

PROSTHODONTIC TREATMENT MODALITIES FOR
MICROSTOMIA PATIENTS: A SYSTEMATIC REVIEW

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FACULTY OF DENTISTRY
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KUALA LUMPUR

2024

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**RESEARCH REPORT SUBMITTED IN PARTIAL
FULFILLMENT OF THE REQUIREMENTS FOR THE
DEGREE OF MASTERS OF ORAL SCIENCE**

**FACULTY OF DENTISTRY
UNIVERSITI MALAYA
KUALA LUMPUR**

2024

UNIVERSITY OF MALAYA
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Title of Project Paper/Research Report/Dissertation/Thesis (“this Work”):

Prosthodontic Treatment Modalities For Microstomia Patients: A Systematic Review

Field of Study: Prosthodontic

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ABSTRACT

Microstomia, a condition characterized by a reduced oral aperture size, posed challenges in dental procedures and prosthetic rehabilitation. This systematic review critically evaluated published case reports and case series related to the prosthodontic treatment of microstomia. The review identified the most common methods for measuring the severity of microstomia and suggested appropriate treatment options tailored to the severity of the condition.

Using the PICO framework, the study reviewed English publications up to May 2024, emphasising only on mouth opening assessments and treatments while excluding temporomandibular joint issues.

The search methodology encompassed multiple electronic databases, including PubMed, Medline, EBSCO host, Scopus, and Web of Science, using keywords and MESH terms: microstomia, limited oral opening, limited oral access, prosthodontic, and prosthesis. Data extraction was followed by a standardised process, verified by multiple reviewers for accuracy.

The findings of this review revealed that the most adopted approach to measure maximal mouth opening (MMO) in microstomia patients was the lip-to-lip technique for edentulous and combination cases, while the interincisal technique was predominantly used for partially dentate patients. After removing all duplicates and screening, the total articles were 82 case reports, of which 15 were excluded due to insufficient details such as a lack of follow-up regarding patient outcomes. The correlation analysis was conducted using the remaining 67 reports. The Fisher Exact Test indicated that there was no significant correlation between the severity of microstomia and the choice of prosthodontic treatment approaches. None of the authors used conventional dentures for

cases with a severely reduced maximum mouth opening (less than 20mm for edentulous and less than 25mm for partially dentate or combination).

In conclusion, the lip-to-lip technique is the most common way to measure maximal mouth opening (MMO) for edentulous and combination cases, while the interincisal technique was preferred for partially dentate patients. There was no significant link between the severity of microstomia and the treatment chosen. Therefore, more research is needed to better understand this relationship.

Keyword : Microstomia, limited oral opening, limited oral access, prosthodontic, prosthesis.

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ABSTRAK

Mikrostomia, satu keadaan saiz bukaan mulut yang kecil, memberikan cabaran dalam prosedur pergigian dan pemulihan prostetik. Kajian sistematik ini menilai secara kritikal laporan kes dan siri yang diterbitkan berkaitan dengan pengurusan prostodontik mikrostomia. Ulasan ini mengenal pasti kaedah paling banyak digunakan untuk mengukur pembukaan mulut dan mencadangkan pendekatan untuk pilihan rawatan yang disesuaikan dengan tahap kondisi tersebut.

Dengan menggunakan rangka kerja PICO, kajian ini telah menyemak penerbitan dalam Bahasa Inggeris sehingga Mei 2024, memfokuskan laporan kes mengenai kaedah pengukuran bukaan mulut dan rawatan sambil mengecualikan isu sendi temporomandibular.

Metodologi carian merangkumi pelbagai pangkalan data elektronik, termasuk PubMed, Medline, EBSCO host, Scopus, dan Web of Science, menggunakan strategi kata kunci yang menumpukan mikrostomia, pembukaan mulut terhad, akses mulut terhad, prostodontik dan prostesis. Pengekstrakan data mengikuti proses piawai, disahkan oleh beberapa penilai untuk ketepatan.

Pendekatan yang paling banyak digunakan untuk mengukur pembukaan mulut maksimum (MMO) pada pesakit mikrostomia adalah teknik bibir ke bibir untuk kes tanpa gigi dan kes kombinasi, manakala teknik interincisal digunakan untuk pesakit yang mempunyai gigi sebahagian. Selepas mengeluarkan duplikasi dan saringan, sejumlah 82 laporan kes dikenal pasti, di mana 15 telah dikecualikan kerana kekurangan maklumat susulan mengenai hasil rawatan pesakit. Analisis korelasi dijalankan menggunakan 67 laporan yang tinggal. Ujian Fisher Exact menunjukkan bahawa tidak ada korelasi yang signifikan antara tahap mikrostomia dan pilihan pendekatan rawatan prostodontik. Tiada penulis yang membuat gigi palsu konvensional untuk kes dengan pembukaan mulut

maksimum yang sangat terhad (kurang daripada 20mm untuk pesakit tanpa gigi dan kurang daripada 25mm untuk pesakit dengan gigi sebahagian atau gabungan).

Kesimpulannya, teknik bibir ke bibir adalah kaedah yang paling biasa digunakan untuk mengukur pembukaan mulut maksimum (MMO) bagi pesakit tanpa gigi dan kes kombinasi, manakala teknik interinsisal lebih disukai untuk pesakit yang mempunyai gigi sebahagian. Tiada kaitan yang signifikan antara tahap keparahan mikroestomia dengan rawatan yang dipilih. Oleh itu, lebih banyak kajian diperlukan untuk memahami hubungan ini dengan lebih mendalam.

Kata kunci : Mikroestomia, pembukaan mulut terhad, akses mulut terhad, prostodontik dan prostesis.

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ACKNOWLEDGEMENTS

In the name of Allah, the Most Gracious and the Most Merciful, Alhamdulillah, all praises to Allah for His blessing in the completion of this research. May Allah's blessing go to our Prophet Muhammad S.A.W, his family and his companions.

First and foremost, I would like to express my deepest appreciation to my supervisors, Dr. Enas Abdalla M.Etajuri, Dr. Siti Fauzza binti Ahmad, Associate Professor Dr. Ros Anita binti Omar, and Associate Professor Dr. Lim Ghee Seong for their generous support and guidance throughout the stages of writing this report. Their invaluable feedback and suggestions have contributed to the success of this project.

I would like to extend my heartfelt appreciation to my incredible colleagues and friends, whose unwavering support, encouragement, and invaluable advice have been instrumental in this journey.

Further, I must express my deepest gratitude to my cherished family, particularly my husband, Ahmad Tawfik bin Adnan, my wonderful children, and my loving mother. Their unwavering belief in me has truly made all the difference in my journey. Their boundless support and profound understanding have served as my anchor, continually lifting my spirits and fuelling my motivation through every challenge I've faced. I am incredibly blessed to have such a solid foundation in my life, nurturing and empowering me every step of the way.

TABLE OF CONTENTS

Abstract	iii
Abstrak	v
Acknowledgements	vii
Table of Contents	viii
List of Figures	xii
List of Tables.....	xiii
List of Symbols and Abbreviations.....	xiv
List of Appendices	xv
CHAPTER 1: INTRODUCTION.....	1
1.1 Background of the study	1
1.2 Problem statement	3
1.3 Aims.....	4
1.4 Objectives	5
1.5 Research questions.....	5
CHAPTER 2: LITERATURE REVIEW.....	6
2.1 Aetiology of microstomia	6
2.2 Epidemiological aspect.....	7
2.3 Normal mouth opening	8
2.4 Clinical characteristics of microstomia	9
2.5 Treatment for microstomia	13
2.5.1 Surgical therapy.....	14
2.5.2 Non-surgical therapy	15
2.6 Prosthodontic treatment.....	17

2.6.1	Impression technique.....	17
2.6.2	Digital impressions.....	19
2.6.3	Maxillomandibular relationship (MMR).....	20
2.6.4	Prosthesis.....	21
CHAPTER 3: METHODOLOGY.....		25
3.1	Protocol and registrations.....	25
3.2	Ethical considerations and funding requisition.....	25
3.3	Study design.....	25
3.4	Eligibility criteria.....	25
3.5	Search methods for identification of studies`.....	26
3.5.1	Information sources.....	26
3.5.2	Search terms.....	26
3.5.3	Search strategy.....	27
3.6	Data extraction and management.....	29
3.6.1	Sources of evidence selection/ screening.....	29
3.6.4	Quality assessment of case reports and case series.....	31
CHAPTER 4: RESULT.....		32
4.1	Critically reviewed case reports and cases series.....	32
4.1.1	Outcome of search strategy and selection of sources of evidence/ reference.....	32
4.1.2	Data extraction sheet.....	34
4.2	The most adopted approach to measure severity of microstomia.....	44
4.3	Correlation between severity of microstomia with prosthodontics treatment.....	45
4.3.1	Classification of microstomia severity according to dentition status.....	45
4.3.2	The correlation.....	46

4.3.3	Correlation between severity of microstomia with prosthodontics treatment in edentulous patients.	48
4.3.4	Correlation between severity of microstomia with prosthodontics treatment in combination cases.	49
4.3.5	Correlation between severity of microstomia with prosthodontics treatment in partially dentate patients.....	50
4.4	Aetiology of microstomia, dentition status and impression techniques	51
4.4.1	Aetiology of Microstomia	51
4.4.2	Dentition status.....	52
4.4.3	Impression technique.....	52
CHAPTER 5: DISCUSSION		56
5.1	Critically reviewed case reports and cases series	56
5.1.1	Overview	56
5.1.2	Method for the critical review of case reports and case series	57
5.1.2.1	Eligibility criteria	57
5.2	The most adopted approach to measure severity of microstomia.....	58
5.3	Correlation between severity of microstomia with prosthodontics treatment.....	59
5.3.1	Recommended prosthodontic treatments based on the severity of microstomia	61
5.4	Implementing Bias Reduction Strategies.....	62
5.5	Limitations.....	62
5.6	Recommendations for future research	63
CHAPTER 6: CONCLUSION.....		64
References		65
APPENDIX A : JBI Critical Appraisal Form (Case Reports)		78

APPENDIX B : JBI Critical Appraisal Form (Case Series)	79
APPENDIX C : The most adopted approach to measure severity of microstomia (edentulous – 48 cases)	80
APPENDIX D : The most adopted approach to measure severity of microstomia (combination - 14 cases)	81
APPENDIX E : The most adopted approach to measure severity of microstomia (partially dentate - 20 cases)	82
APPENDIX F : AETiology of microstomia in 82 case reports and cases series	83
APPENDIX G : Dentition status in 82 case reports and case series	84
APPENDIX H : Preliminary impression technique in 82 case reports and case series ..	85
APPENDIX I : Final impression technique in 82 case reports and case series	86

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LIST OF FIGURES

Figure 4.1 Summary of the Search Strategy	33
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LIST OF TABLES

Table 2.1 The Index of Oral Access by King (2016).....	10
Table 2.2 Classification systems based on diagnosis and management for microstomia patients proposed by Maria et al. (2022).....	12
Table 3.1 List of information sources used in the study	26
Table 3.2 Keyword search.....	27
Table 3.3 Final comprehensive search strategy across selected electronic databases ...	28
Table 4.1 Table extraction sheet	35
Table 4.2 Demographic variables	44
Table 4.3 The severity of microstomia in relation to dental condition compared to the prosthodontic treatment provided	47
Table 4.4 Aetiology of Microstomia and Dentition Status	51
Table 4.5 Impression techniques.....	53
Table 5.1 Proposed prosthodontic treatments according to severity of microstomia	61

LIST OF SYMBOLS AND ABBREVIATIONS

MMO	:	Maximum mouth opening
TMJ	:	Temporomandibular joint
CAD/CAM	:	Computer-aided design & computer-aided manufacturing
SSc	:	Systematic sclerosis
SMF	:	Oral submucous fibrosis
EB	:	Epidermolysis bullosa
MB	:	Moebius syndrome
IOA	:	Index of oral access
DM	:	Diagnosis and management
MMR	:	Maxillomandibular relationship
POM	:	Point of measurement

LIST OF APPENDICES

APPENDIX A : JBI Critical Appraisal Form (Case Reports)	78
APPENDIX B : JBI Critical Appraisal Form (Case Series)	79
APPENDIX C : The most adopted approach to measure severity of microstomia (edentulous – 48 cases)	80
APPENDIX D : The most adopted approach to measure severity of microstomia (combination - 14 cases)	81
APPENDIX E : The most adopted approach to measure severity of microstomia (partially dentate - 20 cases)	82
APPENDIX F : Aetiology of microstomia in 82 case reports and cases series	83
APPENDIX G : Dentition Status in 82 case reports and case series	84
APPENDIX H : Preliminary impression technique in 82 case reports and case series	85
APPENDIX I : Final impression technique in 82 case reports and case series	86

CHAPTER 1: INTRODUCTION

1.1 Background of the study

Microstomia, derived from the Greek words "micros" meaning small and "stoma" meaning mouth, is defined as an abnormally small oral opening ("The Glossary of Prosthodontic Terms: Ninth Edition," 2017). It is also known as reduced oral aperture (ROA), which is a condition that is frequently overlooked despite its detrimental consequences on the masticatory function, patient appearance, speech and overall quality of life (Maria et al., 2022; Srivastava et al., 2022).

Microstomia and limited mouth opening are related but distinct concepts, both terms are often leading to confusion in diagnosis. Limited mouth opening is a broader term that encompasses any restriction in mouth opening, regardless of the size of the oral aperture. This condition can arise from various causes, including temporomandibular joint (TMJ) disorders, muscle spasms, trauma, dental issues, or psychological factors (Edger-Lacoursière et al., 2024; Park et al., 2022). In contrast, microstomia specifically refers to a condition characterised by a reduced oral aperture size, which poses significant challenges for dental prosthetic rehabilitation (Kunusoth et al., 2022). While microstomia falls under the umbrella of limited mouth opening, it is crucial to recognise that not all cases of restricted mouth opening are classified as microstomia (Edger-Lacoursière et al., 2024).

Microstomia and limited mouth opening due to TMJ disorders exhibit distinct clinical characteristics and necessitate different diagnostic approaches for proper management. Microstomia is typically marked by a severely reduced mouth opening, and usually results from congenital or acquired conditions. This condition leads to significant functional challenges, including difficulties with speech, eating, and aesthetic concerns (Ki & Park, 2024; Park et al., 2022). On the other hand, limited mouth opening due to TMJ disorders

is associated with symptoms such as pain, clicking, and restricted jaw movement, with the maximum mouth opening often exceeding 20 mm (Ansar et al., 2022; Balel et al., 2023). The diagnosis of TMJ-related cases follows the Diagnostic Criteria for Temporomandibular Disorders (DC/TMD), which includes a detailed clinical examination and imaging techniques like MRI to assess disc position and joint health (Ansar et al., 2022; Balel et al., 2023). Although both conditions involve restricted mouth opening, microstomia is defined by a significantly smaller oral aperture and is assessed subjectively, while TMJ disorders are diagnosed through established clinical criteria and imaging. Careful distinction is required due to their overlapping symptoms.

Microstomia can significantly diminish a patient's quality of life by impacting multiple aspects of their daily functioning, including physical comfort, social interactions, and emotional well-being (Ki et al., 2020). The condition often leads to poor nutrition, as restricted oral opening hinders food intake and chewing ability, making it difficult to eat and swallow. Consequently, patients may avoid certain foods or meals altogether, which will affect their overall health. Furthermore, speech production is commonly affected, resulting in articulation and pronunciation difficulties that challenge communication, which can hinder both social and professional interactions (Antonarakis et al., 2017).

Beyond the functional impairments, microstomia can negatively affect a patient's confidence and self-esteem. The inability to eat, speak, or smile without difficulty may draw unwanted attention, leading to embarrassment and discomfort (Zweifel et al., 2010). Additionally, maintaining proper oral hygiene becomes difficult with microstomia, heightening the risk of dental problems such as caries and periodontitis, which further diminishes daily functioning (Sahoo et al., 2013). These physical, social, and emotional challenges can contribute to social withdrawal, loss of independence, and even depressive feelings, exacerbating the overall impact on the patient's quality of life (Selvi et al., 2011).

Additionally, constructing dental prostheses for microstomia patients can indeed present unique challenges due to the limited mouth opening. One of the primary hurdles is making an accurate impression, as the limited mouth opening makes it difficult to insert standard impression stock trays. This necessitates the use of modified or sectional impression trays to capture the necessary anatomical details accurately (Subba et al., 2022). Again, the fabrication of a well-fitting prosthesis is critical for restoring aesthetics, comfort, and function, which requires precise recording of anatomical landmarks and the construction of accurate custom trays and diagnostic casts (Maria et al., 2022). It is important to note that reduced tissue support and compromised oral anatomy may also affect the stability and retention of prosthetic devices.

Numerous case reports, including those by Cura et al. (2003), Singh et al. (2014) and Tripathi et al. (2011) have detailed various techniques and methods for denture construction. In a recent systematic review, Patil et al. (2019) conducted an in-depth evaluation of all the prosthetic methods applied in the oral rehabilitation of patients with microstomia. They found that most used prostheses are segmented, collapsed, flexible, and folding dentures, while implant-supported prostheses are the next preferred option. Other types of prostheses were a hinged prosthesis with a swing lock mechanism, resin-bonded bridgework, a complete denture with limited interocclusal space, and a prosthesis with attachment system. According to them, long-term follow-up is necessary for each case to evaluate the success rate of each approach. They concluded that the success of the prosthetic approach mostly relies on the severity of the microstomia and the underlying aetiology of the microstomia.

1.2 Problem statement

Various case reports have explored different techniques for constructing prostheses for microstomia patients. Traditional methods often require modifications, such as the use of

sectional trays. These trays can be easily assembled and disassembled, allowing them to pass through the patient's mouth more comfortably and still provide an accurate impression. (Colvenkar, 2010; Kunusoth et al., 2022). Custom sectional trays also have been employed to make definitive impressions for both maxillary and mandibular arches, ensuring better access and accuracy (Luebke, 1984). In some cases, digital technologies like Computer-Aided Design and Computer-Aided Manufacturing (CAD/CAM) have been utilised to fabricate sectional custom trays and flexible dentures, offering a partial digital workflow that enhances patient's satisfaction and prosthetic outcomes (Zhang et al., 2021). Intraoral scanning combined with custom two-piece impression trays have also been reported to fabricate conventional dentures, demonstrating the integration of digital and traditional methods (Moslemian & Hasanzade, 2023). Despite these advancements, there are no clear guidelines that taking into consideration the severity of the patient's condition. Current practices often depend on modified tools and methods, such as sectional trays, which improve accuracy but may not suit every patient's needs. Some advancements, like digital technologies, have been introduced, but there is not a unified approach that combines these methods based on the severity of microstomia (al-Hadi, 1994). This lack of clear guidelines makes it difficult for clinicians to choose the best approaches for each patient, potentially affecting the quality of care and satisfaction (Carlow et al., 1987). Therefore, there is a need for a straightforward, evidence-based guidelines that combine existing techniques, enabling clinicians to select the most effective treatment approaches for microstomia patients.

1.3 Aims

The aim of this systematic review is to consolidate existing knowledge and to suggest prosthodontic treatment according to the severity of microstomia, to improve the quality of care for microstomia patients undergoing prosthodontic treatment.

1.4 Objectives

1. To critically review published case reports and case series on the prosthodontic treatment of microstomia patients.
2. To identify the most adopted approach to measure the severity of microstomia
3. To assess the correlation between prosthodontic treatment and the severity of microstomia.

1.5 Research questions

1. What is the most adopted approach to measure the severity of microstomia, and what factors contribute to the variation in measurement methods among different authors?
2. Is there any correlation between prosthodontic treatment and the severity of microstomia?

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CHAPTER 2: LITERATURE REVIEW

2.1 Aetiology of microstomia

Microstomia, marked by reduced mouth opening, can arise from various congenital or acquired factors, significantly impacting a patient's functional, psychological, and aesthetic appearance (Srivastava et al., 2022). Congenital microstomia can be a manifestation of systemic and inherited disorders, such as the Treacher Collins syndrome, Pierre Robin sequence, or hemifacial microsomia, The specific aetiologies in these conditions, however, often remain uncertain (Gülses, 2011; Wahle et al., 1992).

Acquired microstomia on the other hand, can results from various aetiologies, primarily involving trauma and pathological conditions. One of the most common causes is facial burns, which lead to hypertrophic and contracture scars, significantly reducing mouth opening and complicating management due to the involvement of perioral musculature (Thakur et al., 2020). Trauma, including electrical and thermal burns, ingestion of caustic substances, and reconstructive lip surgeries, can also result in cicatricial scar formation, further inhibiting adequate mouth opening (Gülses, 2011). Radiation therapy, particularly in the head and neck region, is another significant cause, leading to fibrosis and reduced elasticity of the tissues, thereby limiting mouth opening (Srivastava et al., 2022).

Furthermore, one of the notable manifestations of systematic sclerosis (SSc) is microstomia, due to fibrosis of the skin and mucosa, which complicates mastication, phonation, and oral hygiene (Kao et al., 2021; Ozatik et al., 2022). SSc also known as scleroderma, is a rare autoimmune connective tissue disease characterised by increased collagen synthesis, leading to multi-organ sclerosis, including the skin and joints, which significantly impacts patients' quality of life (Shir-Az et al., 2023).

Submucous fibrosis-induced microstomia is a condition characterised by a significant reduction in the size of the oral aperture due to the progressive fibrotic scarring of the oral soft tissues, which is a hallmark of oral submucous fibrosis (SMF) (Sheela & Rathika, 2017). This chronic and irreversible condition leads to complications such as burning mouth sensation, xerostomia, trismus, and severe limitation of mouth opening, which collectively contribute to the development of microstomia (Samieirad et al., 2018).

2.2 Epidemiological aspect

Microstomia incidence and prevalence vary based on the underlying causes and demographic factors. The condition is notably prevalent among children and adolescents due to their propensity for hypertrophic scarring, which often results from injuries or burns in the perioral region (Ismayilov et al., 2019). In adults, microstomia can arise from various aetiologies, including trauma, chemical burns, repeated excisions of skin cancer, and radiation therapy (Ki et al., 2020).

For example, chemical burns caused by the ingestion of caustic soda, often during suicide attempts, can cause severe oral and perioral contractures, leading to microstomia (Chidzonga, 2021). Additionally, according to Spanholtz and Giunta (2012), 3.7–10.8% of thermal burn admissions are complicated by reduction in the size of the oral aperture.

Joseph et al. (2023) conducted a study based on questionnaires and clinical examinations in three epidermolysis bullosa (EB) specialist centres. Among the 42 EB patients included in the study, 25 (59.5%) had dystrophic EB (DEB), 5 (11.9%) junctional EB (JEB; dermal-epidermal split), and 12 (28.6%) simplex EB (SEB; intra-epidermal cleavage). They found that microstomia was observed exclusively in cases of DEB, with an overall incidence rate of 40.0%.

Alarcón-Sánchez et al. (2024) systematically reviewed the oral findings and comprehensive dental management of Moebius syndrome (MS), noting that out of 124 cases, 15.32% of patients with MS have microstomia.

S. Zhang et al. (2021) assessed the association between oral manifestations and the presence of SSc compared to SSc-free populations. They found that the inter-incisor distance was significantly reduced in SSc patients. Similarly, Shionoya et al. (2020) reported that 70% of patients with SSc develop microstomia due to facial skin and oral mucosa fibrosis. Furthermore, Benz et al. (2021) revealed that the lips are the most commonly affected area in patients with scleroderma, with a prevalence of 57.6%.

Other than that, microstomia is identified by Gondivkar et al. (2020) as one of the primary signs in patients with SMF, a chronic, progressive, and potentially malignant condition that predominantly affects individuals in India (0.62-6.42%), Sri Lanka, China (1-3.03%), Vietnam (0.15-14.4%), and Taiwan (0.086-17.6%)

2.3 Normal mouth opening

Understanding the normal range and variability of mouth opening is vital in clinical practice as it is a diagnostic tool for conditions like trismus and microstomia. The assessment of mouth opening typically involves instructing the patient to open their mouth as widely as possible, followed by measuring the distance between the edges of the central incisors using a ruler or calliper (Mezitis et al., 1989). In a study involving 1,160 healthy Greek adults, it was found that, the mean maximal mouth opening was 52.5 mm for men and 47.1 mm for women (Mezitis et al., 1989). A similar study in another regions, Nepal, reported comparable mean values for mouth opening, highlighting some regional differences but overall consistent results across different populations, as noted by Cox and Walker (1997). This suggests that while there may be slight variations in

mouth opening measurements between regions, there is a general agreement on the average size among healthy adults.

Khare et al. (2012) conducted a study to determine the average mouth opening in 849 adults within the Indian population by measuring the distance between the edges of the incisors. They showed that males have a larger mouth opening compared to females across all age groups, with a significant decrease in mouth opening as age increases. Similarly, Yao et al. (2009) found that maximum mouth opening (MMO) significantly decreased with increasing age, regardless of sex.

While in Malaysia, Shaari et al. (2014) found that the range of mouth opening in adults can vary between 35 to 50 mm, with significant differences observed between males and females. Likewise, a study on the Indonesian population highlighted significant differences in mouth opening between genders and height groups, with males having larger mouth openings than females and taller individuals having larger mouth openings than shorter individuals (Rahmania et al., 2017).

Zawawi et al. (2003) found a strong positive correlation between MMO and 3-finger measurements in their study. They suggested a simple and reliable method to assess normal MMO is to determine the ability to accommodate the width of three fingers in the mouth during examination.

2.4 Clinical characteristics of microstomia

Assessing the severity of microstomia in patients involves both subjective and objective measures, considering individual anatomical features, functional limitations, and underlying causes. Naylor et al. (1984) proposed a method to measure the maximum oral aperture, categorising mild microstomia as an aperture between 41-50mm, moderate between 31-40mm, and severe if it does not exceed 30mm. They emphasised that this

classification is crucial for understanding the extent of the condition and planning appropriate interventions.

King (2016) proposed an index of oral access (IOA) that categorises the severity of the condition by evaluating the treatment access to posterior teeth (Table 2.1).

Table 2.1 The Index of Oral Access by King (2016)

IOA 0 – Normal	<ul style="list-style-type: none"> • Access to all areas of the mouth for all restorative treatment possible • Modification of impression technique or prosthetic design not required
IOA 1 – Mild	<ul style="list-style-type: none"> • Access to molar teeth restricted, complex treatment (endodontic treatment / indirect restorations) may be compromised or not possible • Minor modification of impression technique required to enable impression taking • Minor modification to prosthetic design required to enable insertion and removal
IOA 2 – Moderate	<ul style="list-style-type: none"> • Access to molar teeth for restorative treatment not possible • Access to premolar teeth restricted – complex treatment (endodontic treatment/indirect restorations) may be compromised or not possible • Access to incisors and canines for all treatment possible • Modification of impression trays required to enable impression taking • Modification to prosthesis design required to enable insertion and removal
IOA 3 – Severe	<ul style="list-style-type: none"> • Access to incisor and premolar teeth restricted – complex treatment (endodontic treatment/indirect restorations) may be compromised or not possible • Impression-taking severely compromised and significant modification to trays and technique required to enable impression taking • Significant and complex adjustments to prosthetic design required
IOA 4 – Extreme	<ul style="list-style-type: none"> • Access to all restorative treatment not possible • Impression-taking not possible • Prosthetic rehabilitation modification not possible

The IOA is defined by King (2016) as “a tool designed to increase the management of records, assist in diagnosing and planning restorative dentistry treatment, monitor the progression of diseases, encourage more objective treatment planning, and improve communication among clinicians”.

The IOA does not account for vertical mouth opening, which is crucial for assessing the full extent of functional impairment. For instance, a study on the use of 3D-printed microstomia orthoses demonstrated significant improvements in both horizontal and vertical mouth opening, highlighting the importance of considering vertical dimensions in treatment assessments (Edger-Lacoursière et al., 2024). Additionally, the IOA overlooks the complexities involved in prosthesis fabrication for microstomia patients. Traditional methods are often inadequate, necessitating modified techniques such as sectional impression trays and collapsible dentures to accommodate the limited oral opening (Kunusoth et al., 2022; Maria et al., 2022; Subba et al., 2022).

Maria et al. (2022) proposed a new classification system (Table 2.2) for microstomia that considers both the severity of the condition and the complexity of treatment options. The IOA is complemented by a diagnosis and management (DM) classification system, which considers additional factors such as vertical mouth opening, difficulty in prosthesis fabrication, manual dexterity, and treatment options. This DM classification further refines the treatment approach by categorizing microstomia into mild, moderate, and severe based on vertical mouth opening measurements, ranging from minimally compromised (31-35 mm) to severely compromised (21-30 mm).

Table 2.2 Classification systems based on diagnosis and management for microstomia patients proposed by Maria et al. (2022)

Class	IOA severity	Vertical mouth opening	Accessibility/Visibility	Treatment options	Prosthetic fabrication difficulty	Manual dexterity
DM-1	Mild	Minimally compromised: 31-35 mm	(i) Denture-bearing areas of the mouth are fully accessible and visible (ii) Impressions and JRR can be recorded easily	(i) Conventional removable dentures (ii) Implant-supported prosthesis (iii) Flexible dentures	Not technique sensitive	Adequate
DM-2	Moderate	Moderately compromised: 21-30mm	(i) Denture-bearing areas of the mouth have moderately compromised accessibility and visibility (ii) Moderately difficult to record impressions and JRR (modification of the tray/ technique is required*)	(i) Surgical correction (ii) Prosthodontic mx: (1) Implants supported/retained prosthesis (2) Flangeless prosthesis (3) Sectional complete removable dental prostheses can be used for the locking mechanism (4) Swing lock denture with cobalt-chromium framework	Moderately technique-sensitive, moderately skilled technician required	Fair
DM-3	Severe	Substantially compromised: 10-20 mm	(i) All the denture-bearing areas of the mouth have substantially compromised accessibility and visibility (ii) Extremely difficult to record impressions and JRR	(i) Surgical correction (ii) Prosthodontic mx: Sectional collapsible complete removable dental prosthesis	Highly technique-sensitive, highly skilled technician required	Poor
DM-4	Extreme	Severely compromised: <10 mm	(i) Denture-bearing areas hardly visible (ii) Impressions and JRR not possible	Prosthetic rehabilitation not possible	-	-

The classification system also outlines specific treatment options for each severity level, such as conventional removable dentures for mild cases and more complex prosthodontic solutions like sectional complete removable dental prostheses for moderate cases (Maria et al., 2022).

Despite the practical benefits of Maria et al.'s classification system, it's important to note that it is not evidence-based and may need further validation through clinical studies. The variability in patient's preferences and the subjective nature of aesthetic and functional assessments further complicate the standardisation of treatment protocols for microstomia (Ki & Park, 2024).

2.5 Treatment for microstomia

Microstomia can result from various causes leading to cicatricial scar formation, which affects both function and aesthetics (Nanda et al., 2016). Treatment options for microstomia include surgical, nonsurgical, and combined approaches, with the aim to enhance mouth opening, improve lip function, and achieve better aesthetic outcomes (Nanda et al., 2016).

According to Ki et al. (2020), several recommendations are proposed for treating microstomia. At the initial stages, it is advised to employ conservative treatment methods, such as oral splint appliances, physical therapy, and exercise. These approaches should be maintained until the patient adapts to microstomia and scar maturation is complete. When functional limitation of mouth opening persists despite implementing proper conservative treatment methods, such as oral splint appliances, physical therapy, and exercise, surgical intervention becomes the preferred method of choice. The decision to proceed with surgery is based on the necessity to restore adequate mouth function and improve the patient's quality of life. Selecting the appropriate surgical method is critical for effectively correcting microstomia and should be meticulously tailored to the severity

of the condition. Mild cases may require less invasive procedures, while more severe cases might necessitate complex reconstructive surgeries involving multiple tissue layers. This careful selection process ensures that the surgical intervention addresses the functional limitations and promotes optimal healing and a long-term maintenance of the reconstructed lip (Gülses, 2011).

2.5.1 Surgical therapy

Effective surgical treatment for individuals with microstomia must address two primary issues. First, it should restore the size of the oral opening by releasing the commissural contracture. Second, it should minimise the cosmetic defect caused by the deformation of the oral angle. By tackling both of these problems, surgical intervention can significantly improve both the functional and aesthetic outcomes for the patient (Grishkevich, 2011).

One common surgical approach is commissuroplasty, which aims to restore the oral commissure by correcting contractures. This procedure involves releasing or excising scarred contractures and covering the resulting soft tissue defect with local adjacent tissues, often using mucosal flaps from the oral cheek, which have proven more reliable than other methods (Spanholtz & Giunta, 2012). For patients with severe facial burns, surgical management often includes techniques such as triangular scar excision with mucosal advancement, scar excision followed by wound closure with full-thickness or split-skin grafts, and division of the contracture with closure using rhomboid mucosal flaps. These methods have shown good functional and aesthetic outcomes, with high patient satisfaction and minimal complications (Zweifel et al., 2010). The triangular scar excision and mucosal advancement method is advantageous for its simplicity and effectiveness in achieving good aesthetic and functional outcomes, but it may not be suitable for extensive scarring (Zweifel et al., 2010; Spanholtz & Giunta, 2012).

Another effective technique for post-burn microstomia is trapeze-flap plasty, which involves dissecting contracted scars with a Y-shaped incision and compensating for the scar-surface deficit with a mucosal trapezoid flap. This method addresses the contracture caused by the fold of scar and mucosal tissue, providing a reliable solution for restoring the oral opening size, however, it requires meticulous surgical planning and execution (Grishkevich, 2011). For scleroderma-related microstomia, hyaluronic acid injections have emerged as a novel and safe treatment modality. This method is minimally invasive and can significantly improve the oral aperture and quality of life but may not be as effective for severe cases and requires further large-scale studies to validate its efficacy (Shir-Az et al., 2023).

Each surgical option has its specific indications, and the choice depends on the severity of the microstomia, the extent of scarring, and the patient's overall condition. While commissuroplasty and mucosal flaps offer good functional and aesthetic results, they may not be ideal for extensive scarring, where skin grafting might be necessary despite its aesthetic drawbacks. Trapeze-flap plasty provides a comprehensive solution for severe cases but demands high surgical precision. Hyaluronic acid injections offer a less invasive alternative with promising results, particularly for systemic conditions like scleroderma, though they require further validation (Zweifel et al., 2010; Grishkevich, 2011; Spanholtz & Giunta, 2012; Shir-Az et al., 2023).

2.5.2 Non-surgical therapy

Non-surgical therapies for microstomia encompass a variety of approaches aimed at improving mouth opening and functionality without resorting to surgical intervention. One common method involves using splints and appliances designed to prevent tissue contraction and promote gradual stretching of the oral aperture. These devices can be intraoral or extraoral, static or dynamic, and are often customised to fit the patient's

specific needs. For instance, dynamic splints combined with intralesional injections of triamcinolone acetonide and hyaluronidase have been used effectively in cases of microstomia secondary to facial burns (Thakur et al., 2020).

In young children, microstomia prevention appliances are crucial, especially following oral burns, as they help maintain mouth opening during the healing process. These appliances are designed to be easily adjustable and minimally painful, ensuring better compliance and functional outcomes (Ajmal et al., 2019). Another innovative approach involves the use of a dynamic commissural appliance made from acrylic resin and expansion screws, which provides both horizontal and vertical stretching forces, allowing the patient to control the pressure applied, thus improving mouth opening and functionality (Antonarakis et al., 2017).

For congenital microstomia, non-surgical methods like using oesophageal balloons for dilation, followed by oral splints, have been effective in gradually enlarging the mouth opening without the need for surgery (Rezak et al., 2012). Additionally, photodynamic therapy (PDT) has been explored for its potential to reduce hypertrophic scarring and improve oral aperture by eliminating pathogenic microflora and enhancing clinical parameters (Lopez et al., 2020).

Psychological support and physiotherapy are also integral to non-surgical management, addressing the functional and aesthetic impairments affecting speech, chewing, and social interactions (Ismayilov et al., 2019). In patients with scleroderma, a multifaceted approach combining medical and physical therapies is recommended, although specific guidelines are lacking, and further research is needed to establish long-term efficacy (Gonzalez et al., 2021).

Postoperative adjuvant therapies, such as the use of splints, are essential in preventing the redevelopment of microstomia after surgical interventions, although their long-term use can sometimes cause discomfort (Makiguchi et al., 2014). Additionally, commissure splints and mouth exercises can be helpful in managing microstomia, particularly in edentulous patients who face challenges with denture insertion and removal (Nanda et al., 2016; Srivastava et al., 2022).

Overall, non-surgical therapies for microstomia are diverse and require a tailored approach to meet the individual needs of patients, aiming to improve their quality of life by enhancing oral function and aesthetics.

2.6 Prosthodontic treatment

Prosthodontic treatment in microstomia patients involves addressing the unique challenges posed by a significantly reduced oral opening and altered oral anatomy. Traditional techniques and materials often need to be adapted or replaced with specialised approaches to ensure successful outcomes. A thorough understanding of the patient's specific anatomical constraints and functional limitations is essential for devising an effective treatment plan.

2.6.1 Impression technique

Impression is required to fabricate any dental prosthesis. Standard stock impression trays, used in conventional practice, are often too large to fit comfortably within the restricted oral space. This can result in incomplete or distorted impressions, compromising the final prosthesis's fit and function. Due to the cumbersome nature of inserting and removing impression trays, various modifications to the trays have been used in the past.

These modifications include flexible trays and sectional trays, which are reassembled outside the mouth after the impression is taken (Aswini Kumar et al., 2013). Flexible impression techniques in microstomia are specialised methods designed to accommodate the restricted mouth opening of this condition (Dewan et al., 2015; Samet et al., 2007). For instance, flexible trays made from materials such as silicone or thermoplastic can be bent and adapted to capture accurate impressions of both hard and soft tissues (Samet et al., 2007).

One practical approach for preliminary impression is using segmented trays. These trays are typically divided into two or more segments that can be inserted separately into the mouth and then reassembled intraorally or extra orally using various mechanisms such as magnets, hinges, or Pindex systems (Colvenkar, 2010; Jabbari et al., 2014; Kunusoth et al., 2022). For instance, a reusable sectional handle with magnets allows for easy reassembly and disassembly, facilitating the impression process and saving clinical time (Colvenkar, 2010).

In some cases, modifying standard trays by trimming excess material or adjusting the shape can make them more manageable for microstomia patients. Sowmya et al. (2014) trimmed the tray from the buccal aspect, and the impression compound was adapted in the mouth in those regions manually. These modifications allow dental professionals to use readily available materials while still providing a tailored approach to impression-taking. Additionally, high-viscosity putty impression materials can be used with modified trays to capture detailed impressions in smaller sections. The putty can be easily manipulated within the confined space, ensuring accurate detail capture. Baslas et al. (2014) performed hand manipulation techniques using condensation silicone to acquire the first impressions.

For final impressions, the use of custom sectional trays, designed specifically for the patient's unique oral anatomy, offer the best fit and comfort. It can be made from a variety of materials, including light-cured acrylics, to accommodate different patients' needs and preferences, which can be inserted in segments and then reassembled intraorally to capture an accurate impression. Various techniques, such as Lego-like connectors (Kunusoth et al., 2022), magnets (Aswini Kumar et al., 2013), press button (Colvenkar, 2010) and pins (Kaira & Dabral, 2014) are used to join these segments. The use of these connectors is particularly beneficial as it allows for better stabilisation during border moulding and final impression making, enhancing the fit of the prosthesis (Subba et al., 2022; Thareja et al., 2019).

The advantages of impression-making techniques mentioned above include improved accuracy of the final prosthesis, enhanced patient comfort, and the ability to manage severe cases of microstomia effectively. However, there are also disadvantages, such as the complexity and time required for fabricating and using sectional trays and the potential for impression and cast deformation (Kumar et al., 2016; Maria et al., 2022). Despite these challenges, the advancements in impression-making techniques and materials continue to improve the outcomes for microstomia patients, offering them better functional and aesthetic results.

2.6.2 Digital impressions

Digital impression techniques offer significant benefits for microstomia patients, who often face challenges with traditional prosthetic rehabilitation due to their restricted mouth opening. The use of intraoral scanning (IOS) and computer-aided design and manufacturing (CAD/CAM) technologies has revolutionized the process, making it more efficient and comfortable for both patients and dental practitioners. Intraoral scanning, due to its reduced size compared to conventional impression trays, allows for easier

access and manoeuvrability within the limited oral cavity, thereby minimizing patient discomfort and the risk of impression deformation (Adali et al., 2019). This technique has been successfully employed to create preliminary impressions and edentulous models, which are then printed using 3D printers to fabricate custom trays and dentures (Saygılı et al., 2019).

For instance, Jagielska et al. (2024) described the use of a sectional custom tray designed through CAD/CAM technology to make border-moulded impressions, which still resulted in a retentive dental prosthesis (Jagielska et al., 2024). Similarly, another report highlighted the use of intraoral scanning and 3D printing to create sectional custom trays for definitive impressions, which were then used to fabricate conventional dentures, demonstrating the practicality and effectiveness of digital methods in such cases (Ozatic et al., 2022; Moslemian & Hasanzade, 2023).

The integration of digital technologies not only enhances the accuracy and fit of the prostheses but also reduces the overall treatment time and patient discomfort, as evidenced by multiple successful case reports (Thareja et al., 2019; S. Zhang et al., 2021). Moreover, digital impression techniques have been shown to be particularly beneficial for patients with severe microstomia, such as those with scleroderma, by providing an alternative to conventional methods that often fail to accommodate the anatomical limitations of these patients (Ozatic et al., 2022). Overall, the adoption of digital impression techniques in the prosthetic rehabilitation of microstomia patients represents a significant advancement, offering a more patient-friendly, precise, and efficient approach to dental care (Aswini Kumar et al., 2013).

2.6.3 Maxillomandibular relationship (MMR)

Traditional methods for recording MMR in patients with microstomia often require innovative approaches to ensure accurate prosthetic rehabilitation (Maria et al., 2022).

One effective technique involves using a sectional base that can be inserted in two pieces, enhancing stabilization during border moulding and final impression making, which helps create a better-fitting complete denture (Thareja et al., 2019). Additionally, magnets can be incorporated into the record bases to assist in the assembly and retention of the denture sections intraorally (Sankaran et al., 2023).

Digital techniques also offer promising solutions for MMR. For instance, an optical jaw tracking system can digitally record the maxillomandibular relationship, including maximum intercuspation and centric occlusion, integrating into a 3D virtual patient representation (Revilla-León et al., 2024). This digital approach can be further enhanced by using extraoral digital photographs and volumetric datasets from cone beam computed tomography (CBCT) imaging to create a photorealistic 3D virtual patient, facilitating diagnosis, communication, and patient acceptance of the treatment plan (Kuric et al., 2018).

Establishing the correct jaw relation is crucial, and while many techniques exist, none is universally superior (Özkan et al., 2018). The use of innovative impression techniques, such as interlocking custom trays and folding record bases, can significantly aid in recording accurate maxillomandibular relationships in microstomia patients (Garg et al., 2011).

2.6.4 Prosthesis

For individuals with microstomia, special attention is given to the design and fabrication of complete dentures to ensure proper fit and function within the restricted oral space. Traditional complete dentures often need to be adapted to accommodate the anatomical constraints of microstomia patients. Two effective strategies include shortening the denture flange and reducing the number or size of the teeth (Tayari et al., 2019; Zhang et al., 2021), both of which can markedly improve the comfort and

functionality of the prosthesis for individuals with restricted mouth openings. Furthermore, the incorporation of a long-term silicone soft liner on the tissue side of rigid denture bases, as suggested by Farhang et al. (2011) can alleviate trauma to the delicate tissues, enhancing comfort and reducing the risk of injury.

Sectional dentures present a practical solution for microstomia patients, who encounter significant challenges with conventional dentures due to their limited mouth opening. The primary advantage of sectional dentures lies in their ease of insertion and removal, which is crucial for patients with restricted oral access (Shams et al., 2020). Additionally, these dentures can be customised with simple attachments, such as press buttons, hinges, or magnets, each offering distinct benefits that reduce the need for the patient's manual dexterity (Baslas et al., 2014). Furthermore, sectional dentures enhance patient compliance by minimising tissue impingement and providing a more natural sensation during function (Baslas et al., 2014).

However, there are some disadvantages associated with sectional dentures. The fabrication process can be technique-sensitive and may require specialized skills, which can be a limitation in a predoctoral student clinic setting (Tulunoglu et al., 2018). The use of hinges and other attachments can complicate the design and may interfere with jaw relations and teeth arrangement (Kajave et al., 2015). Moreover, while sectional dentures are durable, the connections between sections, such as dovetails or magnets, may require precise adjustments to ensure proper occlusion and prevent movement of the denture segment (Baslas et al., 2014; Kajave et al., 2015).

Despite these challenges, sectional dentures remain a viable conservative approach for patients who prefer to avoid surgical interventions, offering a balance between functionality and ease of use (Baslas et al., 2014). The use of innovative materials and techniques, such as flexible impression trays and customized hinges, further enhances the

practicality and effectiveness of sectional dentures in managing microstomia (Kajave et al., 2015; Tulunoglu et al., 2018). Overall, while sectional dentures present some fabrication and adjustment challenges, their benefits in terms of patient comfort, ease of use, and cost-effectiveness make them a valuable option for prosthetic rehabilitation in microstomia patients.

Another common treatment option among dentists is the use of flexible dentures. Flexible dentures offer a viable solution for patients with microstomia. The primary advantage of flexible dentures is their ability to be deformed for easier insertion and removal, which is particularly beneficial for patients with limited oral aperture. These dentures are typically made from materials with a lower elastic modulus compared to traditional acrylic resins, making them easily deformable, elastic, and less prone to fracture. Once inserted, they quickly regain their original shape, providing a comfortable fit and enhanced functionality (Egan & Swindells, 2012; Zidani et al., 2018).

Additionally, flexible dentures are lightweight, colourless, and odourless, reducing the risk of allergies and ensuring high resistance to acids and alkalis. Their semi-transparent pink colour blends seamlessly with the gums, offering an aesthetic advantage by making the border between the denture base and gums difficult to distinguish (Zidani et al., 2018). The use of warm water to soften the denture before insertion further aids in achieving a good adaptation with the natural tissues in the mouth, enhancing patient comfort and ease of use (Zidani et al., 2018; Egan & Swindells, 2012).

However, there are some disadvantages associated with flexible dentures. The fabrication process can be complex, requiring precise techniques such as sectional or hinged designs, which may involve intricate mechanisms for connecting the components (Jagielska et al., 2024; Jivanescu et al., 2007). Moreover, while flexible dentures provide a practical solution for insertion and removal, they may not offer the same level of rigidity

and support as traditional dentures, potentially affecting their long-term durability and stability (Jivanescu et al., 2007; Samet et al., 2007).

Implant rehabilitation in patients with microstomia presents unique challenges and benefits. One of the primary advantages of using implants in these patients is the enhanced retention and stability of the prosthesis. For instance, a maxillary complete overdenture supported by dental implants can maintain its retention despite the progressive nature of microstomia, which might otherwise compromise the fit and function of conventional dentures (Langer et al., 1992). Additionally, implants can help distribute occlusal forces more evenly, reducing the risk of mucosal pain and tissue damage, which is particularly beneficial for patients with limited oral access and compromised mucosal health (Jensen & Sindet-Pedersen, 1990). The use of sectional dentures and implant-supported overdentures can also facilitate easier insertion and removal, accommodating the restricted mouth opening and improving the patient's ability to manage their prosthesis independently (Cheng et al., 2006; Langer et al., 1992).

However, there are notable disadvantages to consider. The surgical placement of implants in patients with microstomia can be technically challenging due to limited access and visibility, potentially requiring advanced surgical skills and specialized instruments (Cheng et al., 2006). Furthermore, the progressive nature of conditions like scleroderma can lead to changes in the oral tissues and bone structure over time, potentially affecting the long-term stability and success of the implants (Langer et al., 1992). Postoperative care and maintenance can also be more demanding, as patients with limited oral opening may struggle with adequate oral hygiene practices, increasing the risk of peri-implantitis and other complications (Jensen & Sindet-Pedersen, 1990).

CHAPTER 3: METHODOLOGY

3.1 Protocol and registrations

This systematic review was performed in accordance to Open Science Framework (OSF) and Preferred Reporting Items for Systematic reviews and Meta-Analyses (PRISMA). The protocol was submitted and registered on 13 December 2023. Registration DOI : <https://doi.org/10.17605/OSF.IO/Z67C4>

3.2 Ethical considerations and funding requisition

Ethical approval was not required because this study used acquired and synthesised data from previously published studies. This systematic study was funded by the Dental Postgraduate Research Grant (DPRG), Faculty of Dentistry, Universiti Malaya; Grant Number: UMG016E-2024.

3.3 Study design

In this study, systematic review was conducted. A systematic review is a structured approach to examine a particular clinical question by thoroughly reviewing a wide range of available literature while reducing bias. Systematic reviews are specifically developed to address specific clinical inquiries using the PICO (population, intervention, comparison, and outcome) framework (Smith & Duncan, 2022).

3.4 Eligibility criteria

The inclusion criteria for this study were designed to comprehensively cover all relevant and available literature about microstomia. 1) The study reviewed all case reports and case series available in English until May 2024. 2) The study focused on case reports of microstomia that provide detailed and clear assessments of vertical mouth opening. 3) The study included cases that outline prosthodontic treatments administered to patients with microstomia.

On the other hand, any cases with limited mouth opening related to temporomandibular joint (TMJ) problems were excluded.

3.5 Search methods for identification of studies`

3.5.1 Information sources

Combining search results from many databases improves coverage, recollection, and reduces the likelihood of forming incorrect conclusions during a review (Ewald et al., 2022). Table 3.1 lists various electronic databases that were utilized for conducting a comprehensive search. Each database is accompanied by its respective website link for accessing the advanced search options. The databases included are PubMed, Medline, EBSCO host, Scopus and Web of Science.

Table 3.1 List of information sources used in the study

Electronic Database	Website
Pubmed	https://pubmed.ncbi.nlm.nih.gov/advanced/
Medline	https://web.p.ebscohost.com/ehost/search/advanced
EBSCO host	https://research.ebsco.com/c/vy25p4/search/advanced/
Scopus	https://www.scopus.com/search/form.uri?display=advanced
Web of Science	https://www.webofscience.com/wos/woscc/advanced-search

3.5.2 Search terms

A literature search was conducted by applying specific Mesh terms and keywords, including those that are relevant and connected to: microstomia, limited oral access, limited oral opening, limited mouth opening, prosthodontic and prosthesis. The search strategies mentioned were developed using Boolean operators such as 'AND' and 'OR' to refine and optimise the retrieval of relevant studies from databases. Table 3.2 categorized the search queries into keywords based on the PICO.

Table 3.2 Keyword search

Elements	Keyword
Population	AB Microstomia OR MH microstomia OR AB "limited oral opening" OR AB "limited oral access" OR AB "limited mouth opening"
Intervention	None
Outcomes	AB Prosthodontic OR MH prosthodontic OR AB prosthesis OR MH prosthesis

3.5.3 Search strategy

Table 3.3 provides an overview of the final comprehensive search strategy detailing how the process was structured to ensure thoroughness and effectiveness in retrieving relevant literature.

Initially, a preliminary limited search of PubMed had been conducted, with the keyword selection primarily based on Patil et al. (2019). This search was followed by an examination of the titles, abstracts, and index terms of the retrieved articles. The search strategy had been developed using the PICO framework, beginning with the collection of relevant keywords, synonyms, and index terms. This process also helped identify a preliminary list of key papers, which was subsequently used to assess the effectiveness of the finalized search strategy.

Table 3.3 Final comprehensive search strategy across selected electronic databases

Database	Search Strategies
EBSCO host	AB (AB Microstomia OR SU microstomia OR AB "limited oral opening" OR AB "limited oral access" OR AB "limited mouth opening") AND AB (AB prosthodontic OR SU prosthodontic OR AB prosthesis OR SU prosthesis)
Scopus	((TITLE-ABS-KEY(Microstomia) OR TITLE-ABS-KEY("limited oral opening") OR TITLE-ABS-KEY("limited oral access") OR TITLE-ABS-KEY("limited mouth opening")) AND (TITLE-ABS-KEY(Prosthodontic) OR TITLE-ABS-KEY(prosthesis)) AND (LIMIT-TO (SUBJAREA,"DENT")))
PubMed	(((((microstomia[Title/Abstract] OR ("Microstomia"[Mesh])) OR ("limited mouth opening"[Title/Abstract]) OR ("limited oral opening"[Title/Abstract])) OR ("limited oral access"[Title/Abstract]))) AND (((prosthodontic[Title/Abstract] OR ("Prosthodontics"[Mesh])) OR (prosthesis[Title/Abstract])) OR ("Mandibular Prosthesis Implantation"[Mesh] OR "Maxillofacial Prosthesis Implantation"[Mesh] OR "Dental Prosthesis, Implant-Supported"[Mesh] OR "Dental Prosthesis"[Mesh] OR "Maxillofacial Prosthesis"[Mesh] OR "Mandibular Prosthesis"[Mesh] OR "Denture, Partial, Temporary"[Mesh] OR "Dental Implants"[Mesh])))
Medline	(AB Microstomia OR MH microstomia OR AB "limited oral opening" OR AB "limited oral access" OR AB "limited mouth opening") AND (AB Prosthodontic OR MH prosthodontic OR AB prosthesis OR MH prosthesis)
Web of Science	((((TS=(microstomia)) OR KP=(microstomia)) OR TS=("limited oral opening")) OR TS=("limited oral access")) OR TS=("limited mouth opening") AND (((TS=(prosthodontic)) OR KP=(prosthodontic)) OR TS=(prosthesis)) OR KP=(prosthesis)

A secondary search was then conducted by first reviewer (K.N), an independent, yet meticulous and thorough search of multiple online databases/ search engines, including PubMed, MEDLINE, EBSCO Host, Web of Science, and Scopus, utilizing all identified keywords, synonyms, and index terms. This electronic search spanned from early March 2024 to early May 2024, with the final search performed on May 7, 2024. The complete search strategy is detailed in Table 3.3.

3.6 Data extraction and management

3.6.1 Sources of evidence selection/ screening

The sources of evidence were chosen based on the search strategy and predetermined inclusion and exclusion criteria, as outlined in Table 3.2. Overall, the steps involved were:

1. The literature search involved querying multiple electronic databases and reviewing references of articles and other relevant sources.
2. Duplicates were removed from the dataset.
3. Screening of title and abstracts
4. Screening/assessment of eligibility of full-text articles
5. Compiling the studies to include them for data extraction.

In this study, a single reviewer, K.N., carried out the first two steps, which involved searching databases and registrations and manually searching for journals. The reports retrieved were exported to the Endnote X20 software, and the duplicates, if any, were removed. Subsequently, two reviewers (K.N. and E.A.E.) conducted the screening process starting from Step 3, which involved screening reports to be retrieved, assessing reports for eligibility, and excluding reports based on study types, duplicates, and abstracts. This was done after successfully identifying the list of references from the search. Any differences between the two reviewers were resolved through discussion.

The first reviewer (K.N.) requested via Universiti Malaya Library Publication Supply for full publications that were unavailable online.

3.6.2 Data extraction

The first reviewer (K.N.) created a standardised data extraction sheet. This sheet was subsequently cross-checked by the second, third, and fourth reviewers (E.A.E., S.F.A., R.A.O.). Any discrepancies that arose were resolved through collaborative discussion

among all the reviewers. The first and second (K.N and E.A.E) extracted the following data independently:

1. Author and year of publication
2. Maximum mouth opening (MMO)
3. Aetiology of microstomia
4. Dentition status
5. Primary impression technique
6. Final impression technique
7. The prosthodontic treatment applied for each case

3.6.3 Data synthesis and analysis

The gathered data were systematically arranged into specific thematic categories using IBM SPSS Statistics (Version 26), a statistical software specifically developed for comprehensive data management and descriptive analysis. The data were first examined and classified into overarching thematic categories based on their content and relevance, with each data entry allocated to the category that most accurately reflected its primary theme or subject.

Within SPSS, each thematic category was defined as a variable, and data entries were organised methodically under these variables. This approach ensured precise categorisation and enabled straightforward access for subsequent analysis.

This organisational process allowed for efficient handling of the data, enabling various types of statistical analysis. For example, frequency analysis could be performed to determine the prevalence of each theme, identifying significant trends and patterns within the dataset. Additionally, other statistical techniques available in SPSS could be used to

explore relationships between different themes, measure variability, and conduct cross-tabulations.

By organising the data according to their respective thematic categories in SPSS, the analysis became more structured and coherent. This methodical approach facilitated a comprehensive examination of the data, enhancing the ability to draw meaningful conclusions and insights from the study.

3.6.4 Quality assessment of case reports and case series

To assess the quality and reliability of the included studies, the Joanna Briggs Institute (JBI) Critical Appraisal Tools was utilised. The JBI Critical Appraisal Tools were integral to the systematic review process, providing a structured approach to assess the risk of bias in various study designs, including case series and case reports. These tools consist of ten questions for case series and eight for case reports, each designed to yield 'Yes', 'No', 'Unclear', or 'Not Applicable' responses. The percentage of 'Yes' answers determines the study's risk of bias: high risk with 49% or fewer 'Yes' answers, moderate risk with 50-69%, and low risk with over 70% 'Yes' answers. This categorisation is crucial for ensuring the reliability and validity of systematic reviews, as it directly impacts the confidence in the synthesised findings (Aromataris, 2023; Barker et al., 2023). The importance of critical appraisal in evidence-based practice cannot be overstated, as it helps distinguish high-quality studies from those with methodological flaws, thereby guiding clinical decision-making and policy formulation (Stone et al., 2023).

CHAPTER 4: RESULT

4.1 Critically reviewed case reports and cases series

4.1.1 Outcome of search strategy and selection of sources of evidence/ reference

Figure 4.1 offers a detailed visualization of the flow of information through the various stages of this study. It outlines the initial number of records that were identified and provides a breakdown of how many of these records were eventually included in the review versus those that were excluded. The figure also meticulously documents the reasons behind each exclusion, offering a transparent account of the decision-making process. This comprehensive mapping helps to clarify how the study's final dataset was determined and ensures that the criteria for inclusion and exclusion are fully transparent and well-documented.

The PRISMA flow diagram illustrates the systematic process of identifying, screening, and selecting studies for inclusion in a systematic review. The process began with the identification of studies through databases and registers. A total of 722 reports were initially retrieved from five major databases: PubMed, MEDLINE, EBSCO Host, Scopus, and Web of Science. However, before the screening process could begin, 443 duplicate records were identified and removed. Two reviewers independently screened the remaining studies by evaluating the titles and abstracts to identify relevant articles. This process resulted in the exclusion of an additional 31 reports, leaving 124 unique reports for eligibility assessment.

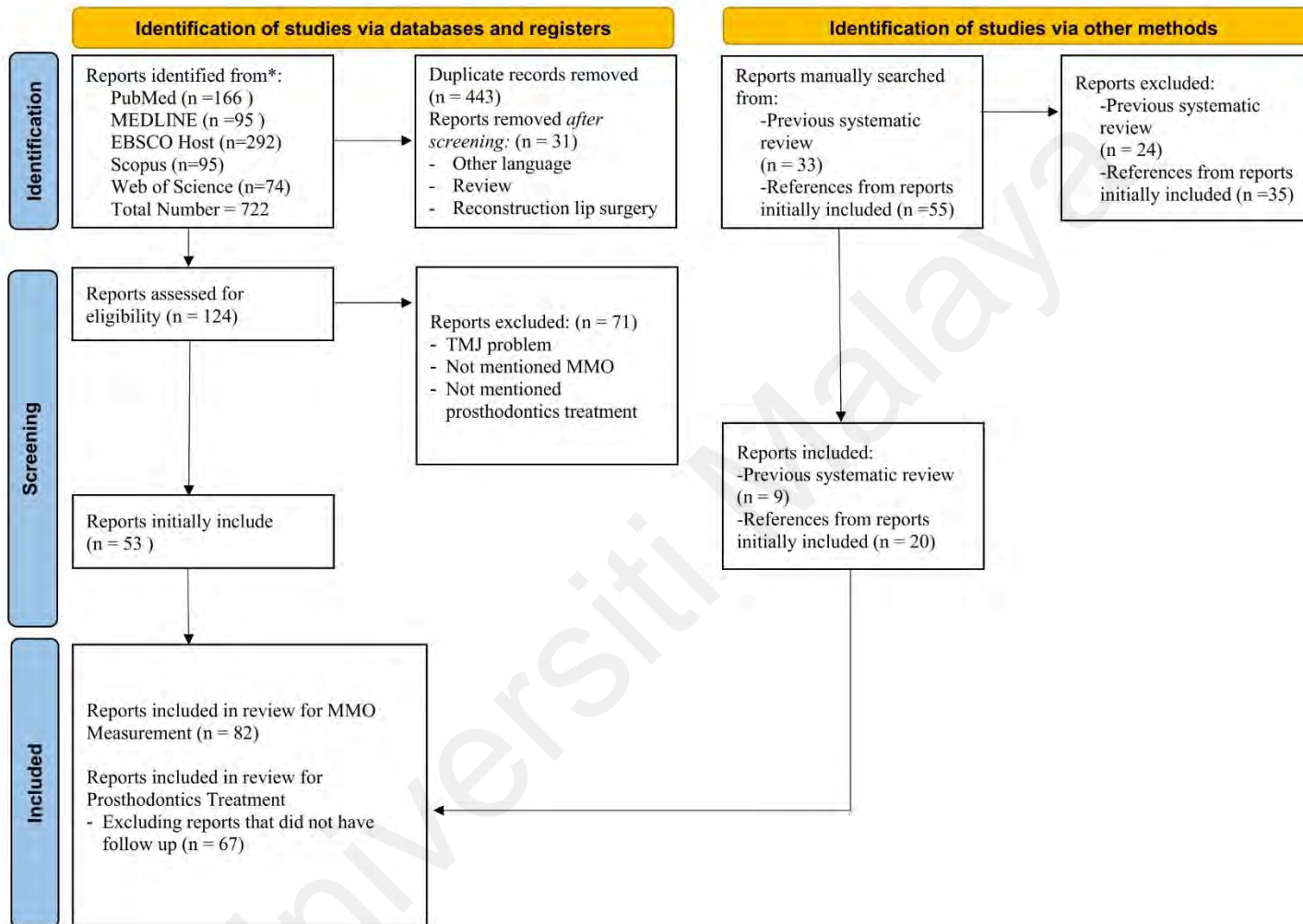


Figure 4.1 Summary of the Search Strategy

The eligibility assessment phase involved a thorough review of the full text of these 124 reports to determine their relevance and suitability for inclusion in the review. Out of these, 71 reports were excluded for various reasons, such as focusing on TMJ problems, not addressing Maximum Mouth Opening (MMO) or prosthodontic treatment, not being case reports or case series, or being published in languages other than the ones accepted by the review. This exclusion process resulted in 53 reports being initially included in the review.

In addition to database searches, the review also involved identifying studies through other methods, such as manual searches from previous systematic reviews and references from initially included reports. This effort added another 88 reports to the pool, though 59 of these were excluded due to redundancy with previous systematic reviews or as references from reports already included. Ultimately, 29 additional reports were included, 9 from previous systematic reviews and 20 from reference lists.

The final step in the process resulted in the inclusion of 82 reports for the review on MMO Measurement and 67 reports for Prosthodontics Treatment, excluding those that did not provide follow-up data. This thorough and methodical approach ensures that the studies included in the systematic review are both relevant and of high quality, providing a solid foundation for the review's conclusions.

4.1.2 Data extraction sheet

All 82 reports were reviewed, and the necessary details were systematically compiled into a table extraction sheet.

Table 4.1 Table extraction sheet

No	Author and Year	MMO	POM	Aetiology	Dentition Status	Primary Impression	Final Impression	Prosthesis	Follow Up
1	Adali et al. (2019)	25mm	lip to lip	surgery	combination	not mentioned	digital	Conventional denture (telescopic crown)	/(3y)
2	Afroz et al. (2012)	26mm	interridge	submucous fibrosis	edentulous	conventional	conventional	Conventional denture	/(6m)
3	Basavanna and Raikhy (2013)	26mm	lip to lip	scleroderma	edentulous	flexible	sectioned	Sectional denture with press buttons	NA
4	Baslas et al. (2014)	25mm	lip to lip	burn	edentulous	flexible	sectioned	Sectional denture with lock and keys	/(3m)
5	Dewan et al. (2015)	24mm	lip to lip	developmental	edentulous	sectional	sectioned	Sectional denture with magnet	/(6m)
6	Dhanasomboon and Kiatsiroj (2000)	30mm	interincisal	scleroderma	partially dentate	flexible	sectioned	Conventional denture	/
7	Farhang et al. (2011)	35mm	horizontal	epidermolysis bullosa	edentulous	flexible	conventional	Conventional denture (with soft reliner)	/(3w)
8	Fatemeh et al. (2021)	26mm	not mentioned	burn	edentulous	flexible	sectioned	Conventional denture	NA
9	Garcés Villalá and Zorrilla Albert (2021)	25mm	lip to lip	scleroderma	combination	not mentioned	not mentioned	Implant with fixed prosthesis	/(10y)
10	Garg et al. (2011)	20mm	not mentioned	radiotherapy	partially dentate	small tray	conventional	Conventional denture	/(1w)
11	Geckili et al. (2006)	25mm	not mentioned	surgery	combination	not mentioned	sectioned	Sectional folded denture with hinge	/(15m)

No	Author and Year	MMO	POM	Aetiology	Dentition Status	Primary Impression	Final Impression	Prosthesis	Follow Up
12	Gauri and Ramandeep (2013)	27mm	lip to lip	submucous fibrosis	edentulous	sectional	combination	Maxilla sectional denture (magnet) and conventional denture (mandible)	/(2y)
13	Givan et al. (2010)	20 mm	lip to lip	surgery	edentulous	shortened flange	sectioned	Sectional folded denture with hinge	NA
14	Gözde Türk and Ulusoy (2015)	23mm	interincisal	scleroderma	partially dentate	sectional	sectioned	Conventional denture	/(4m)
15	Hajimahmoudi and Mostafavi (2014)	30mm	lip to lip	scleroderma	edentulous	flexible	sectioned	Sectional denture with lock and keys (telescopic framework)	/
16	Dikbas et al. (2007)	34mm	horizontal	scleroderma	edentulous	sectional	sectioned	Anterior segment and hinged posterior segment of the maxillary denture. Sectioned mandibular denture (pin)	/(2m)
17	Kaira and Dabral (2014)	36mm	horizontal	scleroderma	edentulous	sectional	sectioned	Sectional denture with press buttons	/
18	Kam et al. (2006)	20mm	interincisal	scleroderma	partially dentate	not mentioned	not mentioned	Sectional denture with magnet	/
19	Kumar et al. (2020)	25mm	lip to lip	developmental	edentulous	sectional	sectioned	Sectional folded denture with hinge	/(1y)
20	Kumar et al. (2012)	25mm	lip to lip	developmental	edentulous	flexible	sectioned	Sectional folded denture with hinge	/(2y)
21	Maria et al. (2022)	20mm	Interridge	surgery	edentulous	combination	sectioned	Hinge-fold denture	/

No	Author and Year	MMO	POM	Aetiology	Dentition Status	Primary Impression	Final Impression	Prosthesis	Follow Up
22	Langer et al. (1992)	28mm	not mentioned	scleroderma	partially dentate	not mentioned	not mentioned	Implant overdenture (maxilla). Implant supported fixed prosthesis (mandible)	NA
23	Moslemian and Hasanzade (2023)	35mm	horizontal	burn	edentulous	digital	sectioned	Conventional denture	/(5y)
24	Paes-Junior et al. (2021)	30mm	interincisal	scleroderma	partially dentate	conventional	sectioned	Acrylic removable partial denture	/(1y)
25	Prasad et al. (2008)	25mm	lip to lip	developmental	edentulous	sectional	sectioned	Sectional denture with press button	/
26	Rathi et al. (2013)	25mm	not mentioned	muscular dystrophy	combination	not mentioned	sectioned	Sectioned folding denture with hinge	NA
27	Ravi et al. (2021)	18mm	lip to lip	submucous fibrosis	edentulous	flexible	sectioned	Sectioned denture with press button	/(6m)
28	Samet et al. (2007)	14mm	interincisal	scleroderma	partially dentate	not mentioned	sectioned	Thermoplastic denture	/
29	Saraf et al. (2014)	22mm	lip to lip	submucous fibrosis	partially dentate	shortened flange	sectioned	Conventional partial denture	NA
30	Satpathy and Gujjari (2015)	23mm	interridge	developmental	edentulous	sectional	sectioned	Maxilla sectioned denture with press button (anterior telescopic). Mandible sectioned denture (cross pin)	/(1m)
31	Saygılı et al. (2019)	25mm	lip to lip	surgery	edentulous	digital	sectioned	Sectioned folding denture with hinge	/(1y)

No	Author and Year	MMO	POM	Aetiology	Dentition Status	Primary Impression	Final Impression	Prosthesis	Follow Up
32	Shams et al. (2020)	25mm	not mentioned	burn	combination	sectional	sectioned	Maxilla sectional denture. Mandible conventional cobalt chrome denture.	/
33	Sharma et al. (2013)	25mm	not mentioned	submucous fibrosis	edentulous	small tray	sectioned	Sectioned folding denture with hinge	/(1y)
34	Sheela and Rathika (2017)	28mm	lip to lip	submucous fibrosis	edentulous	flexible	sectioned	Sectioned denture with press button	/
35	Silvestri et al. (2023)	20mm	not mentioned	surgery	partially dentate	digital	sectioned	Implant supported fixed prosthesis	/
36	Sowmya et al. (2014)	31mm	not mentioned	submucous fibrosis	edentulous	shortened flange	sectioned	Conventional denture (mouth opening improved)	NA
37	Srivastava et al. (2022)	28mm	lip to lip	radiotherapy	edentulous	small tray	sectioned	Sectioned folding denture with hinge	/(6m)
38	Sun et al. (2012)	40mm	horizontal	burn	combination	small tray	sectioned	Sectioned denture (retained by metal ceramic crown)	/(3y)
39	Tayari et al. (2019)	21mm	not mentioned	radiotherapy	edentulous	small tray	sectioned	Conventional denture (reduces size of teeth)	/(1w)
40	Tayari et al. (2019)	27mm	not mentioned	radiotherapy	edentulous	flexible	combination	Conventional denture	/
41	Tayari et al. (2019)	30mm	not mentioned	surgery	edentulous	conventional	conventional	Conventional denture	NA
42	Watanabe et al. (2002)	32mm	horizontal	scleroderma	edentulous	conventional	sectioned	Sectioned folding denture with hinge and magnet	/(2y)

No	Author and Year	MMO	POM	Aetiology	Dentition Status	Primary Impression	Final Impression	Prosthesis	Follow Up
43	Zhang et al. (2021)	40mm	horizontal	scleroderma	partially dentate	not mentioned	digital	Conventional cobalt chrome denture (reduced number of teeth)	/(6m)
44	Zhang et al. (2020)	27mm	lip to lip	scleroderma	combination	digital	digital	Conventional cobalt chrome denture	/(10m)
45	Balakrishnan J et al. (2020)	30mm	horizontal	submucous fibrosis	edentulous	sectional	sectioned	Conventional denture	NA
46	Benetti et al. (2004)	40mm	lip to lip	scleroderma	partially dentate	flexible	sectioned	Sectioned folding denture with hinge	/(3y)
47	Cheng et al. (2006)	28mm	interridge	necrotising fasciitis	edentulous	flexible	conventional	Implant supported overdenture	/(6m)
48	Cheng et al. (2000)	30mm	lip to lip	surgery	edentulous	flexible	conventional	Conventional denture (shortened arch)	/
49	Colvenkar (2010)	25mm	not mentioned	scleroderma	edentulous	flexible	combination	Conventional denture (maxilla). Sectioned denture with magnet (mandible)	/
50	Sahoo et al. (2013)	26mm	lip to lip	scleroderma	edentulous	small tray	sectioned	Conventional dentures	/
51	Egan and Swindells (2012)	30mm	lip to lip	surgery	edentulous	flexible	conventional	Flexible denture	/(6m)
52	Ohkubo et al. (2003)	25mm	not mentioned	rheumatoid arthritis	partially dentate	sectional	sectioned	Sectioned folding denture with hinge and magnet	NA

No	Author and Year	MMO	POM	Aetiology	Dentition Status	Primary Impression	Final Impression	Prosthesis	Follow Up
53	Jain et al. (2019)	16.6mm	lip to lip	radiotherapy	edentulous	sectional	combination	Maxilla sectioned denture with magnet (anterior telescopic). Mandible conventional denture.	/(1m)
54	Jivanescu et al. (2007)	18mm	ridge to incisal	scleroderma	combination	sectional	conventional	Thermoplastic denture (maxilla). No treatment for mandible	/
55	Kajave et al. (2015)	28 mm	interridge	submucous fibrosis	edentulous	combination	combination	Maxilla conventional denture. Mandible sectioned folding denture with hinge	/(6m)
56	Klostermyer et al. (2011)	21mm	lip to lip	burn	combination	sectional	sectioned	Implant with sectional folding overdenture (mandible)	/
57	Kamadajaja and Pertiwi (2019)	25mm	lip to lip	scleroderma	edentulous	small tray	conventional	Conventional dentures	/(6m)
58	Patil et al. (2013)	32mm	horizontal	surgery	combination	sectional	sectioned	Maxilla conventional denture. Mandible sectioned folding denture with hinge	/(6m)
59	Prasad D et al. (2012)	20mm	not mentioned	submucous fibrosis	edentulous	shortened flange	sectioned	Conventional denture	NA

No	Author and Year	MMO	POM	Aetiology	Dentition Status	Primary Impression	Final Impression	Prosthesis	Follow Up
60	Ravindran et al. (2012)	21mm	lip to lip	burn	partially dentate	shortened flange	sectioned	Conventional denture	NA
61	Singh et al. (2014)	23mm	lip to lip	scleroderma	partially dentate	flexible	not mentioned	Thermoplastic denture	/(2y)
62	Singh et al. (2020)	35mm	not mentioned	submucous fibrosis	edentulous	sectional	sectioned	Sectional prosthesis button and pin	NA
63	Siwach and Siwach (2011)	10mm	lip to lip	scleroderma	combination	flexible	sectioned	Sectioned folding denture with hinge	/
64	Tripathi et al. (2011)	15mm	interincisal	radiotherapy	partially dentate	shortened flange	conventional	Conventional denture	/(6m)
65	Tulunoglu et al. (2018)	37mm	horizontal	burn	edentulous	small tray	sectioned	Sectioned folding denture with hinge	/(3m)
66	Yenisey et al. (2005)	40mm	not mentioned	scleroderma	edentulous	flexible	sectioned	Maxilla conventional denture. Mandible sectioned folding denture with hinge	/(6m)
67	Fischer and Patton (2000)	25mm	not mentioned	scleroderma	edentulous	shortened flange	conventional	Conventional denture (minimal interocclusal space)	/(6m)
68	Bidra et al. (2010)	33mm	lip to lip	surgery	edentulous	flexible	not mentioned	Maxillary obturator with 3 magnets placed on flattened nasal extension. (both conventional denture)	/(1y)
69	Cheng et al. (2001)	34mm	not mentioned	surgery	partially dentate		combination	Cobalt chrome partial denture	NA

No	Author and Year	MMO	POM	Aetiology	Dentition Status	Primary Impression	Final Impression	Prosthesis	Follow Up
70	Bilhan et al. (2011)	25mm	not mentioned	developmental	edentulous	flexible	sectioned	Implant supported fixed hybrid prosthesis (maxilla) and implants supported overdenture (mandible)	/(1y)
71	Zidani et al. (2018)	15mm	lip to lip	radiotherapy	combination	flexible	conventional	Thermoplastic denture	/(6m)
72	Jagielska et al. (2024)	20mm	ridge to incisal	scleroderma	combination	digital	sectioned	Thermoplastic mandibular denture. Conventional denture at maxilla	/
73	Jensen and Sindet-Pedersen (1990)	20mm	interincisal	scleroderma	combination	not mentioned	not mentioned	Implant supported bridge (mandible only)	/(2y)
74	McKenna et al. (2012)	19mm	interincisal	scleroderma	partially dentate	sectional		Resin bonded bridge	/
75	Sehgal et al. (2011)	28mm	not mentioned	scleroderma	edentulous	shortened flange	sectioned	Sectional tray with magnet	/
76	Suzuki et al. (2000)	33mm	horizontal	burn	partially dentate	sectional	sectioned	Mandibular denture collapsed with midline hinge and cast swing-lock attachment anterior. Maxilla conventional denture.	/(3y)

No	Author and Year	MMO	POM	Aetiology	Dentition Status	Primary Impression	Final Impression	Prosthesis	Follow Up
77	Zigdon et al. (2011)	30mm	not mentioned	scleroderma	edentulous	not mentioned	not mentioned	Implant supported fixed prosthesis	/(3y)
78	Nam et al. (2012)	40mm	not mentioned	scleroderma	edentulous	not mentioned	not mentioned	Implant with fixed prostheses	NA
79	Patil and Nimbalkar-Patil (2016)	25mm	lip to lip	surgery	partially dentate	shortened flange	not mentioned	Obturator with implant for retention (no teeth)	/(18m)
80	Ravi et al. (2022)	17mm	not mentioned	surgery	partially dentate	sectional	sectioned	Sectioned denture with intraoral magnet	/
81	Sinavarat and Anunmana (2015)	32mm	horizontal	scleroderma	edentulous	sectional	sectioned	Segmental denture with hinge.	NA
82	Subba et al. (2022)	29mm	lip to lip	scleroderma	edentulous	small tray	sectioned	Conventional dentures	/(1m)

4.2 The most adopted approach to measure severity of microstomia

Table 4.2 presents a comprehensive analysis of the various points of measurement used to assess severity of microstomia among three different groups: edentulous, partially dentate, and combination cases (those with both edentulous and partially dentate conditions) indicating the various anatomical landmarks used in the assessment.

Table 4.2 Demographic variables

Sociodemographic Characteristics	Frequency	
	n	%
Point of Measurement (Edentulous)		
Lip to lip	18	46.2
Interridge	5	12.8
Horizontal	7	17.9
Not mentioned	9	23.1
Point of Measurement (Partially Dentate)		
Lip to lip	3	20.0
Interincisal	7	46.7
Horizontal	2	13.3
Not mentioned	3	20.0
Point of Measurement (Combination)		
Lip to lip	6	46.2
Interincisal	1	7.7
Ridge to incisal	2	15.4
Horizontal	2	15.4
Not mentioned	2	15.4
Severity of Microstomia		
Edentulous		
0-19 mm (severe)	3	6.3
20-40 mm (moderate)	45	93.8
Partially dentate		
0-24 mm (severe)	11	55.0
25-40 mm (moderate)	9	45.0
Combination		
0-24 mm (severe)	6	42.9
25-40 mm (moderate)	8	57.1

In this table, most cases reported the use of lip-to-lip technique to measure MMO in edentulous (46.2%) as in Appendix C and combination cases (46.2%) as in Appendix D.

For partially dentate patients, the interincisal technique was the preferred method for measuring MMO (Appendix E). In contrast, it's notable that 29.3% of case reports did not specify the measurement point.

4.3 Correlation between severity of microstomia with prosthodontics treatment

4.3.1 Classification of microstomia severity according to dentition status

The maximal mouth opening (MMO) values presented in all reports are stratified based on the dental status of the subjects. The severity of these values is classified according to parameters wherein the authors did not utilize conventional dentures for the specific measurement of MMO, as illustrated in Table 4.1.

For edentulous patients, there are no conventional denture records available for MMO measurements that fall below 19mm. Given this, measurements of MMO less than 19mm were categorized as 'severe' and consolidated into a distinct group. Another group has been formed for measurements that range between 20mm and 40mm, which are classified as 'moderate.'

In combination cases, the maximum mouth opening continues to impact the authors' selection of prosthodontic treatment. In this study, conventional denture records are not obtainable for measurements below 24mm. Consequently, two distinct groups were created: one group encompasses measurements below 24mm (severe), while the other includes measurements ranging from 25mm to 40mm (moderate).

However, this pattern does not apply to individuals with partial dentition. In these cases, conventional partial dentures typically cover only a specific, limited area of the mouth, rather than providing full coverage. This limited coverage makes the insertion and removal of the dentures more manageable and less complex compared to full dentures.

Consequently, for patients with partial dentition, K.N. has established a group classification that mirrors the approach used for combination cases, where both edentulous and partially dentate arches are present (Table 4.2).

4.3.2 The correlation

From the original set of 82 case reports, fifteen case reports (9 edentulous cases, 5 partially dentate cases, and one combination case) were excluded at this stage because they did not provide any follow-up information for the patients. After exclusions, 67 case reports remain for correlation analysis. Table 4.3 presents an analysis of prosthodontic treatment options in relation to the severity of microstomia and different dentition status.

It categorises patients into three groups: edentulous, partially dentate, and combination cases (having edentulous opposing partially dentate arches). The severity of microstomia is divided into two groups, with Group I indicate severe cases and Group II representing moderate cases, as outlined in Table 4.1. Various prosthodontic treatments are listed, including conventional dentures, sectional dentures, flexible dentures, implant-supported overdentures, and implant-supported fixed prostheses.

Table 4.3 The severity of microstomia in relation to dental condition compared to the prosthodontic treatment provided

Dentition Status	Group	MMO (mm)	Prosthodontic							Fisher Exact Test	p-value
			Conventional denture	Sectional denture	Combination**	Flexible denture	Implant Overdenture	Implant FP	Other		
Edentulous	I	0 - 19	0	1	1	0	0	0	0	7.400	0.526
	II	20 - 40	11	17	4	1	2	1	1		
	Total		11	18	5	1	2	1	1		
Combination*	I	0 - 24	0	1	0	3	1	1	0	7.372	0.176
	II	25 - 40	2	2	2	0	0	1	0		
	Total		2	3	2	3	1	2	0		
Partially Dentate	I	0 - 24	3	2	0	2	0	1	1	4.474	1.000
	II	25 - 40	2	1	1	1	1	0	0		
	Total		5	3	1	3	1	1	1		

*Combination refers to edentulous opposing partially dentate.

**Combination technique refers to using a conventional denture on one side and a sectional denture on the opposing side.

Fisher's Exact Test was applied. Significant at $p < 0.05$.

Group I referred to severe microstomia

Group II referred to moderate microstomia

4.3.3 Correlation between severity of microstomia with prosthodontics treatment in edentulous patients.

Table 4.3 indicates that there is no statistically significant correlation between the severity of microstomia in edentulous patients and the type of prosthodontic management employed, as evidenced by a *p*-value exceeding 0.05. Furthermore, it is observed that none of the authors employed conventional dentures for edentulous patients exhibiting a maximum mouth opening (MMO) of less than 19 mm.

In group I, the range of prosthodontic treatments is somewhat limited. Ravi et al. (2021) chose to utilise sectional dentures with press buttons for both the maxilla and mandible in a patient with MMO of 18mm. In contrast, Jain et al. (2019) favoured a three-segmental denture with a hinge in the middle for the maxilla, supported by the anterior segment, and a conventional denture for the mandible in a patient with an MMO of 17mm.

In group II, a variety of prosthodontic treatments were examined. The most utilised technique was the sectional denture, followed by conventional dentures, a combination of sectional-conventional dentures, and implant-supported prostheses.

Among the studies, only Egan and Swindells (2012) employed flexible dentures made from ClearSplint acrylic, a hybrid acrylic material that can be repaired using chemically cured resin, offering a benefit in case of tooth loss from the prosthesis. On the other hand, Bilhan et al. (2011) conducted dental implant surgery and utilised a fixed hybrid prosthesis in the maxilla, along with an overdenture in the mandible.

4.3.4 Correlation between severity of microstomia with prosthodontics treatment in combination cases.

For combination cases, where one arch is edentulous, the maximum mouth opening still plays a significant role in the author's decision regarding the use of prosthodontic treatment. From Table 4.3, in Group I, which represents more severe MMO (0 - 24 mm), the distribution of prosthodontic treatments is as follows: no patient received a conventional denture, one patient received a sectional denture, no patient received a combination of sectional-conventional dentures, three patients received flexible dentures, one patient received an implant-supported overdenture, and one patient received an implant-supported fixed prosthesis.

In Group I, Siwach and Siwach (2011) constructed an acrylic denture segmented into two sections, featuring a hinge assembly specifically designed for patients with MMO of 10mm. In contrast, three other authors opted to provide flexible dentures to their patients with varying MMO measurements of 20mm (Jagielska et al., 2024), 18mm (Jivanescu et al., 2007), and 15mm (Zidani et al., 2018), respectively. Jensen and Sindet-Pedersen (1990) focused on utilising osseous integrated implant-supported bridge in a patient with an MMO of 20mm, demonstrating successful outcomes in both implant integration and improvement in oral function. Additionally, Klostermyer et al. (2011) emphasised the utilisation of a sectional folding overdenture implant in the mandible of a patient with an MMO of 21mm.

In Group II from Table 4.3, which encompasses less severe microstomia (25-40 mm), the distribution of prosthodontic treatments is different: two patients received conventional dentures, two patients received sectional dentures, two patients received a combination of sectional-conventional dentures, and one patient received an implant-supported fixed prosthesis.

To assess the statistical significance of the distribution of the treatments across the two groups, the Fisher Exact Test was employed. No significant correlation was found ($p=0.106$) between the severity of microstomia and the type of prosthodontic treatment received in combination cases. A pattern is observed in combination cases: where the more missing teeth they have, the less likely they are to use conventional dentures.

4.3.5 Correlation between severity of microstomia with prosthodontics treatment in partially dentate patients.

In Table 4.3, the severity of microstomia for partially dentate patients is divided into two groups: Group I (0-24 mm), indicative of more severe conditions characterised by limited mouth opening, and Group II (25-40 mm), representing less severe conditions.

Within Group I, three patients received conventional dentures (Garg et al., 2011; Gözde Türk & Ulusoy, 2015; Tripathi et al., 2011), two received sectional dentures (Kam et al., 2006; Ravi et al., 2022), and two received flexible dentures (Samet et al., 2007; Singh et al., 2014). McKenna et al. (2012) described a case where they replaced teeth 11 and 21 using a resin-bonded bridge for a patient with a mouth opening of 19mm.

Group II shows a different distribution: two patients received conventional dentures, one received sectional dentures, one received a combination of sectional-conventional dentures, and one underwent an implant overdenture. Langer et al. (1992) used dental implants in the treatment of microstomia patients with 28mm MMO. He constructed maxillary implant-supported overdenture and mandibular implant-supported fixed prosthesis. Statistical analysis using the Fisher Exact Test yielded a p -value of 0.940, indicating no significant statistical correlation between the severity of microstomia and the type of prosthodontic treatment chosen.

4.4 Aetiology of microstomia, dentition status and impression techniques

Information on the aetiology of microstomia, evaluation of the patients dentition status and impression techniques are presented in table 4.4 and table 4.5.

Table 4.4 Aetiology of Microstomia and Dentition Status

Sociodemographic Characteristics	Frequency	
	n	%
Aetiology of Microstomia		
Surgery	14	17.1
Scleroderma	31	37.8
Burn	9	11.0
Submucous fibrosis	11	13.4
Radiotherapy	7	8.5
Developmental	6	7.3
Other	4	4.9
Dentition Status		
Edentulous	48	58.5
Partially Dentate	20	24.4
Combination (C/P or P/C)	14	17.1

4.4.1 Aetiology of Microstomia

In this systematic review, various causes of microstomia were observed (Table 4.4), with scleroderma being the most reported (37.8%). This was followed by surgical interventions (17.1%), submucous fibrosis (13.4%) in addition, radiotherapy (8.5%) was also identified as a potential cause of microstomia in few cases (refer to Appendix F).

Bilhan et al. (2011), Dewan et al. (2015), Kumar et al. (2012), Kumar et al. (2020), Prasad et al. (2008) and Satpathy and Gujjari (2015) reported the microstomia cases as developmental origin, excluding all other potential causes and the patient's medical and dental histories did not contribute to the final diagnosis.

Additional cases reported microstomia condition associated with illnesses such as epidermolysis bullosa (Farhang et al., 2011), necrotising fasciitis (Cheng et al., 2006), muscular dystrophy (Rathi et al., 2013), and rheumatoid arthritis (Ohkubo et al., 2003).

4.4.2 Dentition status

Edentulous cases were the most frequently (58.5%) reported, followed by partially dentate cases (24.4%) and cases with a combination of completely edentulous on one arch and partially dentate on the other arch (17.1%), as shown in Appendix G.

4.4.3 Impression technique

The impression technique cannot be categorized by the severity of microstomia because some reports did not provide detailed and consistent data regarding the methods used to take impressions. This lack of comprehensive data reporting makes it difficult to establish a clear correlation between the impression technique and the severity of microstomia. Table 4.5 outlines the various techniques reported for both primary and final impressions. Additionally, the term "combination" in this table refers to the utilization of both sectional and conventional impressions within a single patient.

Table 4.5 Impression techniques

Impression	Frequency	
	N	%
Preliminary		
Conventional	4	4.9
Sectional	20	24.4
Flexible	21	25.6
Shortened flange	9	11.0
Small tray	9	11.0
Digital	5	6.1
Combination*	2	2.4
Not mentioned	12	14.6
Total	82	100.0
Final		
Conventional	12	14.6
Sectioned	51	62.2
Combination*	6	7.3
Digital	3	3.7
Not mentioned	10	12.2
Total	82	100.0

*Referred to combination of sectional and conventional impression in a patient.

Table 4.5 shows that the most used techniques for preliminary impression were flexible impression or hand manipulation with putty impression material (25.6%). Using flexible impression material techniques allow for better manipulation and adaptation within the constrained space, ensuring that critical anatomical landmarks are captured without causing discomfort or trauma to the patient (Sebastián et al., 2023). For example, in patients with scleroderma-induced microstomia, the rigidity of the skin and mucosa further complicates using conventional trays, making flexible materials a more viable option (Ozatic et al., 2022). The use of sectional stock trays (24.4%), which can be assembled extra-orally after the impression was made is another innovative approach documented as a technique of choice. Less common but notable were modifications of stock tray by shortening the flange and the use of smallest tray or paediatric tray, both representing 11.0% respectively. 6.1% of authors preferred the use of CAD-CAM

technique for constructing individual custom trays for the patients (Jagielska et al., 2024; Saygılı et al., 2019; Silvestri et al., 2023; Zhang et al., 2020).

One significant issue with flexible impressions is the difficulty in achieving a dimensionally accurate impression due to the restricted oral aperture, which can lead to imprecise prosthesis fabrication and poor fit (Colvenkar, 2010; Kunusoth et al., 2022). Maria et al. (2022) emphasised the importance of forming precise custom trays and diagnostic casts to ensure final impression accuracy in microstomia patients. While flexible impressions alone may not suffice for final impressions, the integration of sectional custom trays allow for accurate impression taking by accommodating the limited mouth opening and ensuring that all critical anatomical landmarks are captured (Colvenkar, 2010).

For this reason, 62.2% of authors use sectional custom trays for final impressions, and 14.6% of the authors manage to use conventional custom trays, as in Appendix I. Digital CAD-CAM technology, though less commonly used (6.1% for preliminary impression and 3.7% for final impression), has shown promising results in creating accurate and well-fitting prostheses for these patients. For instance, intraoral scanning has been successfully used for preliminary impressions, and 3D printing has facilitated the creation of custom sectional trays and dentures, leading to high patient satisfaction and successful prosthetic outcomes (Jagielska et al., 2024; Saygılı et al., 2019; Zhang et al., 2021). However, the adoption of digital scanning is hindered by several factors. The cost and learning curve associated with digital technologies can be prohibitive. The need for specialized equipment, such as 3D printers and CAD/CAM software, along with the training required to use these tools effectively, can be a significant barrier for many dental practices (Zhang et al., 2020; Jagielska et al., 2024). An additional 12.2% of the authors either used the

primary impression as the final impression or did not mention any impression technique at all in their reports.

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CHAPTER 5: DISCUSSION

5.1 Critically reviewed case reports and cases series

5.1.1 Overview

Prosthetic treatment of patients with microstomia presents significant challenges due to the difficulty in accessing the oral cavity for dental procedures. Various innovative techniques and materials have been developed to address these challenges. One common approach involves using sectional custom trays for making impressions, which can be particularly effective in capturing the necessary anatomical details despite the limited oral aperture (Maria et al., 2022; Subba et al., 2022). Similarly, intraoral scanning has emerged as a viable alternative to traditional impression techniques, allowing for the creation of accurate models without the need for large trays that cannot fit into a restricted mouth opening (Ozatic et al., 2022; Moslemian & Hasanzade, 2023). In addition to impression techniques, the design of the prosthesis itself is crucial. Sectional, collapsible, and hinged dentures have been developed to facilitate easier insertion and removal, addressing the functional and aesthetic needs of patients with microstomia (Patil et al., 2019; Kumar et al., 2020; Srivastava et al., 2022). The use of intraoral scanning and 3D printing for creating removable partial dentures has also been effective in such cases, providing a more comfortable and precise fit for the patients (Ozatic et al., 2022). Overall, the success of prosthetic treatments in microstomia patients depends on the severity of the condition and the underlying cause. While traditional methods may fall short, the integrating digital technologies, innovative impression techniques, and customised prosthesis designs have shown promising results in improving the quality of life for these patients (Patil et al., 2019).

5.1.2 Method for the critical review of case reports and case series

5.1.2.1 Eligibility criteria

The inclusion criteria for this study were designed to comprehensively cover all relevant and available literature on microstomia. The study reviewed all case reports and case series published in English up to May 2024 to ensure that all pertinent cases were captured. Given the unique nature of microstomia, covering all published data without date restrictions aimed to maximize the collection of relevant information.

The study will focus on case reports of microstomia that provide detailed and clear assessments of vertical mouth opening. This specific criterion is crucial for understanding the extent of the condition. It also included cases that documented prosthodontic treatments administered to patients with microstomia, intending to gather comprehensive information on various treatment approaches and their outcomes, thus contributing to a deeper understanding of effective management strategies for this condition.

On the other hand, the exclusion criteria are set to maintain the study's focus and relevance. Any cases where limited mouth opening is attributed to temporomandibular joint (TMJ) problems will be excluded. This is because TMJ issues represent a different underlying cause for limited mouth opening, which may require different diagnostic and treatment approaches compared to microstomia. By excluding such cases, the study aims to ensure that the data collected is specifically relevant to microstomia, allowing for a more accurate and focused analysis of this condition and its prosthodontic treatment options. This careful delineation between inclusion and exclusion criteria helps to refine the study's scope and enhance the quality of the findings.

5.2 The most adopted approach to measure severity of microstomia

MMO is a critical parameter in diagnosing and managing microstomia, and is essential for evaluating the functional impairment and planning appropriate interventions, including prosthetic rehabilitation and surgical procedures (Sarandha et al., 2015; Sudhir & Kundankumar, 2017).

Most authors in this review use lip-to-lip to measure MMO for edentulous and combination edentulous-partially dentate patients. Clinicians often employ the lip-to-lip measurement method because it provides a straightforward assessment of the functional mouth opening, especially in cases where the incisal edges are not clearly defined or accessible due to dental or anatomical variations (Fatima et al., 2016; Kumar et al., 2020; Sudhir & Kundankumar, 2017). Additionally, lip-to-lip measurements does not require the insertion of any instruments into the mouth, which can be difficult and uncomfortable for patients with limited oral openings (Patel et al., 2013). It is also more accessible to perform in a clinical setting, making it a practical choice for routine assessments (Sudhir & Kundankumar, 2017).

In cases of partially dentate, the interincisal technique is indeed the most preferred method for measuring MMO. This method is particularly useful as it provides a consistent and reliable measurement, which is crucial for accurate clinical assessment and treatment planning.

The various methods for measuring MMO explored in this review most likely because researchers' attempts to accurately capture the maximal extent of mouth opening that patients can achieve at various angles. Despite the diverse approaches to measuring MMO, the key objective is to ensure that prosthetic devices can accommodate maximum mouth opening effectively, without causing harm to the patient. Therefore, although the specific measurement points may differ, the ultimate aim in all studies remains consistent:

to promote the optimal fit and function of prosthetic treatments for individuals with microstomia.

5.3 Correlation between severity of microstomia with prosthodontics treatment

From the original set of 82 case reports, fifteen case reports were excluded at this stage because they did not provide any follow-up information for the patients. A quality case report should clearly describe the clinical condition after the intervention, detailing whether symptoms are present or absent. Presenting the management outcomes helps convey crucial information to clinicians (Moola et al., 2020). The presence of follow-up in many cases underscores the importance of post-treatment evaluations in dental prosthetics. The follow-up periods varied widely, with the shortest being 1 week (Tayari et al., 2019) and the longest spanning 10 years (Garcés Villalá & Zorrilla Albert, 2021). Most cases showed that patients adapted well to the prosthesis without any significant complications. For instance, Mostafavi and Hajimahmoudi (2017) noted a minor ulcer on the patient's mandibular mucosal ridge during follow-up. A 'g' relief was incorporated into the prosthesis for that area, and the patient reported no other ulcerations in subsequent follow-ups.

After exclusions, 67 case reports remain for correlation analysis. The descriptive analysis of all 67 included studies indicated that there was no significant correlation found between the severity of microstomia and the choice of prosthodontic treatment approaches. This suggests that the severity of microstomia does not strongly influence the selection of specific prosthodontic treatments across the studies analysed. This finding could imply that various prosthodontic options may be considered regardless of the degree of microstomia, with treatment decisions influenced more by individual patient factors and clinical considerations rather than the severity of microstomia alone.

Srivastava et al. (2022) and Kumar et al. (2020) highlighted the significance of patient's manual dexterity as one of the factors that contribute the prosthetic treatment options, as reduced dexterity can complicate the insertion and removal of the prosthesis, necessitating designs that are easier to handle, such as collapsible or hinged dentures. Cost considerations also play a significant role, as advanced techniques like CAD/CAM and custom-fabricated sectional trays can be more expensive than conventional methods. However, the long-term benefits in terms of improved function, aesthetics, and quality of life often justify the higher initial investment (Patil et al., 2019). The prosthodontic management of microstomia patients requires a tailored approach that considers all these factors to achieve the best possible outcome in terms of aesthetics, function, and patient satisfaction (Tayari et al., 2019).

The analysis examining the relationship between microstomia severity and prosthodontic treatment approaches, however, revealed an interesting trend regarding the selection of dental prosthetics. It was observed that conventional dentures are often avoided when mouth opening is below a certain limit, suggesting that anatomical limitations affect treatment decisions. This trend highlights the need for flexibility in managing microstomia, with options like flexible dentures or using mini implants when access to the mouth is limited.

5.3.1 Recommended prosthodontic treatments based on the severity of microstomia

From the findings in Table 4.3, a table outlining the relationship between prosthodontic treatments and the severity of microstomia is suggested.

Table 5.1 Proposed prosthodontic treatments according to severity of microstomia

Dentition Status	Group	MMO	Prosthodontic Treatment Options
Edentulous	I	0 - 19	<ul style="list-style-type: none"> • Sectional denture • Sectional denture opposing conventional denture
	II	20 - 40	<ul style="list-style-type: none"> • All prosthodontics treatment with modification
Combination	I	0 - 24	<ul style="list-style-type: none"> • Sectional denture • Flexible denture • Implant-supported prosthesis (sectional overdenture or fixed prosthesis)
	II	25 - 40	<ul style="list-style-type: none"> • All prosthodontics treatment with modification
Partially Dentate	I	0 - 24	<ul style="list-style-type: none"> • All prosthodontics treatment with modification
	II	25 - 40	<ul style="list-style-type: none"> • Depending on number of missing teeth • The greater the number of missing teeth, the more modifications are required.

Table 5.1 proposed a classification system for prosthodontic treatment options based on the patient's dentition status (edentulous, partially dentate, and combination), and the severity of their limited mouth opening (measured in millimetres).

5.4 Implementing Bias Reduction Strategies

To minimise bias in this systematic review, a series of systematic strategies were implemented. Clear and consistent inclusion and exclusion criteria were established to assess relevant studies, reducing selection bias.

To ensure thoroughness, a comprehensive search strategy was executed across multiple databases using relevant keywords and Medical Subject Headings (MeSH) terms related to microstomia and prosthodontics. A standardised data extraction form was created to maintain consistency, with independent data extraction performed by two reviewers to enhance accuracy, resolving discrepancies through discussion to minimise errors and subjective bias.

Adherence to established reporting guidelines, such as PRISMA (Preferred Reporting Items for Systematic Reviews and Meta-Analyses), ensured transparent reporting of the methodology, including search strategy, selection criteria, and data extraction processes for reproducibility. The Joanna Briggs Institute (JBI) Critical Appraisal Tools were employed to evaluate the quality and reliability of the studies, focusing on bias risk in various designs.

Through these meticulous steps, bias was minimised, significantly enhancing the reliability and validity of the systematic review on prosthodontic treatments for microstomia patients, providing trustworthy insights for clinical practice.

5.5 Limitations

This review was limited by inconsistencies in data across case reports and case series. The heterogeneity among the included studies, variability in mouth opening measurement techniques, and the absence of high-quality evidence, such as randomized controlled

trials, constrains the obtainment of comprehensive findings. The reliance on case reports and case series may introduce bias and limit the ability to draw definitive conclusions.

5.6 Recommendations for future research

While current research provides important information on the prosthodontic management of microstomia, there is a significant need for greater consistency and standardization in reporting more details of the cases such as mouth opening measurement techniques and impressions techniques used. To enhance the quality and reliability of future research, it is crucial to implement stricter review processes for case reports and develop standardized measurement and management protocols. Standardized measurement protocols and detailed reporting of impression techniques and treatment outcomes are essential for advancing the field. Addressing these issues will enable future studies to offer more definitive guidance and improve both the functional and aesthetic outcomes for patients with microstomia.

CHAPTER 6: CONCLUSION

1. In conclusion, this study addresses the objectives outlined regarding the prosthodontic management of patients with microstomia. It provides a rigorous analysis of published case reports and series, revealing a wide array of treatment approaches while highlighting the diverse aetiologies of microstomia, with scleroderma being the most frequently documented cause.
2. The lip-to-lip technique was the most adopted method for measuring maximal mouth opening (MMO) in edentulous and combination cases. The interincisal technique remains the preferred choice for partially dentate patients.
3. There was no significant correlation between the severity of microstomia and the type of prosthodontic treatment provided.

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