ACTA OTORHINOLARYNGOLOGICA ITALICA 2022 Jun 30 [Online ahead of print]; doi: 10.14639/0392-100X-N1598

Review

Silent sinus syndrome: systematic review and proposal of definition, diagnosis and management

Sindrome del seno silente: revisione sistematica e proposta di definizione, diagnosi e trattamento

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SUMMARY

Silent Sinus Syndrome (SSS) is a rare disease consisting of a collapse of maxillary sinus walls with concomitant orbital floor descent. Due to its rareness, the literature highlights some confusion on its definition, diagnosis and management. A PRISMA-compliant systematic review was performed on SSS with focus on definition, diagnosis and therapeutic management. Twenty-eight studies were selected, with 276 patients evaluated. The analysis revealed that the leading definition of SSS includes evidence of both enophtalmos and maxillary atelectasia. Although the definition of SSS accepts only spontaneous sinus collapse, the presence of sinonasal diseases and history of facial trauma are starting to be included in the criteria. Most studies (n = 21) considered CT scans satisfactory for diagnosis of SSS, while 7 also performed MR. The majority of SSS were successfully treated with isolated functional endoscopic sinus surgery (n = 17), sparing orbital reconstruction as a rescue procedure in case of non-satisfactory long-term resolution of signs. Although the literature is starting to coordinate on diagnosis of SSS, our review revealed the necessity of consensus on its definition and management.

KEY WORDS: silent sinus syndrome, maxillary atelectasia, enophthalmos, imploding antrum syndrome, silent sinus syndrome management

RIASSUNTO

La sindrome del seno silente (SSS) è una rara patologia caratterizzata da collasso delle pareti del seno mascellare con deiscenza del pavimento orbitario. A causa della rarità di questa sindrome, la letteratura mostra confusione circa la sua definizione, diagnosi e trattamento. È stata condotta una revisione sistematica fedele alle procedure PRISMA sulla SSS con focus su definizione, criteri diagnostici e trattamento (28 studi, 276 pazienti). L'analisi ha mostrato come la definizione predominante della SSS includa l'evidenza sia di enoftalmo che di atelettasia mascellare. Sebbene la definizione originaria accetti solo la presenza di collasso del seno spontaneo, ultimamente la presenza di disturbi nasosinusali e una storia pregressa di trauma facciale vengono inclusi nei criteri diagnostici. La maggior parte degli studi (28) ha considerato le immagini TC sufficienti per la diagnosi radiologica, mentre 7 hanno richiesto anche la RMN.

Il trattamento prevalente è la FESS (Functional Endoscopic Sinus Surgery) (17 studi), lasciando l'intervento di ricostruzione orbitaria come procedura di salvataggio in caso di mancata risoluzione clinica. Sebbene la letteratura inizi a coordinarsi riguardo la diagnosi di SSS, è necessario un consenso riguardo alla definizione e alla gestione terapeutica.

PAROLE CHIAVE: *sindrome del seno silente, atelettasia mascellare, sindrome del seno imploso, enoftalmo, gestione sindrome del seno silente*

Introduction

Silent Sinus Syndrome (SSS) or imploding antrum syndrome is a very rare condition, usually consisting of asymptomatic spontaneous collapse of the sinus walls and floor of the orbit [1]. Due to its rarity, the literature has often

Received: April 2, 2021
Accepted: November 14, 2021

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How to cite this article: Rosso C, Saibene AM, Felisati G, et al Silent sinus syndrome: systematic review and proposal of definition, diagnosis and management. Acta Otorhinolaryngol Ital 2022 Jun 30 [Online ahead of print]. https://doi.org/10.14639/0392-100X-N1598

highlighted the confusion around its definition, diagnosis and proper management.

The aetiology of SSS remains controversial. Patients typically deny preexisting sinus disease or orbitofacial trauma. Most authors postulate that collapse of the inferior orbital wall is induced by negative pressure generated by resorption of gas after a natural ostium occlusion that may occur during the first or second decade of life [2,3].

Clinical appearance usually consists in asymptomatic enophthalmos and altered facial appearance. Nevertheless, diplopia, sinusitis, rhinorrhoea, post-nasal drip, facial pressure, or pain may also be present [1,4].

SSS and Chronic Maxillary sinus Atelectasia (CMA) are terms which have been used interchangeably; the latter can be defined as a persistent and progressive decrease in maxillary sinus volume secondary to inward bowing of the antral walls [5].

Some authors define CMA as SSS, while others sustain that the two are separate clinical entities [6,7]. For example, according to Ende et al, it is possible to consider three stages of CMA: the last, group III, is defined by clinical facial deformity with the onset of ocular disturbances, and may be also defined as SSS [8]. Brandt et al. also argued that SSS should be considered a subtype of CMA [9].

The original definition of SSS dates to 1994 and included only spontaneous enophthalmos not associated with prior trauma or surgery [2]. Nevertheless, many authors have started accepting wider inclusion criteria involving more aetiologies (idiopathic, post traumatic, and iatrogenic) and sustain that they should be included in the definition of SSS [10].

Treatment is also a subject under continuous re-evaluation. At first, surgical treatment consisted in Caldwell-Luc sinus surgery with inferior meatotomy and concurrent transconjunctival repair of the orbital floor. In 1993, Blackwell et al. described endoscopic maxillary antrostomy (FESS: functional endoscopic sinus surgery) in conjunction with a transconjunctival orbital floor repair (OR) in three patients and reported resolution of maxillary disease on follow-up and no recurrence of enophthalmos [1].

Nowadays, the leading treatment of SSS, in order to restore the natural ventilation of the maxillary sinus, appears to be FESS [11]. Instead, the main controversy in literature remains the necessity and timing of the orbital floor reconstruction via a subconjunctival approach. Some authors prefer a one-stage approach, with endoscopic antrostomy and reconstruction of the orbital floor performed at the same time [10,12]. Others believe that a two-stage approach is more convenient, with a delay between antrostomy and the orbital reconstruction of 6 months [13]. Still others consider FESS alone to be the best choice of therapy with less invasiveness and satisfactory results, with no need of OR [6].

To shed light on these controversies, we performed a systematic review of the literature on SSS, with particular focus on definition, diagnosis and surgical approach.

Materials and methods

A PRISMA-compliant systematic literature review [14] was carried out in December 2020 on the Web of Science, PubMed and Scopus databases, using a search strategy for "(Silent Sinus) AND (Maxillary)" and "(Silent Sinus Syndrome) AND (Maxillary)".

We included studies focused on SSS with the following criteria.

Inclusion criteria

- Age 1-100 years.
- Silent sinus syndrome (SSS).
- Chronic maxillary atelectasia (CMA).

Exclusion criteria

- Studies whose main purpose was unrelated to SSS characteristics and management.
- No human patients involved.
- Language other than English, Italian, French, German and Spanish.
- Article accepted but not published.
- Article type: case reports with less than 3 patients, reviews, comments, letters to the editor, book chapters.

Population, Intervention, Comparison, Outcomes, and Study (PICOS) criteria

PICOS criteria [15] for the present review were as follows:

- Patients with CMA or SSS diagnosis.
- Intervention: evaluation of definition, diagnostic criteria and treatment.
- Comparison: comparison of different definitions, diagnosis and therapeutical options (FESS + OR, FESS alone, OR alone, antibiotic therapy, wait & see).
- Outcome: proposal of shared definition, diagnostic criteria and treatment.
- Study design: Retrospective case studies and case series (more than 3 patients) were enrolled in the review.

Data extraction and quality assessment

Two of the authors (CR and CP) independently screened the retrieved studies based on title and abstract; when uncertainty existed in the abstract evaluation, we retrieved and assessed the full text. After completion of all searches, duplicates were removed. Evaluation through full-text screening was then carried out. Critical appraisal led to the selection of 28 studies (Fig. 1). Both

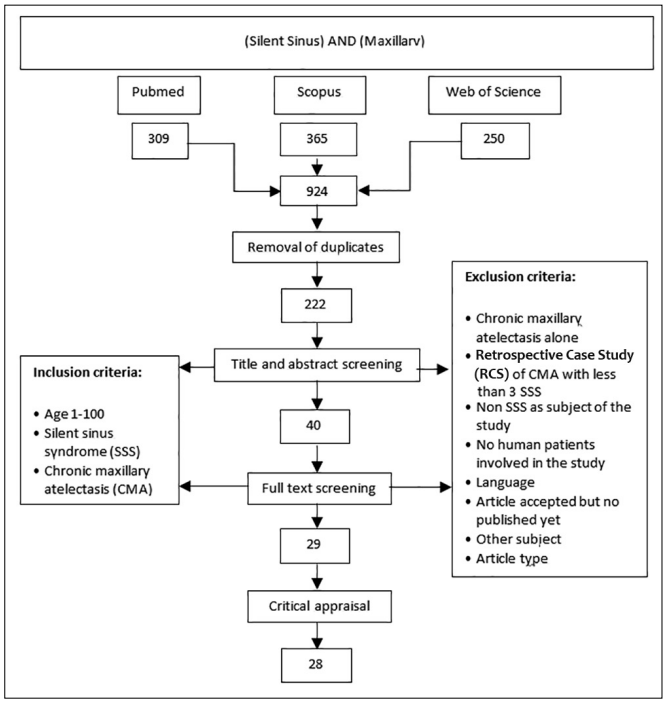

Figure 1. PRISMA flow-chart.

retrospective and prospective studies were included, while case reports and small case series were excluded because of their intrinsically lower level of evidence (the minimum number of patients was arbitrarily set at 3). Published reviews on SSS were similarly excluded, but their reference list was reviewed to identify possible additional studies. A manual search in the reference lists of these articles was performed to identify potentially relevant papers missed during the database search. Differing opinions were resolved by consensus between the two authors. Data extracted and analysed included study design, sample size, mean patient age, diagnostic criteria, associated factors, instrumental diagnosis, surgical approach and timeline, outcomes, complications and follow-up time.

Results

The systematic review retrieved 28 original articles [3,4,6,7,10-13,16-19,21,23-37]. Twenty-one were retrospective case studies (RCS), while 7 were case reports reporting at least 3 cases. A total of 276 patients were evaluated with a mean of 9.8 cases per study (range 3-57). Mean age was 40.4 years, although 3 papers did not report the age of participants (Tab. I). There was a wide variety of diagnostic criteria among studies (Tab. II). The most frequently required findings for diagnosis were evidence of enophthalmos (En) (25 studies), maxillary atelectasia (MA) (23 studies) and hypoglobus (15 studies), while post-traumatic, facial asymmetry and diplopia evidence were less required (respectively, in 3, 3 and 1 studies) as seen in Table II. Moreover, 7 studies did not directly define diagnostic criteria, which were extrapolated from the manuscript.

Nine studies included patients with sinonasal symptoms; 6 included post-traumatic cases, and 1 had post-surgical SSS (Tab. I). Twenty studies diagnosed SSS with CT alone, while 7 performed both CT and MR imaging. One study did not define what imaging was performed (Tab. I).

Surgical approaches consisted in combined FESS and OR in 72 patients, mostly in a one-step surgery. Four studies performed both types of timing-choices (one step or two steps 6 months apart), deciding according to the individual case. FESS alone was performed in 116 patients; other less common therapies were a wait & see approach (57 patients), OR alone (8 patients) and antibiotic therapy (1 patient) (Tab. III). A study by Khon et al. did not define their therapeutical approach in the 22-patient case series [29].

All patients had clinical resolution (CR) or clinical improvement (CI). Only Lin and Brown reported 2 patients who had FESS surgery alone that did not resolve enophthalmos, who then underwent subsequent OR [13,16]. Complications are reported in Table I.

Discussion

SSS is a relatively rare disease that most ENT specialists and ophthalmologists know as an entity, but the sporadic evidence leads to ambiguity on many of its aspects, from definition, to diagnostic criteria to treatment.

Definition

Our systematic review revealed that among the 28 studies evaluated, the most required diagnostic criteria were evidence of enophthalmos (25 studies) and maxillary atelectasia (MA) (23 studies), with 21 requiring both signs. Hypoglobus observation follows as essential criteria in 15 studies, also with evidence of orbital floor remodeling at CT scans (11 studies) and the absence of sinonasal diseases (10 studies) (Tab. II).

Therefore, even if there is much misunderstanding around the definition of SSS, it is possible to assume that most authors agree in having both enophthalmos and MA as necessary criteria. Only 2 studies required only MA in the absence of enophthalmos, considering CMA and SSS as a same entity [4,17].

Post-traumatic silent sinuses were originally excluded from the definition of SSS [18,19], and some recent papers continue

Table I. Baseline data of the included studies.

Study (year)	Study design	Sample size	Mean age (range) in years	Diagnostic criteria	Associated factors	Diagnosis	Surgical approach	Surgical timeline	Outcome	Complications	Follow-up (months)
Behbehani [25] (2006)	RCS	5	36.7 (32-42)	En Hypoglobus MA	Ost-traumatic (1) Chronic sinusitis (4)	CT scans	FESS and OR (5)	One step (5)	100% CI or CR	Residual 1mm En (2) Transient infraorbital hypoaesthesia (1)	24
Bossolesi [18] (2008)	Case reports	4	42 (38-45)	En Opacified maxillary sinus Absence of major sinus pathology Absence of previous trauma, surgery or congenital facial deformity	Chronic sinusitis (2)	End CT MR	FESS and OR (4)	One step (4)	100% CR	None	12-24
Brown [16] (2017)	RCS and review	6	43 (35-52)	En MA Hypoglobus	Chronic sinusitis (1) Post-traumatic (6)	CT	FESS (1) OR (1) FESS and OR (4)	One step (1) Two steps (3)	100% CI	Residual 2 mm En (1) minor diplopia (1)	Not defined
Chariba [11] (2014)	RCS	13	34 (13-61)	En MA	Chronic sinusitis (5)	CT	FESS (13)	N/A	100% CI	Orbital breach (2)	30
Chavez-Montoya [26] (2017)	RCS	3	44 (37-48)	MA Opacified maxillary sinus	Nasal polyposis (1)	CT	FESS (1) FESS and OR (1) Wait & see (1)	One step (1)	100% CI	-	24
Claròs [12] (2015)	Case reports	3	36.3 (25-45)	Not defined En Hypoglobus Low pressure in maxillary sinus	-	CT	FESS + OR (3)	One step (3)	100% CR	None	Not defined
Claròs [10] (2019)	RCS	13	38 (25-53)	Not defined En Hypoglobus Orbital floor resorption MA Absence of trauma and congenital facial deformity	-	CT	FESS + OR (13)	One step (13)	100% CI	None	Not defined
Cobb [21] (2012)	Case reports	3	44 (30-60)	En MA Maxillary opacity	Post-traumatic (2) Post-surgical (1)	CT MR	FESS and OR (3)	One step (2) Two steps (1)	100% CI	None	12 (1) 36 (1) 48 (1)
De Dorlodot [7] (2017)	RCS	4	44 (12-60)	Facial asymmetry En and/or hypoglobus MA Absence of sinonasal symptoms	-	CT	FESS (3) FESS and OR (1)	Two steps (1)	100% CI	Slight enophtalmos in some patients (n° not defined)	Not defined
Eyigor [6] (2016)	RCS	16	42.37 (20-66)	MA En Absence of trauma or sinusitis	-	CT MR	FESS (16)	N/A	Ongoing follow-up	Ongoing follow-up	Ongoing
Farneti [3] (2017)	RCS and review	6	10 (7-14)	Absence of sinusitis Remodeling of orbital floor at CT/MR scans Opacified maxillary sinus Absence of trauma or congenital deformities	-	CT	FESS (6)	N/A	100% CR	Residual headache (1)	18-135
Freiser [13] (2020)	RCS	57	12.5 (3.7-18)	Not defined En Hypoglobus MA	-	CT	FESS (19) Wait & see (38)	N/A	100% CI or CR	Not defined	Not defined
Gaudino [23] (2013)	RCS	6	44 (22-67)	En Diplopia Opacified sinus MA	-	CT MR	FESS (1) FESS and OR (2) Wait & see (3)	One step (1) Two steps (1)	2 FESS: no significant CI 2 FESS and OR: CI	Not defined	Not defined

continues ▶

Table I. Baseline data of the included studies (*follows*).

Study (year)	Study design	Sample size	Mean age (range) in years	Diagnostic criteria	Associated factors	Diagnosis	Surgical approach	Surgical timeline	Outcome	Complications	Follow-up (months)
Illner [27] (2002)	RCS	5	47 (39-65)	Not defined En Hypoglobus Maxillary sinus completely developed Maxillary sinus opacified Infundibulum occluded	-	CT/MR	FESS (3) Antibiotic therapy (1) Wait & see (1)	N/A	Not defined	Not defined	Not defined
Kashima [28] (2016)	RCS	11	39.5 (23-62)	Not defined MA En Hypolgobus	Not defined	Not defined	FESS and OR (11)	One step (11)	100% CI	Residual enophthalmos (1) 1 mm over correction (1) Lower eye-lid retraction (1) Nasolacrimal duct obstruction (1)	9
Kohn [29] (2013)	RCS	22	41.2 (22-70)	En Hypoglobus MA Orbital changes at CT scans	Chronic rhinosinusitis (9) Post-traumatic (10)	CT/MR	Not defined	Not defined	Not defined	Not defined	Not defined
Korn [17] (2009)	Case reports	5	Not defined	MA Orbital floor resorption	Chronic rhinosinusitis (2)	CT	FESS and OR (5)	One step (5)	100% CI	transient infraorbital hypoaesthesia (1) residual diplopia (1)	Not defined
Lee [30] (2018)	Case reports	3	44.6 (37-55)	En Hypoglobus MA Opacified maxillary sinus	Post-traumatic (1)	CT MR	FESS (1) OR (1) Wait & see (1)	N/A	100% CI	residual diplopia (1) Residual enophthalmos (1)	Not defined
Lin [31] (2015)	RCS	9	Not defined	MA Orbital floor resorption En Absence of sinusitis	Post-traumatic (1)	CT	FESS (7) FESS and OR (2)	Two steps (2)	2 FESS alone did not resolved enophthalmos and so underwent OR	none	21.4
Martìnez-Capoccioni [32] (2016)	RCS	20	44.2 (28-67)	En and/or Hypoglobus Endoscopic findings of MA Altered facial appearance Maxillary contraction and orbital enlargement at CT scans	Nasal obstruction (9)	CT	FESS (15) Wait & see (5)	N/A	100% CI	none	6-18
Rose [33] (2003)	RCS	14	41.3 (25-78)	Not defined En Hypoglobus Remodeling of orbital floor Absence of nasal diseases	-	CT	OR (6) Wait & see (8)	N/A	100% CI	none	5-33
Sesenna [34] (2010)	Case reports	3	39 (28-46)	En Hypoglobus MA Absence of sinusitis, trauma and congenital deformities Orbital floor remodeling at CT scans	-	CT	FESS and OR (3)	One step (2)	100% CR	None	10-16
Sivasubramaniam [35] (2011)	RCS	18	Not defined (19-54)	En and/or Hypoglobus MA Altered facial appearance Absence of sinusitis	-	CT	FESS (18)	N/A	78% CR 17% CI	Residual enophthalmos (1)	15-120
Thomas [22] (2003)	RCS	4	32 (27-35)	En Absence of sinusitis MA Opacified maxillary sinus Inferior bowing of orbital floor	-	CT	FESS and OR (2) FESS (2)	Two steps (2)	100% CR	Residual enophthalmos after FESS alone (2) which required second step surgery with OR	Not defined

continues ▶

Table I. Baseline data of the included studies (*follows*).

Study (year)	Study design	Sample size	Mean age (range) in years	Diagnostic criteria	Associated factors	Diagnosis	Surgical approach	Surgical timeline	Outcome	Complications	Follow-up (months)
Vander Meer [4] (2001)	RCS	4	42 (38-47)	En MA Absence of sinusitis, trauma or congenital deformities Remodeling of orbital floor at CT scans	-	CT	FESS and OR (4)	One step (4)	100% CR	none	2-36
Virgin [36] (2008)	RCS	5	42 (22-65)	En MA	-	CT	FESS (4) FESS and OR (1)	Two steps (1)	100% CR	None	24
Wan [37] (2000)	Case reports	3	41 (38-44)	Not defined En Hypoglobus MA Remodeling orbital floor	Chronic sinusitis (1)	CT	FESS (3)	N/A	100% CR	none	N/A
Wise [19] (2007)	RCS Loro si impuntano solo sui reperti diagnostici alla TC	11	46.5 (11-70)	En Opacified maxillary sinus Orbital floor remodelling Absence of chronic sinusitis, trauma or congenital deformities	-	CT	FESS (3) FESS and OR (8)	Not defined	Not defined	none	Not defined

RCS: retrospective case study; En: enophthalmos; MA: maxillary atelectasia; FESS: functional endoscopic sinus surgery; OR: orbital reconstruction; CI: clinical improvement; CR: clinical resolution; End: nasal endoscopy

considering post-traumatic cases not definable as SSS due to their lack of spontaneous development [10,20].

Nevertheless, a retrospective analysis of 6 cases and literature review showed how traumatic SSS management follows the same principles as for spontaneous SSS [21]. In fact, our analysis revealed that recent papers started to include enophthalmos with MA secondary to traumatic events in the group of SSS, explaining that clinical and radiological presentations are comparable, as well as surgical treatment [13,21,22].

Moreover, the association of SSS with sinonasal symptoms is debated. Ten studies considered the evidence of sinonasal symptoms as an exclusion criterion because MA secondary to chronic sinusitis with sinus dysventilation and sinonasal symptoms is, by those authors, considered as CMA and not as SSS [9]. At the same time, the majority of studies do not clarify this point, and include among SSS subjects who have associated factors such as sinusitis, chronic nasal congestion and other sinonasal symptoms of obstruction, even though these are not considered diagnostic criteria (Tab. II).

Finally, 7 studies did not specify diagnostic criteria, which were extrapolated from the clinical description of cases. This risks producing inaccurate studies given that the literature lacks in consensus in SSS diagnosis and the reader may find its definition confusing.

From our analysis, it appears that the leading definition of SSS should include the evidence of enophthalmos and maxillary atelectasia. We believe that presence of sinonasal diseases and history of facial trauma may be reasonable factors to be included in anamnestic data because of the comparable clinical presentation and treatment.

Definition proposal

Our proposal for definition is:

- criteria to diagnose SSS are contemporary presence of enophtalmos and maxillary atelectasia;
- minor associated factors may be the presence of sinonasal diseases and history of facial trauma.

Diagnosis

Most studies only required CT for imaging investigation, since the relevance was to investigate maxillary bony walls atelectasia with possible resorption of the orbital floor. Nevertheless, 7 groups also considered it useful to add MR scans (Tab. I) to evaluate:

- dislocation of extra and intra-conical fat, extrinsic ocular muscles and optic nerve;
- differentiation between mixed signal central secretions and high signal peripheral;
- thick edematous mucosal lining within the maxillary sinus [6,18,23].

Diagnostic proposal

Only CT scan is required. MR imaging can be associated in selected cases, and specifically to evaluate a marked hypoglobus. We propose to add evaluation of extraocular muscle movement and potential diplopia at first ENT clinical evaluation, in order to select symptomatic and therefore the most critical cases [10,20].

Treatment

The review also showed disagreement on management strategies. Seventeen authors proposed combined FESS and OR surgery as the leading therapy to obtain resolution of anatomical impairment and diplopia, if present. Among these, the majority (n = 9) performed both the procedures simultaneously, while 4 preferred to first carry out FESS surgery and to observe a possible progressive improvement of the enophtalmos to decide whether or not to perform additional OR. Eventually, 3 studies performed FESS and either simultaneous or delayed OR according to the criticality of the case (Tab. III).

The reasons which guide each surgical team towards one choice or another are multiple. The main factor is undoubtedly the severity of enophthalmos and hypoglobus, although no study defined a quantitative cut-off which could help in the surgical decision. OR approach is supported by the fact that, although there have been reports of resolution of the progression of enophthalmos by antrostomy alone, it is unclear whether other aesthetic deformities caused by SSS as hypoglobus or superior sulcus deformity also respond. Behbehani et al. believe that delaying orbital implant placement in cases with significant enophthalmos and hypoglobus is unjustified since complications like diplopia or infection are rare with this procedure. Furthermore, simultaneous implant placement also obviates the need for additional anaesthesia and hospitalisation [25]. Cobb et al. maintain that FESS alone may stop the descent of the orbital wall, but there would be no reason to expect that the position of the orbital floor, and thus the globe, would be reversed [21].

On the contrary, Thomas et al. support delayed repair of the orbital floor as in some patients enophthalmos improves with antrostomy alone [22]. Moreover, OR has not been shown to provide any significant restoration in the orbital muscle functions, and because of that diplopia is not corrected [6].

A total of 116 patients underwent FESS surgery alone, being considered as a necessary and sufficient procedure to resolve MA and enophthalmos (Tab. III). If we consider the entire case series of the review, FESS alone appears as the leading therapeutic choice, with only 7 reporting residual enophthalmos (6%), and 4 requiring subsequent OR (3.4%). Moreover, Numa et al. undertook a review of 84 cases and concluded that for patients with SSS diagnosis, uncinectomy alone may be sufficient [24].

FESS + OR follows with 72 cases treated with this management. Wait & see (57 patients) may be a valid alternative in asymptomatic cases or young population [13]. OR alone (8 patients) and antibiotic therapy (1 patient) remain marginal therapeutic options (Tab. III).

This leads us to the conclusion that most of SSS may be successfully treated with isolated FESS surgery, sparing OR as a rescue procedure in case of non-satisfactory resolution of enophthalmos or diplopia.

Treatment proposal

FESS is the leading treatment for SSS and orbital reconstruction should be performed only in selected and symptomatic cases or, if needed, in a second approach when no resolution is seen. The proposed timepoint for a potential postponed OR, according to the literature, is 6 months.

Our review clearly shows the need to develop consensus regarding the definition of SSS and most of all its management. Even if the literature has started to find marginal consensus in recent years, it appears necessary to define shared diagnostic criteria, as well as a shared approach to the best treatment choices with the lowest rate of invasiveness and morbidity. The literature would also

Table II. Summary of diagnostic criteria for SSS.

Not directly defined	Study (year)	E	Hypoglobus	MA	Opacified maxillary Sinus	NO previous trauma, surgery or congenital deformities	No sinonasal symptoms	Post-traumatic	Facial asymmetry	Diplopia	Orbital floor remodeling at CT scan
	Behbehani [25] (2006)	X	X	X							
	Bossolesi [18] (2008)	X			X	X	X				
	Brown [16] (2017)	X	X	X				X			
	Chariba [11] (2014)	X		X							
	Chavez-Montoya [26] (2017)			X	X						
x	Claròs [12] (2015)	X	X	X							
x	Claròs [10] (2019)	X	X	X		X					
	Cobb [21] (2012)	X		X				X			
	De Dorlodot [7] (2017)	X	X	X			X		X		
	Eyigor [6] (2016)	X		X		X					
	Farneti [3] (2017)				X	X	X				X
x	Freiser [13] (2020)	X	X	X							
	Gaudino [23] (2013)	X		X	X					X	
x	Illner [27] (2002)	X	X		X						
x	Kashima [28] (2016)	X	X	X							
	Kohn [29] (2013)	X	X	X							X
	Korn [17] (2009)			X							X
	Lee [30] (2018)	X	X	X	X						
	Lin [31] (2015)	X		X			X	X			X
	Martìnez-Capoccioni [32] (2016)	X	X	X					X		X
x	Rose [33] (2003)	X	X				X				X
	Sesenna [34] (2010)	X	X	X		X	X				X
	Sivasubramaniam [35] (2011)	X	X	X			X		X		
	Thomas [22] (2003)	X		X	X		X				X
	Vander Meer [4] (2001)	X		X		X	X				X
	Virgin [36] (2008)										
	Wan [37] (2000)	X		X							
x	Wise [19] (2007)	X	X	X							X
7	TOTAL	24	15	23	7	6	9	3	3	1	10

E: enophtalmos

benefit from prospective studies on the best outcome in SSS management.

Conclusions

Literature about SSS is controversial and confusing. Our systematic review illustrates that the leading definition of SSS includes the evidence of enophthalmos and maxillary atelectasia by CT. Hypoglobus, presence of sinonasal diseases and history of facial trauma may or may not be associated, although the clinical relevance and management seems to be comparable. FESS alone appears to be the first choice of treatment, since at post-operative follow-up, orbital floor retraction tends to spontaneously reverse with clinically satisfactory results.

Table III. Summary of therapeutic strategies for SSS.

Study (year)	FESS + OR	FESS	OR	Wait & see	Antibiotic therapy
Behbehani [23] (2006)	X (5)				
Bossolesi [16] (2008)	X (4)				
Brown [14] (2017)	X (4)	X (1)	X (1)		
Chariba [11] (2014)		X (13)			
Chavez-Montoya [24] (2016)	X (1)	X (1)		X (1)	
Claròs [12] (2015)	X (3)				
Claròs [10] (2019)	X (13)				
Cobb [19] (2012)	X (3)				
De Dorlodot [7] (2017)	X (1)	X (3)			
Eyigor [6] (2016)		X (16)			
Farneti [3] (2017)		X (6)			
Freiser [13] (2020)		X (19)		X (38)	
Gaudino [21] (2012)	X (2)	X (1)		X (3)	
Illner [25] (2002)		X (3)		X (1)	X (1)
Kashima [26] (2016)	X (11)				
Kohn [27] (2013)	-	-	-	-	-
Korn [17] (2009)	X (5)				
Lee [28] (2019)		X (1)	X (1)	X (1)	
Lin [29] (2015)	X (2)	X (7)			
Martìnez-Capoccioni [30] (2016)		X (15)		X (5)	
Rose [31] (2003)			X (6)	X (8)	
Sesenna [32] (2010)	X (3)				
Sivasubramaniam [33] (2011)		X (18)			
Thomas [20] (2002)	X (2)	X (2)			
Vander Meer [4] (2001)	X (4)				
Virgin [34] (2008)	X (1)	X (4)			
Wan [35] (2000)		X (3)			
Wise [17] (2007)	X (8)	X (3)			
TOTAL	17	17	3	7	1
TOTAL PER CASES	72	116	8	57	1

Acknowledgements

We gratefully thank the Librarians from the Università degli Studi di Milano, Biblioteca del Polo Centrale. Without their help, we would not have been able to locate and analyse a significant number of the studies included in this systematic review.

Conflict of interest statement

The authors declare no conflict of interest.

Funding

This research did not receive any specific grant from funding agencies in the public, commercial, or not-for-profit sectors.

Authors' contributions

CR made a substatial contribution to the conception and design of the article, to the acquisition, analysis and interpretation of data. AMS and GF critically revised the article and gave the final approval of the version to be published. GP made a substatial contribution to the conception and design of the article and gave the final approval of the version to be published.

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