

Invited Review Paper  
TMJ Disorders

# Synovial chondromatosis of the temporomandibular joint: a case description with systematic literature review

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*L. Guarda-Nardini, F. Piccotti, G. Ferronato, D. Manfredini: Synovial chondromatosis of the temporomandibular joint: a case description with systematic literature review. Int. J. Oral Maxillofac. Surg. 2010; 39: 745–755. © 2010 International Association of Oral and Maxillofacial Surgeons. Published by Elsevier Ltd. All rights reserved.*

**Abstract.** Synovial chondromatosis (SC) of the temporomandibular joint (TMJ) is a rare disease characterized by the presence of calcified loose bodies within the joint, and few systematically gathered data are available about its epidemiology. The aim of this paper was to describe a case of SC of the TMJ, and to carry out a systematic review of the literature on epidemiology over the past decade. A case of a 53-year-old female with the classical triad of signs and symptoms of SC (pain, swelling, restricted mouth opening) is described. A systematic search in the National Library of Medicine's PubMed Database was performed. 155 cases were described in 103 publications. Most dealt with single case reports. Females are affected more than males with a 2.5:1 ratio and the mean age of patients was about 46 years. Late diagnosis is common and in most cases more than 2 years elapsed between symptom onset and surgical intervention. Open TMJ surgery is the treatment of choice, since less invasive techniques, such as arthroscopy, allowed complete removal of the loose bodies only in about half of cases. A single recurrence was described, confirming the benign nature of the disease.

**Key words:** temporomandibular joint; synovial chondromatosis; systematic review; TMJ surgery.

Accepted for publication 26 March 2010

Synovial chondromatosis (SC) is a cartilaginous metaplasia of the mesenchymal remnants of the synovial tissue of the joints. Its main characteristic is the formation of cartilaginous nodules that may be pedunculated and/or detach from the synovial membrane, becoming loose bodies within the joint space<sup>36</sup>. The aetiology of SC is not fully understood and cases are divided into primary SC (without identifiable aetiological factors)

and secondary SC (with a known aetiology). Secondary cases are often thought to be related to previous trauma, repetitive microtrauma or degenerative arthritis. Primary cases cannot be associated with any supposed aetiological factors and are considered to be more aggressive<sup>23</sup>.

SC usually affects large synovial joints, such as the elbow and knee, and it is uncommon in the temporomandibular

joint (TMJ)<sup>30</sup>. The main clinical symptoms are preauricular swelling, pain, limitation of jaw movement and crepitus joint sounds. The diagnosis is made by computerized tomography (CT) and magnetic resonance imaging (MRI)<sup>22</sup>. Surgery is the therapeutic choice to remove nodules and loose bodies from the joint space. Several approaches have been described, depending on the extension of the lesion with respect to the cranial fossa<sup>10</sup>.

The first reported case of SC of the TMJ was in 1933, when Axhausen reported metaplastic chondrogenesis in the synovial membrane<sup>4</sup>. After that, reports were characterized by inconsistencies of nomenclature, until the 1980s when the term 'SC' was adopted<sup>32,40,54</sup>. A literature review of the period up to 1997 showed that 74 cases of SC of the TMJ were described, mainly as isolated case reports<sup>55</sup>. More recent studies have attempted to summarize the literature data<sup>3,30,58</sup>, but none has satisfied the need for a systematic assessment, and the findings are inconsistent regarding the total number of SC cases.

The aim of the present paper is to describe a case of SC of the TMJ and to carry out a systematic review of the literature over the past decade to update the epidemiological data.

### Case report

A 53-year-old woman was referred because of severely restricted mouth opening and pain in the TMJ area. She presented a slight swelling in the right preauricular area, she was unable to open her mouth more than 20 mm and the midline of the mandible was deflected slightly to the right side (Fig. 1). Symptoms had been present for about 1 year before the appointment and the restriction of mouth opening had worsened progressively. For about 6 months, the patient had worn an oral appliance prescribed by a general dental practitioner on the basis of an



Fig. 1. Preoperative mouth opening with deflection on the affected side and slight preauricular swelling.

unspecified diagnosis of 'craniomandibular dysfunction' without experiencing any improvement. After that, the family physician had treated her with anti-inflammatory drugs (ketoprofene 80 mg, twice a day for 7 days) but her symptoms had remain unchanged and she was then referred to the authors' tertiary centre. During history taking, she denied any past trauma or diagnosis of any rheumatological conditions. Physical examination revealed that, despite the preauricular swelling, neither dysfunction of the facial nerve nor hearing disturbances were present. Joint sounds on the right side were absent and the contralateral joint was negative on palpation. An orthopantomogram seemed to reveal one small calcification around the condyle, so further studies using imaging techniques were requested.

MRI showed an hypoechoic area located anteriorly to the condyle in the closed mouth position and located posteriorly to the condyle in the open mouth position (Fig. 2). CT confirmed the presence of at least two loose bodies within the right TMJ and the diagnostic hypothesis of SC was made. The patient was scheduled for surgery.

A classical surgical approach to the TMJ was chosen owing to the impossibility of removing wide loose bodies by means of arthroscopy alone. Under general anaesthesia, the right TMJ was exposed using a preauricular incision. Immediately after opening the joint capsule, some transparent and viscous fluid came out of the joint space. A large calcified loose body was present in the upper

joint space and was removed (Fig. 3), another fibrous micronodular loose body was found around the condyle (Fig. 4). Two large loose bodies were detected within the TMJ, in contrast to the large number of small bodies described in other cases (Fig. 5). Synovectomy was performed to allow clearance of the joint compartment from any potential residual of metaplastic activity. Post-surgical histological examination revealed chondrometaplasia of the synovial membrane and confirmed the diagnosis of SC of the right TMJ, classified as third stage according to Milgram's classification (Fig. 6)<sup>36</sup>.

The postoperative course was uneventful and only painkillers and antibiotics were prescribed. The day after surgery, a postoperative mouth opening of 28 mm was recorded, with no motor deficit on the right side of the face. Vigorous physiotherapy was started 1 week before surgery, and follow-up assessments were scheduled at 1 week, 1 month, 3 months, 6 months and 1 year. At the 1 year appointment, a maximum mouth opening of 43 mm was recorded, with no signs of recurrence (Fig. 7).

### Literature review

On 30 July 2009 a systematic search in the National Library of Medicine's PubMed Database was performed to identify all peer-review papers in the English literature dealing with cases of SC of the TMJ. The studies included for discussion in the review were clinical studies describing surgical interventions performed on one or more new cases of SC of the TMJ with

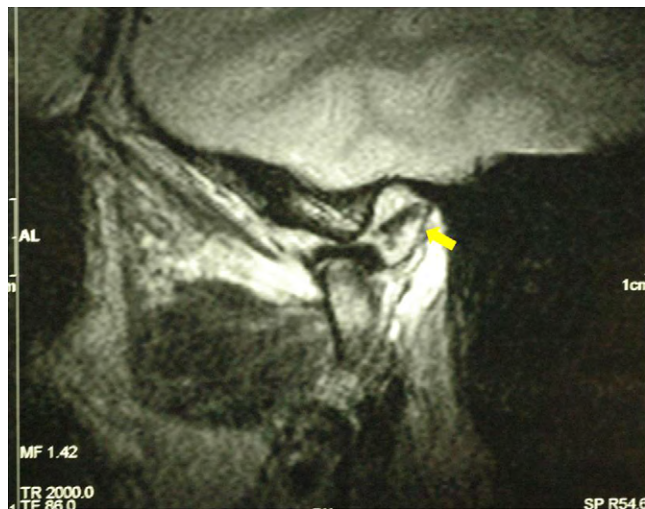


Fig. 2. MRI revealing a large hypoechoic area (yellow arrow) in the posterosuperior compartment of the right TMJ. (For interpretation of the references to color in this figure legend, the reader is referred to the web version of the article.)

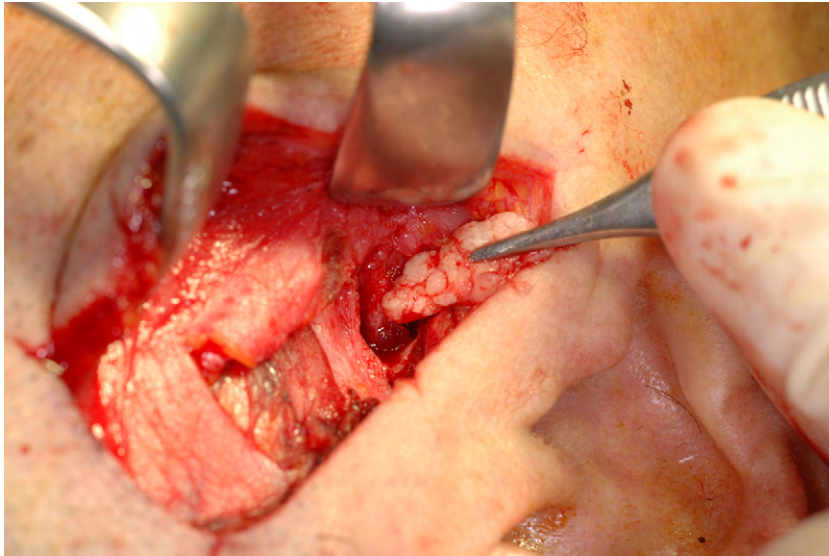


Fig. 3. Removal of a large calcified loose body from the joint space.

respect to the last comprehensive review carried out by VON LINDERN et al.<sup>55</sup>.

The search strategy comprised four steps: a search based on the Medical Subjects Headings (MeSH) of the PubMed database; a word terms search within the PubMed database; a search within PubMed related articles to find the selected ones; a search within the references lists of the selected articles.

The following MeSH terms were used to identify a list of potential papers to be included in the review.

- TMJ: an articulation between the condyle of the mandible and the articular

tubercle of the temporal bone. Year introduced: 1997.

- SC: rare, benign, chronic, progressive metaplasia in which cartilage is formed in the synovial membranes of joints, tendons sheaths, or bursae. Some of the metaplastic foci can become detached producing loose bodies. When the loose bodies undergo secondary calcification, the condition is called synovial osteochondromatosis. Year introduced: 1990 (previous indexing: chondroma, 1966–1989).

The search was limited to papers in the English language published later than 01/



Fig. 4. Removal of a second large loose body, which had a fibrous micronodular aspect.

01/1998 and the combination of the two MeSH terms allowed the identification of 23 citations, the abstracts of which were read to select articles to be retrieved in full text. After abstract reading, 12 papers were selected for inclusion in the review.

In the second step of the literature search the combined word terms 'TMJ' and 'SC' were used to identify other potential papers to be included in the review. Limits were set as described above. This search strategy provided a list of 49 new citations, the abstracts of which were read to select articles to be retrieved in full text. After abstract reading, 30 additional papers were selected for inclusion in the review.

A search within the PubMed related articles for each of the included papers and a hand-made search within the references lists of the included papers were performed, but no additional relevant papers were identified.

A total of 42 papers were selected for inclusion and discussion in this review on the basis of abstract reading. Retrieval of full texts was possible for 38 papers, while in four cases the full text of the paper was not retrieved because the authors were unavailable because of address changes or did not respond to the authors' request.

For each of the included studies, the following data were recorded for discussion: sample size; age of the patient at the time of surgical intervention; male-to-female ratio; right-to-left joint ratio; clinical signs and symptoms; duration of symptoms before intervention; diagnostic imaging technique; type of surgical technique; number of loose bodies extracted from the joint space; follow-up period at the time of last observation after intervention; presence/absence of recurrence (Table 1). Papers available only in abstract form were included in the table to provide as much comprehensive data as possible on the prevalence and demographic features of the disease; most of them provided sufficient information but little information on the other features of the cases was available, and is thus missing from the table.

## Results

### Number of cases and demographic features

The selected studies accounted for 80 new cases of SC of the TMJ. Only 8 studies described more than one case, and 2 of them described only two cases of SC. The age of the patients ranged between 12 and 81 years in the 38 studies for which age data were available (77 cases from 38 full texts and



Fig. 5. This case of SC was characterized by the presence of only two large loose bodies.

one abstract), with an approximate mean age of 46.3 years. Only one case of a patient under 18 years affected by SC was described. Females are affected almost three times more than males, with a ratio of 56:20 (76 cases from 38 full text papers). The right and left TMJs seem to be affected equally; 40 cases to the right and 37 to the left joint (76 cases from 38 full text papers). Only a single new case of bilateral involvement was reported, even if surgical data are available only for one joint.

**Signs and symptoms**

The three cardinal signs and symptoms of SC of the TMJ were confirmed to be pain

in the preauricular area (69 out of 76 cases for which data are available), swelling, facial asymmetry or joint deformity (62/76 cases) and impairment in jaw function (49/76). Other less frequent symptoms were, in order of prevalence: occlusal changes, joint sounds, trismus, headache, sensory disturbances, facial nerve palsy.

The duration of symptoms before intervention ranged from 1 month to 15 years; the time span between symptom onset and surgery was reported in only 64 cases (26 studies). 20 of the 26 studies that provided such data described cases with symptoms lasting from 2 years or more before correct diagnosis was made and surgery was performed.

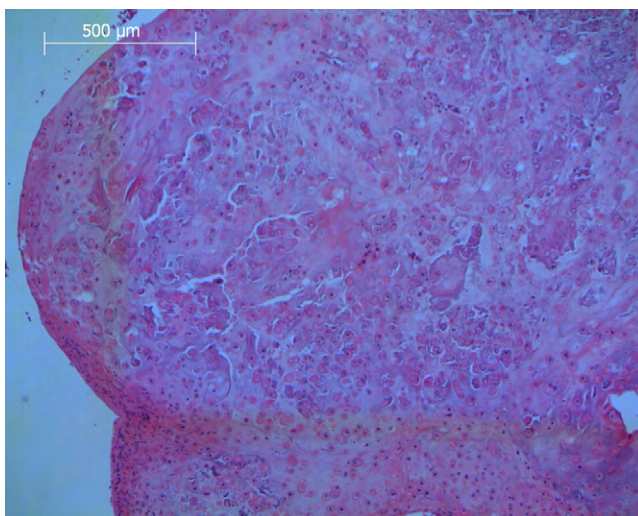


Fig. 6. Histopathological findings showing chondrometaplasia of the synovial membrane.



Fig. 7. Mouth opening at the 1 year follow up.

**Diagnostic imaging techniques**

The type of imaging techniques used for diagnosis and surgical planning was specified for 74 cases. CT and MRI were the most common techniques, being used in 56 and 54 cases, respectively. Orthopantomography was used additionally in 31 cases. Other less frequently used techniques were arthrography (4 cases), cone-beam CT (3 cases) and sagittal tomography (2 cases).

**Surgical techniques**

Most studies described open surgery interventions, which were performed in 63/76 cases. Five of those cases were descriptions of computer-assisted surgery. Synovectomy was the most frequent procedure associated with loose body removal from the joint space, often accompanied by discectomy and, less frequently, by condylectomy. Precise data on the frequency with which these procedures were performed could not be gathered due to the paucity of studies that provided descriptions of the surgical technique. In a few cases (4/76) an intracranial extension was described and repair of the cranial fossa was needed. Arthroscopy was used in 11/76 cases, followed by open surgery in 4 cases to allow removal of all loose bodies and declared by the authors to be only partially successful at removing loose bodies in one case, thus accounting for a 6/11 (55%) success rate for arthroscopy when used alone. Arthrocentesis was used in one case, while in one case the type of surgical intervention was not specified.

Table 1. Summary of data from the studies included in the review. *Legends:* F = female; M = male; R = right; L = left; MIO = mouth interincisal opening; MR = magnetic resonance; CT = computerized tomography; AR = arthrography; OPT = orthopantomography; CBCT = cone-beam computerized tomography; ST = sagittal tomography; N.S.: = not specified.

Study's first author and year	Sample size	Age	F/M ratio	R/L joint	Clinical signs and symptoms	Duration of signs and symptoms	Diagnostic imaging technique	Surgical technique	Nr. of loose bodies	Follow-up span (months)	Recurrence
CAI, 2009 <sup>8</sup>	1	21	1 F	1 L	Pain, reduced MIO, click, concurrent pigmented villonodular synovitis	N.S.	MR	Arthroscopy	Multiple (unspecified)	13	No
PENG, 2009 <sup>42</sup>	1	58	1 F	1 R + L	Slight diffuse swelling, reduced MIO, preauricular tenderness	N.S.	OPT, CT	Open surgery with diskectomy	Multiple (unspecified)	N.S.	N.S.
ZHA, 2009 <sup>59</sup>	1	44	1 M	1 L	Crepitation, deflection, hard 3.5 cm mass in the preauricular area	3 years	CT, MR	Open surgery with no diskectomy or condylectomy	Multiple (90–95)	60	No
ADACHI, 2008 <sup>2</sup>	1	25	1 F	1 L	Trismus, TMJ pain exacerbated with function	9 months	N.S.	Arthrocentesis	Multiple (unspecified)	N.S.	N.S.
FERNANDEZ-SANROMAN, 2008 <sup>13</sup>	5	34–56	3 F, 2 M	3 R, 2 L	Pain (5/5 cases), occlusal changes (4/5), swelling (3/5), dysfunction (3/5)	34–48 months	OPT, MR, CT	Arthroscopy (3) or open surgery (2)	Multiple (unspecified)	16–108	No
HOHLWEG-MAJERT, 2008 <sup>22</sup>	5	12–66	4 F, 1 M	1 R, 4 L	Preauricular swelling and pain in 4/5 cases, only pain in 1 case	1–36 months	OPT (1) MR (4) CT (3) CBCT (2)	Computer-assisted surgery	Multiple (unspecified)	0–132	No
HONDA, 2008 <sup>24</sup>	1	35	1 F	1 R	TMJ pain and reduced MIO	10 years	MR, AR, CBCT	Arthroscopy (only with partial removal of loose bodies)	Multiple (15)	12	No
KADEMANI, 2008 <sup>27</sup>	1	55	1 F	1 R	Preauricular swelling, open bite on the affected side, headache	N.S.	OPT, CT	Open surgery with condyloplasty e glenoid fossa repair	Multiple (unspecified)	N.S.	N.S.
SEMBRONIO, 2008 <sup>49</sup>	1	28	1 F	1 R	Pain and swelling	N.S.	MR	Arthroscopy plus open surgery to allow removal of all loose bodies	Multiple (unspecified)	N.S.	N.S.
YOKOTA, 2008 <sup>58</sup>	1	52	1 M	1 R	Pain, swelling, difficulties to open the mouth	N.S.	CT, MR	Combined transzygomatic temporal skull base and preauricular approach (condylectomy and cranial fossa repair)	Multiple (unspecified)	N.S.	No
ACAR, 2007 <sup>*.1</sup>	1	72	–	–	–	–	–	–	–	–	–
BALLIU, 2007 <sup>6</sup>	1	31	1 M	1 R	Painful swelling	N.S.	CT, MR	Open surgery	Single mass (cartilagineous nodules surrounded by synovia)	0	No

Table 1 (Continued)

Study's first author and year	Sample size	Age	F/M ratio	R/L joint	Clinical signs and symptoms	Duration of signs and symptoms	Diagnostic imaging technique	Surgical technique	Nr. of loose bodies	Follow-up span (months)	Recurrence
D'SOUZA, 2007 <sup>11</sup>	1	63	1 F	1 L	Pain, swelling, crepitus, history of past unresolutive interventions	10 years	CT	2 Open surgeries, the second with synovectomy, diskectomy and interposition of dermis-fat graft (programmed for a condylar resection)	Multiple (unspecified)	~72	Yes
LIEGER, 2007 <sup>30</sup>	1	42	1 F	1 R	Pain on function, slight deflection on mouth opening	5 years	OPT, MR, CT	Open surgery with synovectomy	Multiple (unspecified)	9	No
MANDRIOLI, 2007 <sup>*,33</sup>	1	–	–	–	–	–	–	–	–	–	–
REYES MACIAS, 2007 <sup>45</sup>	1	54	1 F	1 R	Pain on jaw movement, deformity of the TMJ	5 years	N.S.	Open surgery	Multiple (unspecified)	8	No (infiltration with steroids to control pain after 1 month)
XU, 2007 <sup>56</sup>	1	49	1 F	1 L	Pain, headache, swelling	7 years	MR, CT	Open surgery	Multiple (unspecified)	24	No
HAMMODEH, 2006 <sup>*,20</sup>	1	–	–	–	–	–	–	–	–	–	–
HUH, 2006 <sup>25</sup>	1	74	1 M	1 L	Swelling with intermittent pain	2 years	OPT, CT, MR	Open surgery with high condylectomy	Multiple (unspecified)	N.S.	N.S.
ARDEKIAN, 2005 <sup>3</sup>	11	19–72	8 F, 3 M	6 R, 5 L	TMJ pain (10/11 cases), swelling (8/11), reduced MIO (3/11)	1–180 months	OPT, CT, MR	Open surgery with synovectomy (11/11) and diskectomy (7/11) with temporalis flap (6/11)	Multiple in 7/11 cases, no in 4/11 (diagnosis confirmed by histologic examination)	12–120	No (one case of ortho surgery needed to correct occlusion after SC)
HAMILTON, 2005 <sup>19</sup>	1	66	1 F	1 L	Asymptomatic preauricular mass	2 years	CT, MR	Unsuccessful aspiration followed by open surgery	Multiple (unspecified)	N.S.	N.S.
MARTIN-GRANIZO, 2005 <sup>34</sup>	1	49	1 F	1 L	Pain, swelling, joint sounds	6 months	CT, MR	Open surgery after unsuccessful arthroscopy	Multiple (over 200)	12	No
MUPPARAPU, 2005 <sup>39</sup>	1	65	1 F	1 R	Premature dental contact in the left and open bite in the right side	18 months	CT, MR	Open surgery with steel plate to repair cranial perforation	Multiple (unspecified)	3	No
SARLANI, 2004 <sup>47</sup>	1	35	1 F	1 L	Limited MIO, asymmetry of the TMJ	7 years	MR	Open surgery with partial parotidectomy	Multiple (109)	6	No
HOLMLUND, 2003 <sup>23</sup>	9	24–81	7 F, 2 M	6 R, 3 L	TMJ pain and impaired mandibular function	8–72 months	OPT (9/9 cases), MR (4/9), CT (3/9), AR (1/9)	Open surgery	Multiple (3–80)	12–194	No

ISHII, 2003 <sup>26</sup>	1	38	1 F	1 L	Pain, swelling, reduced MIO	3 years	CT	Open surgery, calcification of the lateral pterygoid muscle left in place to avoid excessive bleeding	Multiple (unspecified)	120	No
ROOTKIN-GRAY, 2003 <sup>46</sup>	1	56	1 M	1 R	Pain, swelling, trismus	N.S.	ST	Open surgery	Multiple (110)	18	No
SHIBUYA, 2003 <sup>50</sup>	1	51	1 F	1 R	Pain, reduced MIO	N.S.	ST	Open surgery with diskectomy	Multiple (unspecified)	24	No
AYDIN, 2002 <sup>5</sup>	1	39	1 M	1 L	Intermittent pain, mass over the zygomatic arch	3 years	CT, MR	Open surgery with disk repositioning	Multiple (unspecified)	24	No
SHIBUYA, 2002 <sup>52</sup>	1	57	1 F	1 R	Pain, reduced MIO, mandibular deflection to the affected side during movement	2 years	MR, AR	Arthroscopy	Multiple (unspecified)	10	No
VON LINDERN, 2002 <sup>55</sup>	8	26–71	5 F, 3 M	4 R, 4 L	Typical symptoms not present in all cases	N.S.	CT, MR	Open surgery with synovectomy and diskectomy	Multiple (unspecified)	N.S.	N.S.
GAY-ESCODA, 2001 <sup>14</sup>	1	42	1 M	1 L	Pain that increase with movements	3 years	CT	Open surgery with partial synovectomy and remodeling	Multiple (4)	60	No
KOYAMA, 2001 <sup>29</sup>	2	47	2 F	2 L	Pain, mandibular deviation toward the other side at rest	N.S.	CT	Open surgery with diskectomy	Multiple (up to 95)	N.S.	N.S.
YILDIZ, 2001 <sup>57</sup>	1	39	1 M	1 L	Facial asymmetry, mobile perauricular mass	4 years	MR, CT	Open surgery	Multiple (unspecified)	N.S.	N.S.
MIYAMOTO, 2000 <sup>37</sup>	2	21–37	2 F	2 R	Pain, reduced MIO, deflection on the affected side during mouth opening	15–120 months	MR (1 case), CT (1 case)	Arthroscopy (1 case) or open surgery (1 case)	Multiple (2–20)	10–15	No
PETITO, 2000 <sup>43</sup>	4	31–63	4 F	3 R 1 L	Pain, limited MIO, swelling	1–24 months	CT (2/4 cases), MR (1/4), AR (1/4)	Open surgery	Multiple (unspecified)	0–36	No
REDDY, 2000 <sup>44</sup>	1	64	1 M	1 L	Infratemporal swelling	N.S.	CT, MR	Open surgery with cranial fossa repair	Multiple (unspecified)	N.S.	No
SHIBUYA, 2000 <sup>*,51</sup>	1	–	–	–	–	–	–	–	–	–	–
ERCOLI, 1998 <sup>12</sup>	1	54	1 F	1 R	Pain, swelling, occlusal changes, joint sounds	N.S.	OPT, CT, MR	N.S.	Multiple (unspecified)	3	No
GIL-SALÙ, 1998 <sup>15</sup>	1	65	1 F	1 L	Sensory disturbances, pain, peripheral facial nerve palsy	8 years	CT, MR	Open surgery with skull base repair	Single giant mass	15	No
KARLIS, 1998 <sup>28</sup>	1	45	1 F	1 R	Pain, swelling, history of multiple trauma	N.S.	CT	Open surgery	Multiple (unspecified)	N.S.	N.S.
LOUIS, 1998 <sup>31</sup>	1	32	1 F	1 R	'Fullness' in the preauricular area	6 months	CT, MR	Unsuccessful arthroscopy followed by open surgery	Multiple (15–20)	24	No

\* Indicates the studies for which full text could not be achieved.

A large variability in the number of loose bodies removed from the joint space was described, ranging from two large bodies to over 200 small nodules. In most cases, the exact number of loose bodies was not provided, and only some generic descriptions about their composition (calcified nodules) and location (mainly in the upper joint compartment) were given. In two cases, a single giant mass was described.

#### Follow up and recurrence rate

Follow-up data were provided only for 58 cases, ranging from 1 month to 18 years. The recurrence rate was low, with only one case described in the literature. In two cases, corrective surgery was needed some weeks after the intervention. In one of those cases, injection of steroids was needed to achieve good pain relief, and in the other case, orthognathic surgery was performed to correct dental occlusion. The postoperative course was uneventful and easily controlled with common pain killers and antibiotics and symptom improvement was achieved in most cases (54/58).

#### Discussion

A systematic assessment of the literature on SC of the TMJ over the past decade allowed the identification of 80 new cases since the last comprehensive review by VON LINDERN *et al.*<sup>55</sup>. 154 cases have been described in 102 publications (Table 2). Most studies dealt with single case reports, and multiple case series describing from 2 to 11 patients were seldom reported (15/102 publications). Females are affected almost three times more than males, and no changes in this trend could be identified by a comparison of the most recent and past studies.

Both reviews described only studies in which surgery was performed as well as a post-surgical histopathological confirmation of diagnosis. This type of inclusion criterion might have led to the exclusion of

some other potential cases of SC and be the reason for some of the discrepancies in the reviews of prevalence of SC of the TMJ<sup>27,30,47</sup>. The summary of data from the present review, which systematically assessed all available publications from 1998 to the present, and that by VON LINDERN *et al.*<sup>55</sup>, which covered most papers published before 1998, represent the best available evidence for describing the epidemiological features of the disease.

SC usually occurs in large joints, such as the knee or shoulder. Its prevalence is very low, with less than 300 cases being reported in the literature<sup>41</sup>. It is a benign, chronic, and progressive condition that does not seem to undergo spontaneous resolution, since the presence of multiple loose bodies with the joint space interferes with function and requires surgical removal. In the case of larger joints, the classical triad of signs and symptoms is represented by restricted joint range of motion (65%), pain (57%), swelling (46.5%), which are also the main signs and symptoms identified in cases of SC of the TMJ<sup>3</sup>. Pain and swelling were present in most cases selected for this review, with a prevalence of up to about 90 and 80%, respectively, while limited range of motion was described in about 65% of patients.

Symptoms are often unspecific and hard to relate to SC, so late diagnosis is common<sup>10</sup>. Data on the duration of symptoms before diagnosis and intervention were not always reported, but the percentage of cases with long-lasting symptoms seems to be very high, since about 80% of the studies providing such data described cases with symptoms lasting for more than 2 years. Advanced imaging techniques, such as MRI and CT, may be helpful for depicting joint changes and the presence of loose bodies, suggesting the diagnosis of SC<sup>21</sup>. MRI may be useful for depicting the nodules in the early stages of formation, before ossification<sup>4</sup>, and for planning surgery in the early stages.

Surgery has always been recommended as the therapy of choice<sup>10,35</sup>, but some authors advocate less invasive techniques, such as arthroscopy and two-needle arthrocentesis, to remove the loose bodies from the joint space<sup>9</sup>. Data from the present systematic review did not support the hypothesis that minor surgery may be enough to treat SC. The reported success rate for arthroscopy was no better than 55%, since in almost half the cases complete removal of loose bodies from the joint cavity was not achieved by arthroscopy and needle aspiration alone, and open surgery was needed to clear the synovia thoroughly. Such observations are in line with current suggestions that TMJ arthroscopy has little place in the modern era of TMJ surgery, because it has no advantage over arthrocentesis in terms of efficacy and none over open surgery in terms of post-surgical course<sup>16,17</sup>.

In many cases, additional procedures, such as total synovectomy, discectomy and condylectomy were performed for loose body removal alone; the choice has to be based on the stage of the disease<sup>10</sup>. From a histopathological viewpoint, the disease has been described in terms of three distinct and progressive stages based on the phases of the metaplastic process<sup>7,36</sup>. During the first stage of the disease there is active synovial metaplastic activity without loose bodies; the second stage is characterized by the presence of metaplastic nodules plus loose bodies; in the third stage there are multiple loose bodies but no signs of active synovial disease<sup>36</sup>. In cases characterized by an advanced stage of the disease, in which degeneration and calcification of the loose bodies has taken place and there is an inactive synovial membrane, aggressive surgery with synovectomy might not be necessary due to the non-proliferative nature of the disease at this stage. Although calcifications and ossifications of the nodules might have affected the disk and/or reached extra-articular structures, requiring extensive joint clearance as well. The literature data did not provide useful information on the correlation between the stage of the disease and the type of surgical intervention. It seems that the surgeon's experience is the main determinant of the additional procedures to be performed as well as loose body removal.

Once the loose bodies have been removed, the recurrence rate of SC appears to be very low. Only one case of recurrence has been documented in the literature<sup>11</sup>. This may suggest that sponta-

Table 2. Summary of data from the present systematic review of the literature and the one by VON LINDERN *et al.* (2002)<sup>55</sup>.

	This review	Review by Von Lindern <i>et al.</i> , 2002 <sup>10</sup>	Total
Nr. of studies reviewed	42	60	102
Single case reports	34	53*	87
Total cases	80	74	154
Male:female ratio	20:56**	23:51*	43:107 (1:2.5)
Left:right joint ratio	37:40***	31:37****	68:77 (1:1.1)

\* = data were recounted on the basis of Table 1 of the original report.

\*\* = data are available only on 76 cases.

\*\*\* = 1 bilateral case.

\*\*\*\* = data were available on 68 cases.



neous inactivation of the metaplastic process occurs in most cases and that surgery may be considered a radical and definitive treatment for most SC patients, since recurrence was not reported even in cases with incomplete removal of calcifications<sup>26</sup>. A differential diagnosis of chondrosarcoma should be considered because of the life-threatening features of chondrosarcoma, which is a rare primary malignancy of the TMJ, described in less than 20 cases<sup>48</sup>. Owing to the aggressive nature of SC cases for which a known aetiology cannot be identified, it was suggested that primary cases may be neoplastic in origin and secondary cases metaplastic in origin, thus representing mild and benign tumoral variants<sup>53</sup>. Malignant transformation of SC is very rare, and only two cases have been reported in other joints<sup>18,38</sup>, but some histopathological signs of SC, such as cellular atypia, may easily be misinterpreted as signs of malignancy<sup>53</sup>.

The case report described in the present paper shares several aspects with most of the literature cases. Late diagnosis was a feature, in line with literature suggestions, and imaging techniques provided an accurate depiction of the joint and helped to plan surgery. Open surgery was selected as the most suitable procedure to remove the large loose bodies identified with CT, and the uneventful postoperative course supports the benign prognosis and successful therapeutic outcome described for all cases of SC of the TMJ.

Epidemiological data gathered by this systematic review may help researchers identify some shortcomings to be addressed in future studies. Knowledge on SC of the TMJ will be augmented by an improvement in the quality of the literature on this disease. Information on the stages of the disease was lacking in many of the reported cases included in this review as well as a detailed description of the surgical technique used. Staging the disease using the three-stage classification is information that is easy to collect and it should help to link the histopathological findings with the extension of the disease and the type of surgical procedure. Better information about the patient's history, in terms of anamnesis and symptom duration, should be provided in future studies, to identify the potential risk factors for the onset and progression of the disease. The case report described here shows how difficult it is to identify risk factors. No study has conducted a detailed investigation of risk factors.

In conclusion, SC is a rare and benign disease that rarely affects the TMJ. The

case of a patient presenting with the classical signs and symptoms of SC has been described and a systematic literature review over the past decade has been presented. 155 cases of SC of the TMJ were described, with a 2.5:1 female:male ratio and a mean age of about 46 years. A single case of bilateral localization as well as a single case of disease in a patient under 18 years were reported. A few cases of aggressive behaviour of the disease, with invasion of extra-articular structures were described, but information is lacking on the relation of this extra-articular extension with time. Owing to the non-specific symptoms, late diagnosis is common and in general over 2 years occurred between the symptom onset and surgical intervention. Surgery is the treatment of choice. Additional procedures of synovectomy, diskectomy and condylectomy have been used in some cases with no evidence-based relation with the aetiology or the stage of the disease. Minor surgery techniques, such as arthroscopy, have a low success rate owing to the difficulty of removing all the loose bodies using needle aspiration and should not be recommended as a treatment tool on the basis of the systematic assessment of the literature. The recurrence rate is very low, with only one case reported in the literature. All the information gathered through this systematic assessment of the literature should be used to relate the potential risk and prognostic factors to the least aggressive surgical intervention.

#### Funding

None.

#### Competing interests

None declared.

#### Ethical approval

Not required.

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