus preventing the skeletal changes and the secondary degenerative changes. The first line of treatment of SCCH consists of nonsteroidal anti-inflammatory drugs (NSAIDs). Antibiotics and corticosteroids have also been used but are ineffective or have a short-lived effect. Over the last 2 decades, evidence has been accumulating for a beneficial effect of intravenous bisphosphonates (6–11), and there have been a few positive case reports about the use of anti-tumor necrosis factor α (12–14).

SCCH was first described as a separate entity in 1974 by Sonozaki et al (15) in Japan and shortly thereafter in 1975 by Köhler et al (16) in Germany. By the end of 2007 (the year our study was conducted), some 120 articles on SCCH had been published in the international literature. However, it took until 1985 before the first 30 articles had appeared. In 1987, Chamot and colleagues coined the term SAPHO (synovitis, acne, pustulosis, hyperostosis, and osteitis) for a syndrome in which SCCH was associated with generalized joint and skin manifestations (17). Since then, some 300 publications have dealt with the SAPHO syndrome, including cases with SCCH, although no longer as a separate entity. The scarcity of publications related to SCCH is likely to have had an impact on diagnostic delay. This influence is further discussed in the Results section.

A consistent finding among the various publications on SCCH is that it is an ill-known syndrome, which may remain unrecognized for years (2,18–20). Although SCCH is therefore considered to be a rare disorder, it is also clear that it is largely underdiagnosed due to the lack of awareness of treating physicians for the disorder (19–24). Pa-

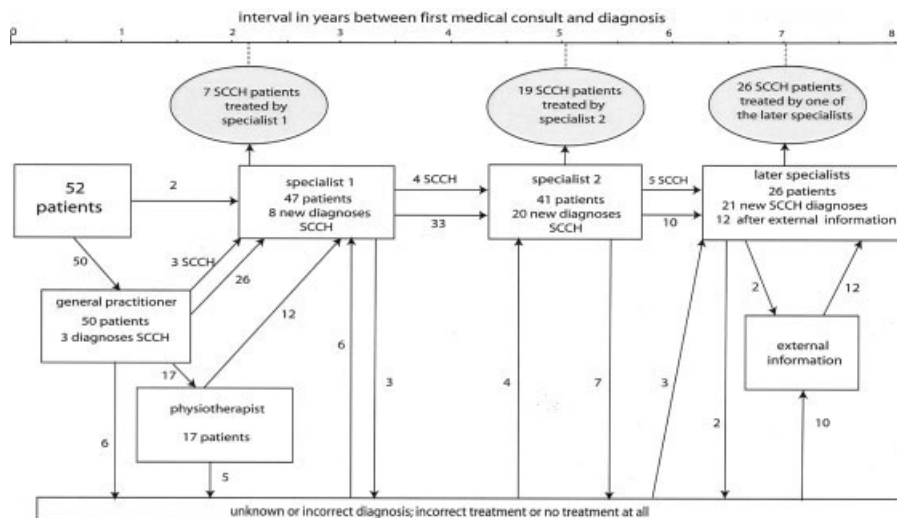


Figure 3. Flow diagram of the trajectories followed by 52 patients from first consultation to establishment of the diagnosis of sternocostoclavicular hyperostosis (SCCH). The definitively diagnosed patients are classified in 3 groups (shaded ellipsoids). Dotted lines indicate the 3 groups' mean durations of diagnostic delay.

tients with this syndrome have therefore been described to go through a “diagnostic odyssey” (21) before a correct diagnosis is made (25–27).

The objective of our study was 2-fold: 1) to examine the diagnostic paths of patients with a definitive diagnosis of SCCH, and 2) to evaluate the impact of the diagnostic delay on various aspects of quality of life.

PATIENTS AND METHODS

The study was initiated and conducted by the Institute of Psychology of Leiden University and followed the ethical standards of the American Psychological Association. The study was partially sponsored by the Dutch SCCH Patients Association (online at: www.scch.nl). Forty-three patients were recruited from the 55 members of this organization. Most patients were under regular clinical control of one of the authors (NATH) at the Department of Endocrinology and Metabolic Diseases of the Leiden University Medical Center (LUMC), a tertiary referral center for patients with SCCH. Nine additional patients who were not members of the Patients Association were directly recruited by one of the authors (NATH) from patients under her control in the LUMC.

Fifty-two patients with a definitive diagnosis of SCCH were interviewed in their homes by 1 of 3 trained interviewers: 2 psychology master students and 1 last-year medical student. We estimated that these 52 respondents encompassed the majority of Dutch patients with a diagnosis of SCCH and were representative of this population. Written informed consent was obtained from all of the patients prior to the start of the interviews.

Interviews were conducted using a structured set of questions on age, sex, age at the time of the first manifestations of SCCH, nature and localization of these manifestations, and details of the diagnostic path followed until the diagnosis of SCCH was definitively established. Responses to the questions were recorded by the interviewers on protocol sheets, and voice recorded if permission for

this was granted prior to the interview. At a later date, responses were digitally coded and double checked with the voice recordings whenever necessary. During the interview, respondents were also asked to complete several questionnaires, including the Dutch language version of the McGill Pain Questionnaire (MPQ-DLV) (28) and the Dutch versions of the Brief Illness Perception Questionnaire (BIPQ) (29) and the Medical Outcomes Study Short Form General Health Survey (SF-20) (30).

The responses to the open questions were categorized by 2 of the authors (WAvdK, SAC). Interobserver agreement was good to perfect, as Cohen's kappas ranged from 0.701 to 1.00 (quartiles: 0.829, 0.943, and 0.977).

RESULTS

The respondent group consisted of 46 women (88.5%) and 6 men (11.5%), ages 24–79 years (mean \pm SD 53.2 \pm 13.75 years, median 56 years). Figure 3 shows the diagnostic pathways followed by our respondents before a definitive diagnosis of SCCH was established.

Diagnostic path. *Symptoms at presentation.* All but one of the 52 patients presented with pain, limitation of movement, or other symptoms in the sternocostoclavicular and/or shoulder region. Pain in the sternum or clavicular region was reported by 13 patients (25%), swelling of the sternum or clavicular region with or without pain by 15 patients (28.8%), anterior chest pain by 7 patients (13.5%), shoulder pain by 10 patients (19.2%), back pain by 2 patients (3.8%), and additional nonspecific symptoms by 5 patients (9.6%). The oldest manifestation dated back 49 years; the most recent one occurred 2 years ago (median 11 years). Age at the time of the first symptom ranged from 15 to 72 years (mean \pm SD 39.0 \pm 14.83 years, median 41.5 years). Twenty-eight patients (53.9%) reported current or past skin manifestations (15 PPP, 2 acne, and 11 unknown). Only 1 patient (1.9%) presented solely with skin problems

(PPP). Eight of the 51 patients who initially presented with bone manifestations simultaneously had signs of PPP. In those cases, the connection between the bone and skin manifestations was not made by the patients or by their physicians.

The general practitioner's approach. Fifty of the 52 patients consulted their general practitioner (GP) with one or more of the above mentioned clinical manifestations after a mean \pm SD of 4.13 ± 8.95 months from the start of symptoms (median 2 weeks, with a minimum of 1 day and a maximum of 3 years). Two patients bypassed their GPs and consulted a specialist directly. The results below concern the action taken by the patient's own GP who was consulted first (some patients changed doctors or consulted one or more substitute GPs).

The patients' GPs suspected a diagnosis of SCCH in only 3 (6%) of 50 cases. In 22 cases (44%), the patients reported that their GP "did not know what was wrong" and did not provide a diagnosis. The remaining patients reported that their GPs suggested "excessive strain" (n = 8 [16%]), "rheumatism, arthrosis, or a bone problem" (n = 5 [10%]), "bursitis or inflammation of the clavicle" (n = 3 [6%]), "psoriasis or furuncle" (n = 2 [4%]), "carpal tunnel syndrome" (n = 1 [2%]), "malignancy" (n = 1 [2%]), "pneumonia" (n = 1 [2%]), or "fibromyalgia" (n = 1 [2%]). In 3 cases (6%), the patient reported that their GP told them that he could find "nothing wrong," that the symptoms were "related to stress," or that they were "imagined."

The actions taken by the GPs consisted of referral to a specialist (n = 16 [32%]), referral to a physiotherapist (n = 8 [16%]), starting medication followed by referral to a specialist (n = 11 [22%]), medication followed by referral to a physiotherapist (n = 9 [18%]), start of medication followed by no further action (n = 3 [6%]), and no action at all, or what was reported by patients as "rejection" (n = 3 [6%]). Medications consisted of NSAIDs (n = 12 [24%]), local corticosteroid injections (n = 6 [12%]), antibiotics (n = 3 [6%]), or unknown pharmaca (n = 2 [4%]).

The specialists to whom the patients were referred belonged to various disciplines: rheumatology (n = 8 [16%]), internal medicine (n = 8 [16%]), orthopedics (n = 6 [12%]), dermatology (n = 2 [4%]), general surgery (n = 1 [2%]), neurology (n = 1 [2%]), or unknown (n = 1 [2%]).

Effects of physiotherapy. Seventeen patients were referred by their GP for physiotherapy. Only 2 patients reported a temporary improvement. Eight patients did not benefit from treatment, and 4 patients reported worsening of their symptoms. Data are not available for the other 3 patients. Twelve of the 17 patients initially referred for physiotherapy were subsequently referred to a specialist.

The first specialist's approach. Of the 50 patients who went to a GP, 39 were referred to a specialist by their own GP (immediately or after the start of medication and/or physiotherapy); 6 patients were referred by a substitute GP, a new GP, or a specialist they were already consulting for a different disease. Two patients bypassed their GPs, consulting a specialist directly. In total, 47 (90.4%) of the 52 patients were thus eventually seen by a specialist, whereas 5 were not referred beyond the care of their GPs (including physiotherapy).

The diagnosis of SCCH was confirmed in 3 cases and

established in 8 additional cases (Figure 3). In the remaining cases, the specialists' diagnosis was "unknown or not supplied" (n = 20 [38.5%]), "rheumatism or arthrosis" (n = 7 [13.5%]), "malignancy" (n = 2 [3.9%]), "bursitis" (n = 2 [3.9%]), "herniated nucleus pulposus of the neck" (n = 2 [3.9%]), "psoriasis" (n = 1 [1.9%]), "polymyalgia rheumatica" (n = 1 [1.9%]), and "old age" (n = 1 [1.9%]).

The 47 patients examined by the first specialist were submitted to a variety of diagnostic procedures: 25 patients (53.2%) had blood tests, 34 (72.3%) had radiographs, 14 (29.8%) had a CT scan, 16 (34%) had a technetium bone scintigraphy (bone scan), 8 (17%) had magnetic resonance imaging (MRI), and 7 (14.9%) underwent a biopsy of the bone swelling. Six patients (12.8%) had both a bone scintigram and a CT scan. In only 1 of those patients, the diagnosis of SCCH was established.

In 25 cases, the first specialists' treatments consisted of NSAIDs (n = 8), local or systemic corticosteroid therapy (n = 9), bisphosphonates (n = 7), or unknown medication (n = 1). Twenty-two patients received no treatment. Thirty-seven of the 47 patients (including 4 patients with a confirmed diagnosis of SCCH and 17 with an "unknown" diagnosis) were eventually referred to a second specialist: a rheumatologist (n = 12), an endocrinologist (n = 11), an orthopedic surgeon (n = 5), a general surgeon (n = 3), an internist (n = 2), a dermatologist (n = 1), a cardiologist (n = 1), and an infectious diseases specialist (n = 1). One additional patient could not recall the field of the specialist to whom she was referred. Three patients were not further referred despite the absence of a diagnosis, with 2 patients not receiving treatment.

The second specialist's approach. Through a variety of paths, 41 patients eventually consulted a second specialist (Figure 3). The diagnosis of SCCH had already been confirmed in 4 patients, and the diagnosis was established de novo in 20 patients. In the remaining 17 patients, the diagnosis was "unknown" (n = 9), "rheumatism or arthrosis" (n = 2), "nothing to find, stress, or imaginary complaints" (n = 2), "bursitis" (n = 1), "psoriasis" (n = 1), "side effects of silicone breast implants" (n = 1), or "varying diagnoses" over time (n = 1). Ten of those 17 patients were referred to a third specialist, 1 was treated symptomatically, and 6 received no medication.

For some of the 41 patients examined by the second specialist, data on the diagnostic tools used by this specialist were missing. For 32 patients, it is known that 23 (71.9%) of them had blood tests, 22 (68.8%) had radiographs, 21 (40.4%) had bone scans, and 8 (25%) had a biopsy. Fifteen (51.2%) of 31 patients had a CT scan and 14 (46.7%) of 30 patients had MRI. Ten (32.3%) of 31 patients had both a bone scintigraphy and a CT scan, and 8 of those patients (80%) were now diagnosed as having SCCH.

The third and later specialist's approach. Thirteen patients with an unknown or incorrect diagnosis eventually consulted a third or subsequent specialist covering various disciplines. A definitive diagnosis of SCCH was ultimately established in 9 of these 13 patients. Ten (76.9%) of the 13 patients had a blood test, 6 (46.2%) had radiographs taken, 6 (46.2%) had a CT scan, 12 (92.3%) had a bone scan, 5 (38.5%) had MRI, and 1 patient (7.7%) had a biopsy. In all

8 patients who had both a bone scintigraphy and a CT scan, the diagnosis of SCCH was established.

Fate of the last group of undiagnosed patients. The 12 remaining patients in whom the diagnosis was still not established after a lengthy round of consultations were eventually also diagnosed after external information became available. Eight patients heard about SCCH by chance through medical acquaintances not involved in the patient's care ($n = 5$) or from friends, relatives, or colleagues who knew a patient with SCCH ($n = 3$). Four patients took independent action such as consulting doctors while on vacation ($n = 2$), searching the Internet ($n = 1$), or writing an open letter to a woman's weekly ($n = 1$).

All of the diagnoses were eventually made by a specialist. With the information obtained from external sources, 7 patients contacted their GPs, who referred 2 of them to an internist, 2 to a rheumatologist, and 3 to the senior author of this article (NATH). This author was personally contacted by 4 other patients without mediation of a physician, and 1 patient was referred to her by another specialist.

Time interval between first consultation and establishment of diagnosis. In our population of SCCH patients, the first diagnosis was established in 1988, 13 years after the first publications appeared in the Western literature on the syndrome. By then, some of the patients had symptoms for more than 10 years, with the longest delay in diagnosis documented in a patient who had symptoms for 36 years before the diagnosis was established. The shortest time interval between the patient's first consultation and establishment of the diagnosis of SCCH was 1 month, with a mean \pm SD diagnostic delay of 5.6 ± 5.9 years and a median delay of 3.5 years. The 25% quartile was exactly 1 year; the 75% quartile was equal to 8.75 years.

There was a significant, albeit small, negative correlation between diagnostic delay and year of diagnosis ($r = -0.289$, $P = 0.037$) (Figure 4A). The number of new diagnoses varied from 0 to 6 (mean \pm SD 2.6 ± 1.7) diagnoses per annum, with the majority of patients with longstanding manifestations diagnosed between 1988 and 2000, and the delay in diagnosis being shorter thereafter.

Figure 4B shows the cumulative frequencies of publications on SCCH and SAPHO syndrome (with or without reference to SCCH) between 1974 and 2007. We see a slow growth of SCCH articles that accelerates a little until 1989 and then tapers off to 1 or 2 publications per year. After the presentation of SAPHO syndrome by Chamot et al in 1987 (17), we see an accelerating growth of SAPHO syndrome articles that presently amount to some 20 publications each year. In the figure, we have indicated the original articles by Sonozaki et al (15), Köhler et al (16), and Chamot et al (17). Although the first article was in English, it was published in a Japanese journal. The latter 2 articles appeared in German and French, respectively, in German and French journals with limited circulation outside these 2 countries. Therefore, those original articles may have been less influential than later work by the same or other authors published in English in more internationally accessible English language journals. We also have marked the appearance, in 1994, of the first Dutch article (31) in the *Nederlands Tijdschrift voor Geneeskunde*, a Dutch

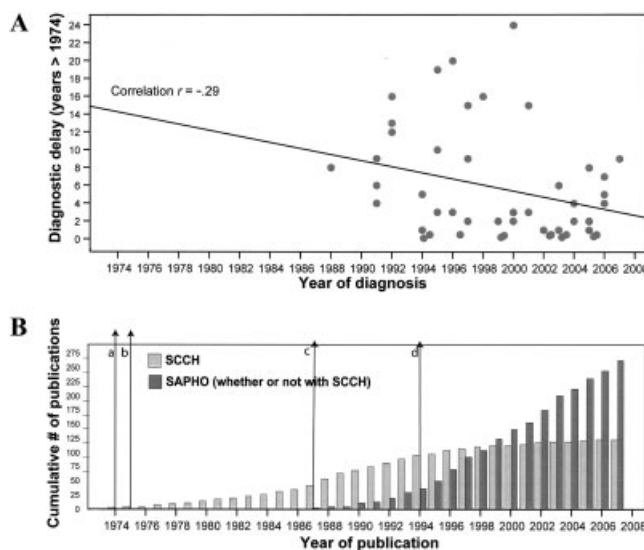


Figure 4. A, Relationship between year of diagnosis and diagnostic delay. B, Cumulative number of publications on sternocostoclavicular hyperostosis (SCCH) and SAPHO (synovitis, acne, pustulosis, hyperostosis, and osteitis syndrome; whether or not including SCCH). a = Sonozaki et al (15); b = Köhler et al (16); c = Chamot et al (17); d = Pais et al (31).

general medicine journal read by virtually all of the doctors in The Netherlands, as well as by many doctors in Belgium. The juxtaposition of Figures 4A and B suggest that what is needed is a certain critical mass in the literature together with one or more key publications before new knowledge is assimilated and awareness is significantly increased.

Impact of diagnostic delay. There was a significant correlation between diagnostic delay and various measures of pain, quality of life, and social functioning after correcting for age ($P < 0.05$, 1-tailed).

Impact of diagnostic delay on pain. The visual analog scales for minimum pain and maximum pain of the MPQ-DLV and the affective pain rating index derived from a set of pain-describing adjectives in the same instrument were positively correlated with the duration of the interval between the first manifestations and diagnosis ($r_p = 0.383$, $P = 0.003$; $r_p = 0.236$, $P = 0.047$; and $r_p = 0.251$, $P = 0.038$, respectively).

Psychological impact of diagnostic delay. An important psychological trauma reported by two-thirds of the patients was the perception that they had not been taken seriously by their treating physician(s). Eight patients (15.4%) felt that this had often been the case. Seventeen respondents (33%) had experienced the response of their GPs to their problem as neglect (their symptoms being ignored or dismissed) or rejection.

A longer delay in diagnosis was also associated with higher emotional distress, as measured by the BIPQ scales emotions, a scale summarizing illness-induced anger, fear, distress, and dejection ($r_p = 0.254$, $P = 0.036$), and consequences, i.e., the perceived negative influence of the illness on one's life ($r_p = 0.266$, $P = 0.030$).

Patients who had to wait longer for the correct diagnosis

to be established reported more health-related impairment of their social activities, such as visits to friends and family, and more impairment of their ability to fulfill activities required in their homes or by their employment. After correcting for age, there was a negative correlation between diagnostic delay and the SF-20 scales role fulfillment (i.e., the absence of health-related impairment of ability to work or conduct housekeeping chores; $r_P = -0.315$, $P = 0.012$) and social functioning (i.e., the absence of health-related impairment of social activities; $r_P = -0.280$, $P = 0.025$).

Socioeconomic impact of diagnostic delay. Several findings indicate that in SCCH, diagnostic delay may also have socioeconomic effects and an impact on a patient's income. Of the 52 respondents, 10 (19.2%) had retired, 19 (36.5%) were in full- or part-time employment, 10 (19.2%) were unemployed, and 13 (25%) were receiving social benefits for permanent disability (regulated by national law). After excluding retired patients and correcting for age, there was a significant difference ($P = 0.0002$) in the duration of the diagnostic delay between employed and unemployed patients, the latter group including the permanently disabled patients. The means of the diagnostic delay were 84.3 months and 31.0 months, respectively, for the unemployed and employed patients.

The relationship between delay and perceived role fulfillment was corroborated by the finding that in the 32 respondents who were employed, those who had experienced shorter periods of diagnostic delay worked more hours per week, as shown by the significant negative correlation, after correcting for age, between duration of the diagnostic delay and number of hours worked per week ($r_P = -0.463$, $P = 0.004$).

DISCUSSION

In a recent and elaborate report by the European Organisation for Rare Diseases (EURORDIS), based on the responses of 12,000 patients with 16 different rare diseases, it was documented that approximately 25% of the patients had to wait for more than 3 years until the correct diagnosis was established, 41% were initially misdiagnosed, 7% were told that their symptoms were psychological or psychiatric, 18% experienced rejection by at least 1 health care professional, and 18% sought answers on their own to arrive at the correct diagnoses (32,33). The results of the present study match this picture to a surprising degree: 40% of our patients were initially misdiagnosed, 4% were told that their problems were psychological, 35% had felt rejected by doctors or nursing staff, and in 23%, the correct diagnosis was only established after the patients obtained information from sources outside their regular medical circuit. Compared with the EURORDIS data, diagnostic delay was more serious in the case of SCCH because 50% had to wait between 3.5 and 36 years for a correct diagnosis.

It is perhaps not surprising that a rare disorder such as SCCH, which usually presents with nonspecific manifestations, may not be readily recognized by members of the medical profession who have not come across the syndrome before. It is unfortunate, however, that in 50% of

cases it took more than 3.5 years, and that in 25% of cases the diagnostic delay exceeded 8 years, with a dramatic delay of 24 years incurred by one patient, taking into consideration that the first publications on the syndrome only appeared in the Western literature in the mid-1970s.

Our data confirm the findings from previous publications acknowledging the difficulties encountered in the diagnosis of this syndrome and the fact that it may go unrecognized for years. Our data additionally demonstrate that the sometimes substantial delays in diagnosis have important psychological and socioeconomic consequences, which are reflected in a decrease in quality of life, although the causal mechanism for this is probably complex. Lack of recognition of the syndrome and delay in diagnosis has meant that in keeping with previous reports, some of our patients also went through a diagnostic odyssey (21), having to be subjected to a range of medical procedures, including surgery, before they were correctly diagnosed and adequate treatment could be initiated. SCCH remains an ill-recognized disease despite its characteristic clinical features. A low level of awareness of the disorder leads to a delay in diagnosis, which has a significant impact on various aspects of quality of life. Awareness should be raised for this disorder, enabling timely diagnosis and initiation of treatment to prevent the irreversible physical and psychological sequelae associated with the protracted untreated state.

More publications on SCCH would certainly help disseminate more knowledge about this rare disorder. Increasing awareness of its manifestations would lead to standard inclusion in the differential diagnosis of conditions affecting the sternoclavicular joint characterized by pain and swelling of the sternocostoclavicular region (5). In the ideal case, we envisage medical guidelines in which SCCH is incorporated.

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AUTHOR CONTRIBUTIONS

All authors were involved in drafting the article or revising it critically for important intellectual content, and all authors approved the final version to be submitted for publication. Dr. van der Kloot had full access to all of the data in the study and takes responsibility for the integrity of the data and the accuracy of the data analysis.

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