INTRODUCTION

Talipes equinovarus or clubfoot (OMIM-119800) is a gross deformity of the foot presented at birth. The word talipes is derived from talus (ankle) and pes (foot). Talipes denotes the clublike appearance of the foot and exists in various subtypes; talipes equinovarus (TEV) being the most prevalent one. The incidence of TEV is about 1/1000 live births per year. It is the seventh most prevalent congenital birth anomaly and the most common of the musculoskeletal system. Globally the burden of this birth defect affects more than 150,000 infants every year. Among all the cases born worldwide, 80% live in low- and middle-income countries. It is depicted in 5,000 years old Egyptian hieroglyphs and firstly reported by Hippocrates 400 years BC. 

TEV can occur as an isolated entity, usually termed as idiopathic, or as a syndromic condition. In its syndromic presentation, it arises in many neurological, neuromuscular and paralytic disorders. Both idiopathic and syndromic conditions can be milder or severe. It generally has a sporadic occurrence but familial cases showing segregation in several generations are also reported. Its etiology is considered to be
a combination of genetic and environmental factors. TEV has a highly negative impact on the life of the subject. If left untreated it may result in dependency on others for performing the daily activities, resulting in heavy economic burden on the family and the country.

For prenatal diagnosis, ultrasonography is considered the most reliable and majority of the cases can be diagnosed after 17 weeks of gestation. Treatment of TEV comprises both surgical and non-surgical methods and is effective in the early years of life. The Ponseti method remains the most popular non-surgical technique.

The present study was aimed to assess the status of scientific literature on TEV published from Pakistan, to get an insight into the trends in knowledge over the years, and to highlight the study gaps in this area, hence to provide directions for further research.

**METHODS**

A review of the literature was conducted from November-2019 to January-2020 and all the papers fulfilling the inclusion criteria and published by the Pakistani researchers were considered. The search strategy adopted was an article title/keyword/abstract-based search using the following key terms: ‘talipes congenital’, ‘clubfoot/congenital clubfoot’, ‘talipes equinovarus/congenital talipes equinovarus’ in Pakistan. TEV reported under the study title of birth defects, musculoskeletal disorder, and congenital foot deformities, were included. PubMed, PakMediNet, Medline, Embase, Science Direct, and Google Scholar were the search engines employed for literature search. The pertinent information including authors, institute, study setting, duration, sample size, target population, age group, goals, and management approach, was extracted. Data were maintained in Excel sheet.

**RESULTS**

**Journals, time era and study setting:** A total of 63 articles were retrieved; of these 56 (89%) studies were published in local journals while 7 (11%) were published in international journals. The highest number of studies (n=10) were published in J Pak Orthop Assoc.

Extensive studies were conducted during the period 2011-2014 (n=28), followed by 2015-2019 (n=17). The highest number of studies were conducted in Sindh (n=27), followed by Khyber Pakhtunkhwa (n=21) and Punjab (n=14) (Fig.1).

**Incidence, prevalence and epidemiology:** The studies reporting true birth-prevalence of TEV in Pakistan are scarce. Its estimated incidence was 6,000–7,000 cases/year; i.e., 1.4:1,000 livebirths and 1.5/1,000 livebirths.**Studies showing risk SNPs, twin studies and effect on consanguinity and familial attributes, remain to be elucidated in multi-ethnic/multi-lingual Pakistani cohorts.**

**Genetics:** Indeed, no study has been published from Pakistan which could highlight the genetic underpinning of TEV. Hence, information regarding the inheritance pattern(s) of TEV, its genetic mapping, gene identification, association studies showing risk SNPs, twin studies and effect of consanguinity and familial attributes, remain to be elucidated in multi-ethnic/multi-lingual Pakistani cohorts.
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Fig. 1: A. Mapping of number of studies on TEV published from various cities.
B. Number of studies published on TEV depicted on bi-annual bar-chart.
Table I: Summary of representative studies carried out in Pakistan on TEV.

| Reference       | Institute                                      | Design          | Duration       | Sample | Study domain          | Theme/management plans                                      |
|-----------------|------------------------------------------------|-----------------|----------------|--------|-----------------------|-------------------------------------------------------------|
| Din, 2004(39)   | Hayatabad Med Complex, Peshawar                | Prospective     | 1998-2000      | 96     | Non-operative         | Kite-Lovell technique                                       |
| Khan and Chino-| Karachi                                        | Prospective     | 2000-2004      | 15     | Operative             | Double zigzag incision as single-stage procedure; neglected cases |
| noy, 2006(15)   | Prospective-Descr. |                |                |        |                      | Turcos procedure, serial casting, neglected cases            |
| Humail et al.   | Dow Uni of Health Science, Karachi             | Prospective-Descr. | 1998-2004      | 360    | Operative & Non-oper. | Consenative management                                      |
| 2009(16)        | Karachi                                        | Prospective     | 2000-2004      | 50     | Non-operative         | Case history, clinical parameters                           |
|                 | Birmingham                                     | Cross-sectional | 18 months      |        |                      | Split tibialis anterior and posterior tendon transfer        |
| Sami et al.     | Mayo Hospital, Lahore                          | Cross-sectional | 2005, 2009     | 20     | Operative             | Revision surgery, PMR, Turco’s, neglected/re-lapsed cases   |
| 2010(38)        | Liqout Uni of Med Sci, Jamshoro                | Prospective-Descr. |                |        |                      | One stage posteromedial release                             |
| Ahmed et al.    | Abbasi Shaheed Hospital, Karachi               | Retrospective,  | 2006-2008      | 13     | Operative             | Percutaneous tendo Achilles lengthening                     |
| 2011(20)        | Liaqut Uni of Med Sci, Jamshoro                | Observational-Descr. |                | 49     | Non-operative         | Ponseti method                                              |
| Makhdoom et al. | Abbasi Shaheed Hospital, Karachi               | Observational-Descr. |                |        |                      | Ponseti vs.Turco’s posteromedial                            |
| 2011(21)        | Mayo Hospital, Lahore                          | Cross-sectional | 2008-2010      | 60     | Operative & Non-oper. | One stage posteromedial release                             |
| Inam et al.     | Hayatabad Med Complex, Peshawar                | Comparative     | 2008-2010      | 45     | Operative             | One stage posteromedial release                             |
| 2012(11)        | Khyber Teaching Hospital, Peshawar             | Prospective     | 2008-2011      | 23     | Operative             | Percutaneous tendo Achilles lengthening                     |
| Khan et al.     | Khyber Teaching Hospital, Peshawar             | Retrospective   | 2009-2010      | 70     | Non-operative         | Ponseti method                                              |
| 2013(36)        | Benazir Bhutto Hospital, Rawalpindi            | Prospective     | 2010-2011      | 55     | Non-operative         | Ponseti method                                              |
| Zia et al.      | Indus Hospital, Karachi                        | Descriptive     | 2012           | Parents | Non-operative         | Cost-effectiveness of Ponseti                               |
| 2013(35)        | Jinnah Postgrad. Med Centre, Karachi           | Cross-sectional | 2009-2010      | 70     | Non-operative         | Achilles tendon tenotomy in Ponseti                         |
| Hussain et al.  | Khyber Teaching Hospital, Peshawar             | Prospective     | 2012           | Parents | Non-operative         | Ponseti method                                              |
| 2014(10)        | Jinnah Postgrad. Med Centre, Karachi           | Cross-sectional | 2009-2010      | 125    | Non-operative         | Ponseti method                                              |
| Khan et al.     | Hayatabad Med Complex, Peshawar                | Prospective-Descr. |                | 28     | Non-operative         | Accelerated Ponseti, neglected cases                        |
| 2014(18)        | Hayatabad Med Complex, Peshawar                | Prospective     | 2013-2014      |        |                      |                                                             |
to-conservative was (2:2) and (6:1), respectively, while in 2010-2014 and 2015-2019, the ratio is (10:19) and (1:13), respectively. The latest studies majorly covered the conservative treatment domain.

Various conservative treatment methods were in practice in Pakistan. Before 2006, Kite method and surgical treatment were in practice. Later, Ponseti and accelerated Ponseti methods became popular.\(^\text{18,24}\) However, no study was reported regarding the French method. Ishaque (2009) has reviewed the conservative management of TEV.\(^\text{25}\)

Studies also reported various surgical measures, i.e., Turco procedure, Window procedure, Posteromedial release, Subtalar release, Modified Turco’s Posteromedial release, etc. Pirani scoring was commonly used to assess the severity level. Irfan and Mehboob (2013) carried out ultrasonography for prenatal detection of TEV in 1,000 expecting women in Lahore.\(^\text{27}\)

**DISCUSSION**

To the best of our knowledge, it is the first review that assembled the available Pakistani literature on TEV regarding its natural history, study designs and management. The chronological arrangement of published reports has revealed that this malformation has gained some attention among the researchers over the years. However, the researchers have predominantly explored a relatively narrow domain and many of the fundamental questions on TEV remain to be elucidated (see below). There has been no comprehensive study reporting the true prevalence and incidence of TEV,\(^\text{28}\) thus the burden of this disorder remains unknown. Traditionally, the hospital-based studies mainly focus on the treatment-seeking group while the pattern of malformation in the general population remains obscure. Further, most of the literature has been published from Khyber Pakhtunkhwa and Sindh provinces and the representative data from the...
tertiary care institutes of other cosmopolitan cities across Pakistan are deficient.

Here, the researchers have primarily focused on the treatment/management of TEV. Both conservative and operative management have been practiced, however, the trend has shifted from operative to conservative management (Table-I). Follow up studies have been conducted, but short-term follow up should be complemented with long-term follow up in order to assess treatment outcome. Mismanagement, noncompliance, natural history and severity of disease are responsible for the recurrence.

The younger/pediatric population remains the focus of most of the studies. There are several potential hurdles in Pakistan including poverty, lack of awareness, lack of specialized clinics/doctors, that delay the treatment of TEV at younger ages. Hence, the neglected or late cases are overrepresented, culminating an increased prevalence of this malformation. Interestingly, unilateral cases were highly presented in majority of the reported cohorts which is contrasting to the other studies where bilateral TEV cases comprised 50% of samples. Hence, investigations are required in order to understand the underlying cause(s) of this discrepancy. Bilateral clubfoot may result from an increased load of genetic factors.

The etiology of TEV includes both genetic as well as environmental factors. Genetics has a clear causative role in a substantial number of TEV cases. So far, no study was reported from Pakistan concerning gene identification, gene mapping, genetic mutation and mode of inheritance. Family history, consanguinity, familial attributes, etc. were evaluated in only a few studies. In familial cases, the degree of relatedness plays a key role as first degree relatives are more prone to disorder as compared to distant relatives; these factors are largely unaddressed in Pakistan. Besides this, studies reporting the role of maternal, and environmental factors and seasonal variation are scarce. Further, TEV has 33% concordance among monozygotic twins while 3% in dizygotic twins. This area also remains to be explored in Pakistani cohorts.

Clubfoot has a negative impact on the life of patient; if it is left untreated it may cause dependency on others for performing the daily activities, difficulties in ambulation and lifelong disability. It causes heavy economic burden not only on the family but on the country as well. An estimated 80% of such cases can be seen in developing countries.

Limitations of the study: All of the clinical parameters of TEV reported in the literature were not covered due to limitation of space. Studies on clubfoot related to prenatal ultrasonographic detection; awareness, perception and attitude towards clubfoot; perspectives from the caregivers’ standpoint; laboratory/motor electrophysiological studies, and surgical approaches, were not included.

CONCLUSION

The hotspot of TEV research in Pakistan is its treatment and management, predominantly the Ponseti method. This study reveals that there is a scarcity of research on various important aspects of TEV in Pakistan, and its epidemiology, prevalence, etiology, risk factors, associated anomalies, maternal and obstetric factors, birth parameters, molecular diagnostics, etc., need to be elucidated. Moreover, large scale population-based studies are required for a broader overview of the malformation. This review highlights marked dearth of scientific evidence on TEV required for awareness, policy-making and relevant public health action.

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Authors’ Contribution:

SM conceived, designed and planned study and also responsible and accountable for the accuracy or integrity of the work.

KM & ZS did data collection and manuscript writing.

KM, ZS & SM edited, reviewed and approved manuscript.