

Temporal Bone Endometriosis — A Multidisciplinary Approach. A Clinical Case

Nazarii Brotskyi^{1*}, Tetiana Tatarchuk², Kateryna Plaksiieva³,
Alexander Fetsych⁴, Volodymyr Ostrianko⁵

1. Private practice, Kyiv, Ukraine.
2. Professor, Senior Scientist, Medical Center "Verum"; Institute of Pediatrics, Obstetrics and Gynecology of the National Academy of Medical Sciences of Ukraine; State Scientific Institution "Center for Future Technologies of the National Academy of Sciences of Ukraine", Kyiv, Ukraine.
3. PhD Student, Medical Center "Verum"; Institute of Pediatrics, Obstetrics and Gynecology of the National Academy of Medical Sciences of Ukraine; State Scientific Institution "Center for Future Technologies of the National Academy of Sciences of Ukraine"; Educational and Scientific Center "Institute of Biology and Medicine", Kyiv, Ukraine.
4. PhD Student, Department of Therapeutic Dentistry FPGE, Danylo Halatsky Lviv National Medical University, Lviv, Ukraine.
5. Associate Professor, Department of Orthodontics, Shupyk National Healthcare University of Ukraine, Kyiv, Ukraine.

Abstract

At present, not a single case of endometriosis of the temporal bone has been described in medical science, as well as the association of endometriosis with dysfunction of the temporomandibular joint and the development of degenerative joint disease, as well as the treatment of these diseases with gestagens.

This study presents a patient with primary complaints of constant acute pain in the left TMJ (temporomandibular joint) aggravated by any movements of the lower jaw, inability to open the mouth wide, and persistent headaches. The first-listed diagnosis was TMJ dysfunction and degenerative joint disease, and the final diagnosis was temporal bone endometriosis and secondary joint dysfunction. An articular splint helped alleviate initial symptoms, and the final relief came after placing on progestogen therapy. Joint dysfunction has a polyetiological nature and requires an integrated approach in diagnosis and treatment to achieve a clinical result.

Case Report (J Int Dent Med Res 2022; 15(3): 1305-1310)

Keywords: Invasive endometriosis, endometrial implants, TMJ, TMD.

Received date: 17 May 2022

Accept date: 03 July 2022

Introduction

Even though the etiology of various types of temporomandibular joint dysfunction is still debatable, certain generally accepted factors are identified in the dental community non the less. Chief among these factors has always been malocclusion (incorrect interdental contacts between teeth in static or dynamic), which led to a CO-CR discrepancy (that is, the mismatch of the jaw's position in the usual bite and in the musculoskeletal stable ratio, in which the articular condyles are in the correct position in the articular fossae, and the maxillofacial muscles are in the correct uniform tone), causing the lower jaw to move into a forced position and producing the corresponding symptoms. Many

other causes are also identified, such as neurological disorders, postural disorders, traumatic lesions, etc. However, only rare sources mention that dysfunctions often arise from dyshormonal conditions¹.

Only a few studies have mentioned that fibrous cartilage, covering articular surfaces, has estrogen receptors, responsible for the normal conduction of synovial fluid through cartilage and, consequently, its normal trophism^{2,3}.

Therefore, considering the joints from a non-dental side is uncommon and is thought unprovable in the dental world. However, as practice shows, approximately 85% of dysfunctions are of a polyetiological nature, quite often not related to dentistry at all.

The presented case illustrates a rare joint pathology mainly caused (despite real dental etiological factors) by endometriosis of the temporal bone (namely, the articular tubercle). The case is truly unique since no such condition has yet been described.

*Corresponding author:

Nazarii Brotskyi,
Private practice, Kyiv, Ukraine.
E-mail: opdihatopa@gmail.com

Materials and methods

Patient Information

Patient E. sought medical assistance at a dental clinic with complaints of constant acute pain in the left TMJ (temporomandibular joint), which intensified with any movement of the mandible, the inability to open her mouth wide, and constant headaches. Complaints began four years ago when the patient first felt a crunch and discomfort in the joint, which increased over time. The patient could not identify a possible cause. As a variant of the reason, she named wearing a bracket system in 2010, but an asymptomatic period of 5 years did not allow her to hold that. The patient underwent several courses of splint therapy with different doctors who tried to rehabilitate her in the concept of Rudolf Slavicek by anterization (shifting the jaw to the anterior therapeutic position), which, however, did not bring even temporary relief.



Figure 1. a) Frontal facial photos; b). Side facial photos; c) Intraoral photos.

According to the patient, she was somatically healthy, with negative family anamnesis.

Clinical Findings

Objective examination by a dentist revealed:

Horizontal type of the skull bone growth
 2nd class, 1st subclass according to

Engle

- VDO reduction
- Chin shift to the left
- Micrognathia of the lower jaw
- Counterclockwise transversal rotation of the upper jaw
- Extrusion of 37 and 47 teeth
- Front teeth protrusion of both jaws
- No interdental contact in the frontal area

Enlargement of the nasolabial angle (see Figure 1, Figure 2)

Opening the mouth by 28 mm with deflection to the left

Palpation: pain in the temporomandibular joints in 8 zones according to Mariano Rocabado; bilateral pain in masticatory, temporal, medial pterygoid, lateral pterygoid, mandibulohyoid, digastric muscles, more significant on the left; pain in the left trapezius and sternocleidomastoid muscles.

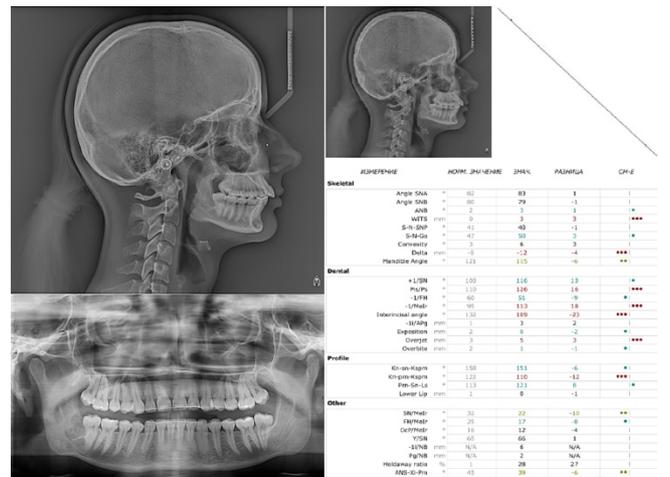


Figure 2. Orthopantomography, teleradiography, cephalometry.

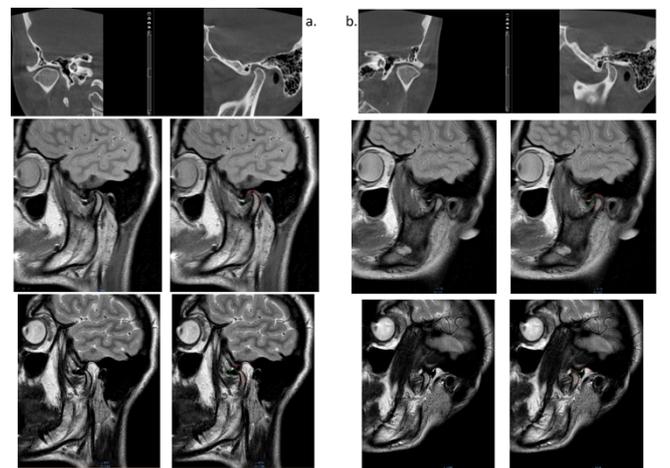


Figure 3. Computed tomography and magnetic resonance imaging of the TMJ. a. Right TMJ. b. Left TMJ.

Diagnostic Assessment

CBCT of the temporomandibular joint with a closed mouth revealed a posteroinferior position of the right condyle, degenerative joint disease of the right joint in the form of regressive remodelling, posteroinferior position of the left

condyle, degenerative joint disease of the left joint in the form of progressive remodelling (formation of exostosis on the articulating surface of the condyle in the form of a spike).

MRI of the TMJ without contrast showed an incomplete anteromedial dislocation of the articular disc of the right TMJ with reduction, synovitis of the right TMJ; complete anteromedial dislocation of the left TMJ articular disc without reduction and block, synovitis of the left TMJ (see Figure 3).

Therapeutic Intervention

The patient was prescribed a combined treatment:

1. Registration the “bite do jour” position using the front Okeson deprogrammer (see Figure 4).

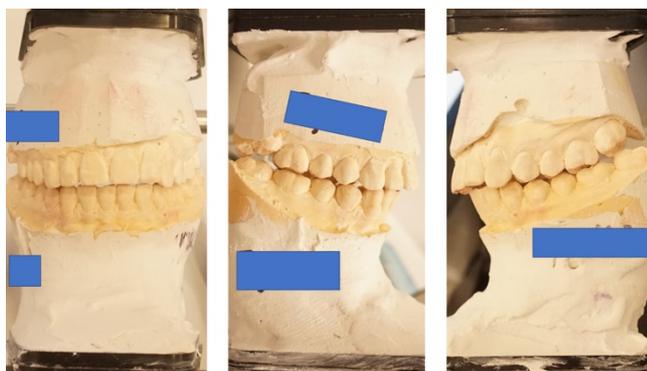


Figure 4. Centric do jour.

2. Fabrication of a three-section myorelaxant splint for the upper jaw to be worn according to the standard scheme (posterior segment — around the clock, anterior segment — during sleep). The period of wearing the splint was not determined since it was necessary not only to remove the symptoms but also to remodel the articular heads to a stable form.

3. Drug therapy: Arcoxia (etoricoxib) 60 mg, 2 times a day for 14 days; Omez (omeprazole) 20 mg, 2 times a day for 14 days; Myorix (cyclobenzaprine hydrochloride) 15 mg, 2 times a day for 7 days.

Significant relief was observed on the 3rd day after the splint therapy started, and on the 14th day, the symptoms disappeared completely. By grinding the splint's occlusal contacts, total muscle relaxation and asymptomaticity of the stomatognathic system were achieved.

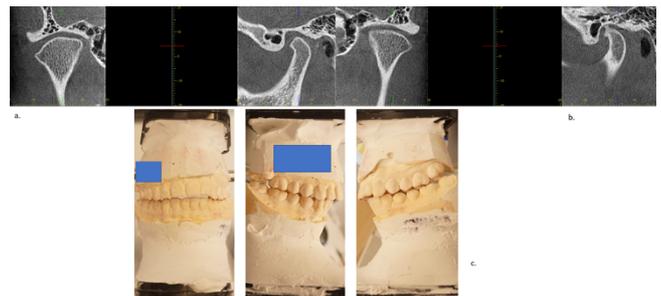


Figure 5. The situation on the 5th month of splint therapy. a. Right TMJ. b. Left TMJ. c. Gypsum coating.

However, on the fifth month of treatment (see Figure 5), the patient complained of pain along the back of the neck, so she was sent to a course of therapeutic massage and exercise therapy to eliminate it. The patient felt significant relief, but a small part of the symptoms remained, so she arbitrarily decided to go to a chiropractor. According to the patient, her cervical and lumbar vertebrae were manually reset, which led to acute pain and the inability to maintain postural balance. Pain in the TMJ and muscles returned to the initial level.

It was decided to send the patient for a consultation with a neurosurgeon, who prescribed her drug therapy (dynostat (parecoxib), mydocalm (Tolperisone), veroshpiron (spironolactone)). The therapy brought little relief but caused acute allergic dermatitis, so it was prematurely terminated.

All this time, the patient was morally depressed and reacted aggressively to any treatment.

Given the recurrence of pain and the ineffectiveness of this type of splint therapy, it was decided to transfer the patient to a full myorelaxant splint on the lower jaw with incisor and canine guidances on the 8th month of treatment (see Figure 6).

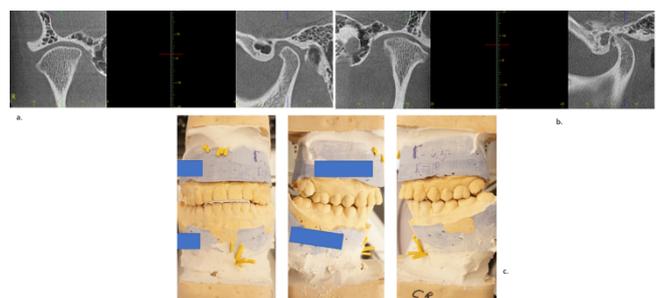


Figure 6. The situation on the 8th month of splint therapy. a. Right TMJ. b. Left TMJ. c. Gypsum coating.

Wearing a new splint relieved the patient's symptoms on the seventh day, but mild pain persisted and periodically increased substantively for several days.

After 2.5 months of wearing the new splint, the patient began to complain of sudden sharp pain in the lower abdomen and was taken to the admission department with a preliminary diagnosis of acute appendicitis. In the admission department, this diagnosis was ruled out and the patient was hospitalized in the gynaecological department. It turned out that the patient was suffering from painful menstruation since menarche, which was corrected by NSAIDs. According to the anamnesis, the patient was last examined by a gynaecologist 1.5 years ago, when she was diagnosed with algodysmenorrhea and prescribed Yaryna (a combined estrogen-progestogen contraceptive — Ethinyl estradiol 0.03 and drospirenone 3 mg).

In the gynaecological hospital, the pelvic ultrasound detected endometrioid cysts of both ovaries, approximately 3.0 cm in diameter, for the first time. After correction of the pain syndrome, the patient was discharged and prescribed Regulon (a combined estrogen-progestogen contraceptive — desogestrel 0.15 mg and Ethinyl estradiol 0.03 mg) for up to 6 months.

The patient took Regulon for two months, but the clinical picture had not changed. It was decided to send the patient to the second consultation with a gynaecologist, who stopped taking Regulon and transferred her to Dienogest 2 mg.

Despite the expected worsening of articular symptoms, which is a common side effect of taking progestogens, the pain syndrome completely disappeared. Normalization of TMJ function was obtained on the 14th month of treatment (see Figure 7).

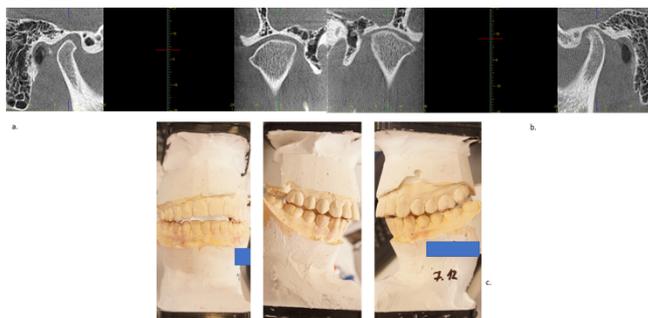


Figure 7. The situation on the 14th month of splint therapy. a. Right TMJ. b. Left TMJ. c. Gypsum coating.

Due to this phenomenon, a retrospective study of the time of the patient's pain symptoms intensification was conducted, which revealed that periods of exacerbation during the treatment most often occurred on the 3rd-6th days of the menstrual cycle. Also, control TMJ CT scans performed during splint therapy revealed a cavity in the articular tubercle, which was empty at the beginning and had a blackout inside upon the CBCT scan made in the first days of the menstrual cycle (see Figure 8).

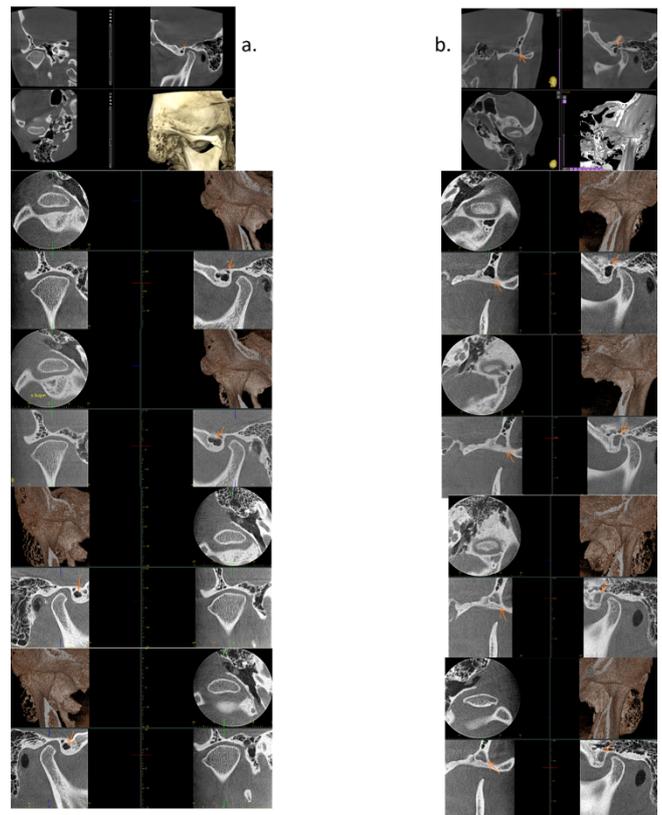


Figure 8. The TMJ CT dynamics. The arrow indicates the area of interest. a. Right TMJ. b. Left TMJ.

For clinical verification of the diagnosis, it was decided to make a control MRI of the TMJ without contrast on the 3rd day of the menstrual cycle, which revealed a cavity in the articular tubercle of the left TMJ filled with a large amount of fluid (see Figure 9).

Clinical data, the cyclicity of exacerbations associated with the menstrual cycle, data from additional research methods and, most importantly, the response to Dienogest therapy let us conclude that extragenital endometriosis was the cause of articular dysfunction.

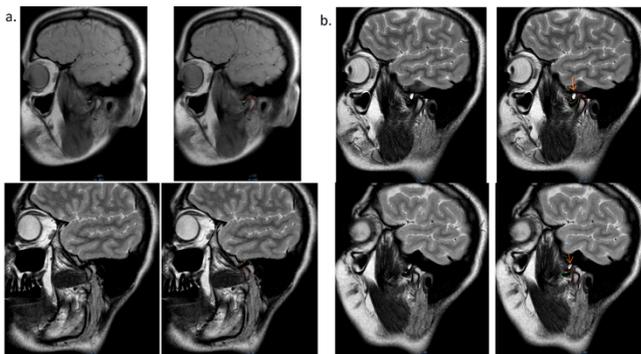


Figure 9. The MRI of the TMJ at the end of splint therapy. The arrow indicates the area of interest. a. Right TMJ. b. Left TMJ.

Results

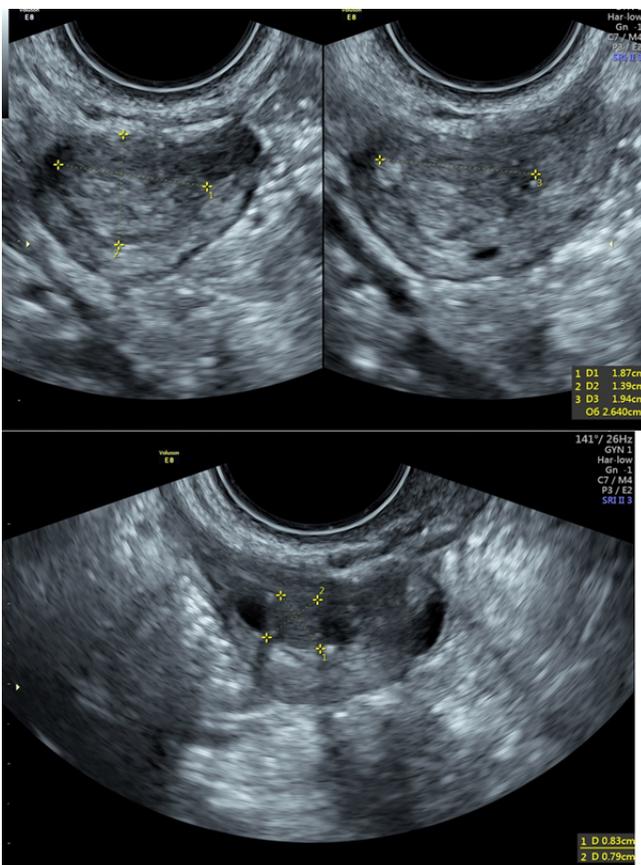


Figure 10. Ultrasound imaging of ovaries with endometrioid cysts. Typical appearance: rounded hypoechoic unilocular masses with hypoechoic 'ground glass' content.

After 6 months of taking Dienogest, menstruation became less painful, but there was no amenorrhea induced by this drug. Despite the expected worsening of articular symptoms, which is a common side effect of taking progestogens, complete disappearance of pain and involution of

the formation on the left articular head were achieved within three months.

According to the pelvic ultrasound, there is a decrease in the size of ovarian cysts from the initial level (approximately 30.0 mm in diameter) at the time of the follow-up examination: 19.0 x 14.0 x 19.0 mm in the right ovary and 8.0 x 8.0 mm. in the left one (see Figure 10).

Considering the insufficient effectiveness of Dienogest in the management of pelvic pain and the lack of reproductive plans of the patient in the nearest future (she planned pregnancy in 1-2 years), it was decided to prolong the use of Dienogest until the start of pregnancy planning, and also to administer Goserelin 10.8 mg s/c.

Discussion

Endometriosis is a condition in which the endometrium (inner layer of the uterus) is present in organs other than the uterus itself. According to the latest systematic review, the incidence of endometriosis in the population ranges from 0.8 to 28.6% with an overall estimate of 4.4%. When analyzing distinct populations, endometriosis was found in 33.5% of women undergoing surgery for benign gynaecological conditions, in 23.8% of infertile women, and in 49.7% of women with chronic pelvic pain^{4,5}.

Endometriosis is an estrogen-dependent inflammatory disease that causes pelvic pain and uterine bleeding and often leads to infertility⁶.

Endometriosis usually occurs in the myometrium (also called adenomyosis), ovaries (endometrioid cysts), ligamentous apparatus of the uterus and on the surface of the peritoneum, less often in the intestines, bladder, ureter, abdominal wall, chest cavity, and other organs⁷.

We should note that statistical data on the frequency of this disease relates to pelvic endometriosis, and there are no statistical data on extragenital endometriosis, given the rarity of this pathology, the diversity and unpredictability of localizations, and the frequent mimicry of other pathological conditions⁸.

Very often, the only symptom that can help in the formation of the diagnosis is the cyclicity of clinical manifestations (associated with both the pathophysiology of endometriosis and the affected organ)⁹. So, for example, with endometriosis of the chest, catamenial hemothorax or hemoptysis occurs. Recurrent pneumonia is also possible. With endometriosis

of the navel, the formation begins to increase and hurt before and during menstruation¹⁰.

That is why multidisciplinary collaboration is so substantial in the diagnosis and management of this condition.

The only guideline regarding extragenital endometriosis published in Japan in 2020 concludes that medical therapy is effective in the treatment of rectosigmoid endometriosis, bladder endometriosis, catamenial haemoptysis (when symptoms are mild or moderate); surgical treatment: endometriosis of the intestine or ureter with hydronephrosis, endometriosis of the navel (however, medication can also be considered)¹¹.

At the moment, the treatment of endometriosis lies in the suppression of ovulation to create a hypoestrogenic state and decidualization of endometrioid heterotopias, which can be achieved by continuous use of progestogens and, in severe cases, GnRH agonists¹².

In this case, as in most cases of extragenital endometriosis that we observed, it was the cyclical nature of the clinical manifestations of TMJ dysfunction that was detected retrospectively, the simultaneous presence of genital endometriosis, and the effectiveness of Dienogest, intended for its treatment, in reducing TMJ pain, prompted thought about endometriosis of the temporal bone¹³.

This clinical case represents a unique variety of extragenital endometriosis — endometriosis of the temporal bone. This once again reminds us of the "omnipresence" of this condition and the need to include a range of gynaecological diseases in the differential diagnosis of maxillofacial pathology.

Conclusions

Temporomandibular joint dysfunction is a complex polyetiological disease, often associated with non-dental causes. Therefore, a systematic assessment of the patient's body is a critical part of the examination, which allows establishing the correct diagnosis and choosing the right treatment tactics.

At the same time, extragenital endometriosis can theoretically affect any organ system, causing characteristic, often non-specific symptoms. Therefore, understanding the specifics of the pathology of all body systems is

important for a gynaecologist in the diagnosis and treatment of extragenital endometriosis.

Declaration of Interest

The authors report no conflict of interest.

References

1. Robinson JL, Soria P, Xu M, et al. Estrogen Promotes Mandibular Condylar Fibrocartilage Chondrogenesis and Inhibits Degeneration via Estrogen Receptor Alpha in Female Mice. *Sci Rep*, 2018; 8: 8527.
2. Puri J, Hutchins B, Bellinger LL, Kramer PR. Estrogen and inflammation modulate estrogen receptor alpha expression in specific tissues of the temporomandibular joint. *Reprod Biol Endocrinol*, 2009; 7: 155.
3. Abubaker AO, Raslan WF, Sotereanos GC. Estrogen and progesterone receptors in temporomandibular joint discs of symptomatic and asymptomatic persons: a preliminary study. *J Oral Maxillofac Surg*, 1993; 51(10): 1096-100.
4. Parazzini F, Roncella E, Cipriani S, et al. The Frequency of Endometriosis in the General and Selected Populations: A Systematic Review. *J Endometr Pelvic Pain Disord*, 2020; 12(3-4): 176-189.
5. Rogers PA. Priorities for endometriosis research: recommendations from an international consensus workshop. *Reprod Sci*, 2009; 16: 335-466.
6. Hudson N. The missed disease? Endometriosis as an example of 'undone science'. *Reprod Biomed Soc Online*, 2021; 14: 20-27.
7. Davis AC, Goldberg JM. Extrapelvic Endometriosis. *Semin. Reprod Med*, 2017; 35: 98-101.
8. Andres MP, Arcoverde FVL, Souza CCC, Fernandes LFC, Abrão MS, Kho RM. Extrapelvic Endometriosis: A Systematic Review. *J Minim Invasive Gynecol*, 2020; 27(2): 373-389.
9. Mederos MA, Villafaña N, Dhingra S, et al. Pancreatic endometrial cyst mimics mucinous cystic neoplasm of the pancreas. *World J Gastroenterol*, 2017; 23(6): 1113-1118.
10. Sharma A, Apostol R. Cutaneous Endometriosis. In: *StatPearls*. Treasure Island (FL): StatPearls Publishing; 2021.
11. Hirata T, Koga K, Kai K, et al. Clinical practice guidelines for the treatment of extragenital endometriosis in Japan, 2018. *The journal of obstetrics and gynaecology research*, 2020; 46(12): 2474-2487.
12. Ministry of Health. *Diagnosis and Management of Endometriosis in New Zealand*. Wellington: Ministry of Health; 2020.
13. Murji A, Biberoğlu K, Leng J, et al. (2020) Use of dienogest in endometriosis: a narrative literature review and expert commentary. *Current Medical Research and Opinion*, 2020; 36(5): 895-907.