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RESEARCH ARTICLE

WRITER'S CRAMP - A MAJOR CONUNDRUM: REVIEW ARTICLE

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Abstract

Background: Writer's cramp (WC) is commonly seen in professional workers where writing is the main part of their work. It shows bimodal age of distribution, affects both genders.WC has an incidence rate of approximately 2.7 per million population.WC is diagnosed basically on clinical presentation and history.Functional improvement is the main aim of treatment in the case of WC.Treatment strategies consist of Pharmacological, Surgical, and Non-pharmacological strategies.

Objectives: This article highlights clinical features, epidemiology, etiology, available treatment and cure rate with respect to available treatment strategies, and the impact of the writer's cramp on the social, economic, and psychological behavior of patients.

Methods: This literature has been compiled after scrutinized analysis and consultation with various neurologists along with thorough research from trusted and verified sources.

Results: It is observed that patients with writer's cramps start using their non-dominant hands to get relief from dystonia. In severe cases, dystonia occurs in non-dominant hands also after some time. Patients with writer's cramps experience difficulty with day-to-day activities like writing, combing hair, and sewing.

Conclusion: Very few recent studies are available for writer's cramps and the sample size is also small. Treat Writer's cramp patient as handicapped, require a large sample size, case-control study, long-duration follow up period.

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Introduction:-

Disease is a medical condition affecting human function or structure negatively. An impairment is defined as "any loss or abnormality of a psychological, physiological or anatomical structure or function. Impairment can be temporary or it can be permanent. It may be visible, invisible, regressive, or progressive. Because of impairment, the affected person may be unable to carry out certain activities considered normal for his age, sex etc. This inability to carry out certain activities is termed "disability".” Handicap is defined as "a disadvantage for a given individual, resulting from an impairment or a disability, that limits or prevents the fulfillment of a role that is normal (depending on age, gender, and social and cultural factors) for that individual". For example, an accident is a disease, which causes loss of lower extremities means impairment. Due to loss of lower extremities person is not

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able to walk causing disability. As the affected person is unable to walk, he loses his job and become unemployed, and lives the rest of his life as a handicapped person.[1]

But the condition of our topic i.e. writer's cramp doesn't follow the above-mentioned pattern. Dystonia can be defined as patterned involuntary repeated muscle contractions mostly causing abnormal posture. It can be classified as primary and secondary, according to distribution it is classified as focal, multifocal, and generalized, according to the age of onset, it can be childhood or adult.

In primary dystonia, gene mutations involved are DYT1-DYT16. Childhood dystonia shows an autosomal dominant pattern of inheritance which commonly involves DYT1 with onset at approximately 14 years of age. Childhood dystonia is seen commonly in Ashkenazi Jewish people. It starts from the arms or feet and gradually progresses to other extremities. Postural deformities can be seen in severe cases. The severe form is common in hereditary cases. Dopa responsive dystonia (DRD) commonly affects those below 12 years of age and causes foot dystonia, resulting in walking difficulties. Patients with DRD show diurnal variations. Gait worsens during the day and it improves during rest at night hours. Mutation for DYT6 is observed in Amish families. DYT6 dystonia starts in cervical, and brachial muscles. As the disease progresses it becomes generalized and causes speech impairment. DYT11 shows dystonia in combination with psychiatric disturbances[2].

The common form of dystonia is focal dystonia. It affects females predominantly between the age of 40-and 60 years. Blepharospasm, Cervical dystonia, Oromandibular dystonia, Limb dystonia, and Spasmodic dysphonia are the main types of focal dystonia. Limb dystonia involves upper or lower extremities. It can be a writer's cramp or a musician's cramp. Approximately 30% of people have other body parts involved with extremities in focal dystonia.

Secondary dystonia occurs due to ingestion of some drugs or due to some other disorders [2]. It can be seen in Wilson's disease, brain tumor, stroke, toxins, Hartnup disease, Huntington's disease, multiple sclerosis, spinal cord injury, antidepressants, psychotropic drugs, and neuroleptic drugs. If the cause of secondary dystonia is treated then secondary dystonia is also cured [2].

In 1713,Ramazzini, the father of occupational medicine wrote about consistent symptoms which are related to the writer's cramp [3]. In 1864, Solly gave the terminology "scrivener's palsy" [4]. "Dystonia" term got widely accepted after 1911 till that period it was known as the disease "Oppenheim- Ziehen"[5]. Wimmer entitled dystonia as a syndrome in 1929 [6].

In most cases, Limb dystonia starts between 34-and 42 years of age with women predominance. Writer's cramp is the most common type of limb dystonia or task-specific dystonia[7]. The pathophysiology of the writer's cramps is unclear [7].But it can be due to repeated work, maybe due to genetic factors,environmental factors, defects in basal ganglia, dopamine increase, or due to contraction of muscles constantly [8].In the writer's cramp initial characteristic seen is an abnormal contraction of the muscle. Physiological study of writer's cramp shows reciprocal, surrounding, intracortical, interhemispheric inhibition, increased sensorimotor plasticity, cortical excitability becomes abnormal which causes excessive muscle contraction resulting in loss of ability for motor control [9].

Various epidemiological studies show that the frequency of focal dystonias is between 5% - and 19 % while the prevalence is 1.4/1,00,000 population. Writer's cramp (WC) is commonly seen in professional workers where writing is the main part of their work such as clerks, teachers, etc. In most cases, it starts at around 30-40 years of age. At starting phase of WC, the person feels heaviness in the fingers or forearm during writing which affects writing fluency. Even though pain is not a common symptom in WC, the patient holds the pen abnormally and forcefully due to dystonic contractions of the forearm and hand muscles. In WC, different patterns are observed, most common is index finger and thumb excessive flexion, wrist flexion, and hand at supination position. Approximately 50% of patients also experienced upper extremity tremors. Mostly it affects the dominant hand [10].

At beginning of the illness, the patient can write short stretches till dystonia or cramp appear. To overcome this situation, patients start to shorten their writing time. Many complain about the difficulty to hold a pen during writing or impaired script or handwriting. In some cases, it is observed that patients also experienced difficulty doing other tasks like brushing, combing, and knife handling. To overcome this problem much patient change their way of pen handling or starts to use a pen with a grip. Some even start using the opposite hand. Around 5% who started using unaffected contralateral arm develop dystonic features [10].

WC is diagnosed basically on clinical presentation and history. Electromyography (EMG), Nerve conduction for forearm muscle can be useful. It shows the crescendo phenomenon. Other investigation methods used are transcranial magnetic stimulation (TMS), computed tomography(CT), and magnetic resonance imaging (MRI). For functional studies, fluorodeoxyglucose positron emission tomography (FDG-PET) is also done in case of conflicting results. DYT1 gene mutation testing is also useful in DYT1 gene mutation suspected cases [10].

Functional improvement is the main aim of treatment in the case of WC. Abnormal posture correction and relief from discomfort are also important in WC management. Some patients start using the non-dominant hand, some started using a pen with a hard grip. Some patients try acupuncture, massage therapy, biofeedback, hypnosis, herbal therapy, or homeopathy. Though these treatment modalities are effective in the short run, the symptoms of writer cramps return eventually. For the treatment of writer's cramp pharmacological, surgical intervention is also used. Pharmacological treatment includes anticholinergics, dopamine agonists, benzodiazepines, baclofen, and dopamine antagonists. Botulinum toxin in injection form is the treatment of choice in case of writer's cramp. It is safer compared to other oral drugs. But for the botulinum toxin injection, the correct muscle needs to be chosen. It can be given with non-pharmacological treatments or with oral drugs. Surgical treatment for writer's cramp includes deep brain stimulation (DBS), repeated transcranial magnetic stimulation (rTMS), and thalamotomy [9].

In dystonia patients, psychiatric disorders are highly observed which affect the quality of life. Many show features of anxiety disorders, depression, and social phobia. Patients with writer's cramp start using the non-dominant hand, a special pen. In severe cases, due to hand abnormal posture, they could not perform writing or combing hair. It causes anxiety, depression, or social phobia in writer's cramp patients[11].

Aim & Objective:-

This Review article highlights clinical features, epidemiology, etiology, available treatment and cure rate with respect to available treatment strategies, and the impact of the writer's cramp on the social, economic, and psychological behavior of patients.

Methodology:-

This literature has been compiled after scrutinized analysis and consultation with various neurologists along with thorough research from trusted and verified sources.

Discussion:-

Dystonia is a hyperkinetic movement disorder resulting in abnormal posture due to the involuntary contraction of a certain group of muscles. It can be divided into primary dystonia and secondary dystonia. Primary dystonia is mostly genetic whereas the main clinical feature is dystonia. It can be young-onset or adult-onset while secondary dystonia is due to drug-induced, brain trauma-related. It is further classified as focal, multifocal, and generalized dystonia. Focal dystonia is further classified into Blepharospasm, Cervical dystonia, Oromandibular dystonia, Limb dystonia, and Spasmodic dysphonia [12].

It is seen that WC has bimodal age distribution at onset from 20 to 45 years while peak incidence mostly occurs at adult age and affects both genders [13-14,15,16]. It commonly involves the dominant hand [17]. It is found that there is a strong association between WC and time spent on writing, myopia, and head trauma [16]. Approximately 5 to 20% of patients have a family history of WC [18,19]. It is found that two variants rs11655081and rs61999318 (p. I493T) of the arylsulfatase G gene (ARSG) have an association with Writer's cramp [20,21]. DYT1 is a rare cause of WC [22]. In some cases, it is observed that even emotional stress causes Writer's cramps[23]. Environmental factors also contribute to developing WC. Other risk factors include anxiety, perfectionism like personality traits and joint mobility, hand size, and drugs such as amitriptyline, imipramine, and paroxetine [24-30]. Secondarily it is found in associated with spinal muscular atrophy, Parkinson's disease, and multiple sclerosis [13]. Some occupational neurologists suggest diabetes could be one of the possible causesof Writer's cramp [31]. WC is seen in professions like typists, hairdressers, tailors, pistol shooters, etc. who repeatedly do movement [32-34].

The writer's cramp is idiopathic, involuntarily muscle contraction and spasm of fingers of upper extremities during writing. It can be accompanied by pain or may be painless. It is classified as simple, progressive, and lastly dystonic. In the simple type, the patient experiences difficulty during writing. In addition to symptoms of the simple type, patients of progressive type show difficulties to perform other simple activities like lifting a fork or using a spoon

while eating. Dystonic type patients having similar symptoms of progressive type from the start of disease also experience involuntary muscle contractions [13-14]. Solly, in 1864 rechristened writer's cramp as scrivener's palsy [35].

Several theories regarding the pathophysiology of scrivener's palsy are based on the concept of repetitive,over-use, stereotyped hand patterns [36-37]. There are mainly three pathophysiological theories behind writer's cramps (WC) which are abolished reciprocal inhibition [38,39], abnormal plasticity [40], and sensory functional abnormality [41].

Patient with writer's cramp complains about the difficulty in writing at the start of the disease, stiffness in the muscles of the arm, fingers, forearm, and occasionally, the shoulder.It can be accompanied by pain, tingling, or prickling sensation. Mostly pain and paresthesia developed at a later stage with carpal tunnel syndrome. Approximately 33% of patients show tremors during writing in the affected hand. In progressive and dystonic types, the patient also experiences difficulty performing other tasks.To overcome difficulty during writing patient displays gripping of the pen excessively, sometimes with an elevation of the elbow and ulnar deviation.Neurological examination mostly shows tendon reflexes, sensation, and muscle strength as within normal limits[13-14,18].

WC has an incidence rate of approximately 2.7 per million population and a prevalence rate found between 16- 68 /1000000 [42-43].

In one study it was seen that approximately 17 % of patients stopped writing completely and started using typewriters, and dictating aids, around 17% started using another hand instead dominant hand, while approximately 60-65 % continued to use the dominant hand but held the pen with the tight grip. Only one patient changed her profession while others continue their profession. It also affects a patient's psychological status[14].

Diagnosis of Writer's cramp is mostly based upon clinical history, neurological examination at rest, and performing activity [43]. Electromyography [EMG], Brain MRI, and spinal MRI can be performed if certain specific symptoms are present. If family history and young-onset of disease present then testing for DYT1 gene mutation will be done[44-45]. For clinical assessment writer's Cramp Rating Scale(WCRS), Arm Dystonia Disability Scale (ADDS) can be useful[47-49].

The main goal of treatment of WC is to improve the patient's writing ability and decrease spasms, tremors, and abnormal posture. Treatment strategies consist of Pharmacological, Surgical, and Non-pharmacological strategies [44].

Pharmacological treatment includes oral medicines and botulinum toxin injections. Oral medicines mostly used are anticholinergic drugs, GABAergic, dopaminergic, dopamine depleting actions, and benzodiazepines [44, 50]. Profit due to anticholinergic drugs is seen in approximately 20% of patients[13]. 50% of patient who was on propranolol, diazepam, and primidone for tremors had positive effects [51]. The most beneficial treatment for Writer's cramp is considered botulinum neurotoxin injections but they have a temporary effect that lasts around 84 days [52-55]. In one study it was proven that around 50% of patients still showed good results with botulinum toxin injections [56]. Discontinuation from treatment was observed due to loss of positive effect, travel distance, and muscle weakness [57]. One case report shows that intramuscular lidocaine injection as a block improved WC but it lasted only 12-24 hours. No side effect was seen [58].

Surgical treatment is preferred in cases who are not responding to pharmacological treatment. Two surgical interventions are available: thalamotomy and deep brain stimulation (DBS).In Taira and Hori case series report, it was observed that 16% of the patient shows post thalamotomy transient complications such as mild hemiparesis, and dysarthria. Resolving of symptoms was seen in around 84% of patients. After 36 months of follow up only 4% of patients showed recurrence of the disease. But thalamotomy is a costly procedure [59-61]. Fukaya et al. in their DBS study showed good results. Around 86-90% of symptoms of WC resolved after DBS. Adverse events seen with DBS were infection and electrode migration, electrode breakage, or misplacement. DBS requires lifetime maintenance and battery replacement. Therefore DBS is not a good option in the case of young patients [62-63].

Non-pharmacological treatment of WC consists of the use of adaptive devices, physiotherapy, using the thicker barrel and point pen, occupational therapy, cooling technique, relaxation therapy, biofeedback, acupuncture, herbal therapy, and massage therapy [44,64]. Various studies show that altering hand posture reduces symptoms of WC. In writing orthotic devices (WOD), the technique of altering hand posture is used for the treatment of WC [65]. Singam

et al. reported that in 15 subjects studied with WOD, 2 Subjects do not complete the study as they were unable to adapt to the device. It was seen that quality scores improved due to WOD. This device is cost-friendly, easy to use and handle, and non-invasive. But it requires a large sample size study for the conclusion of benefits [66]. One study shows that cooling of the upper extremity in the case of WC is an easy, cheap additional treatment option but it gives only temporary relief[67]. Acupuncture, herbal treatment, massage, occupational therapy, splints, and devices can be beneficial in some patients but it is not given adequate results [44].

As it is a rare disease and not much study was done. Until we are getting exact results from other treatment strategies botulinum toxin and DBS will be the most effective line of treatment in WC[44].

It is observed that patients with writer's cramps start using their non-dominant hands to get relief from dystonia. In severe cases, dystonia occurs in non-dominant hands also after some time. Patients with writer's cramps experience difficulty with day-to-day activities like writing, combing hair, and sewing. In some cases, it is found that they are even unable to do signatures which leads to cheques getting rejected/bounced and results in a serious outcome. All this disability develops anxiety and depression in patients with WC and even causes social phobia. Some changes job or loss job and become unemployed. Some patients experienced difficulty performing routine tasks so they started living life as handicapped [8, 11, 14]. WC comes under occupational neuroses (ICD-9, F300.8) and other specified neurotic disorders (ICD-10, F48.8). Very few recent studies are available for writer's cramps and the sample size is also small. Treat Writer's cramp patient as handicapped, require a large sample size, case-control study, long-duration follow up period [68].

Conclusion:-

Very few recent studies are available for writer's cramps and the sample size is also small. Treat Writer's cramp patient as handicapped, we require a large sample size, case-control study, long-duration follow up period.

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