Hearing loss is the second most common disability comprising of 18.9% of persons with disability. Hearing disability is the highest (0.60%) when compared to other disabilities, among children between 0-4 years (0.28%) and 5-9 years (0.32%) (Census of India, 2011). In addition to the reported incidence of congenital hearing loss is 5.6 per 1000 births (Nagapoornima et al., 2007), India is also burdened with the highest prevalence of children with chronic suppurative otitis media (World Health Organisation [WHO], 2004), which when unresolved is a common cause of acquired hearing loss.

Acoustic stimulation in the first 18 months is crucial for the development of central auditory pathway (Finitoz, Albright, O'Neal, 1998 as cited in Grill et al., 2006, page 1). If hearing impairment in a child is not identified early and intervened, the child tends to have speech and language delay. This delay is not easily recovered when rehabilitation is provided later than 2 years of age (Grill et al., 2006). Further, the child’s education, community living and social well-being are also affected (Smith, 2009; Downs, Yoshinaga-Itano, 1999). More than 50% of persons with hearing impairment are illiterate and unemployed in India (National Sample Survey Organisation [NSSO], 2002). As per WHO- South-East Asia Region report on state of ear and hearing care, hearing loss accounts for 2.3% of all Disability Adjusted Life Years (DALY) in this region, which includes India (WHO, 2009). Early identification and intervention has the potential to improve language outcomes, which is likely to result in a 10% decrease in special education costs and a 75% decrease in vocational rehabilitation costs (Keren, Helfand, Homer, McPhilips, Lieu, 2002). Therefore, early identification and intervention is recommended, to capitalize on the critical age during which there is maximum neural plasticity of the brain.
Universal Newborn Hearing Screening (UNHS) is the recommended protocol by organizations such as the Joint Committee on Infant Hearing (JCIH) for early detection and intervention of congenital hearing loss. In UNHS, every baby born is screened for hearing loss before hospital discharge. National level organizations such as the Centre for Disease Control and Prevention, American Academy of Pediatrics (USA) and National Health Service (UK) recommend hearing screening at school entry level for identification of late onset or progressive hearing loss. Other approaches include targeted screening, where only those children at risk for hearing loss are screened. However, universal screening for hearing loss was emphasized as 42-70% of children were likely to be missed in risk-based screening (Hadad, 2004 as cited in Jewel, Varghese, Singh & Varghese, 2013, page 212).

Infant hearing screening programs are not widely adopted in developed countries, unlike in low or middle-income countries like India. As a result, a large number of children do not receive early identification or intervention. In rural West Bengal, India, the average age of suspicion of hearing loss was 1.5 years, the first visit to the doctor was at a mean age of 2.4 years and consultation with an audiologist was at a mean age of 9.3 years (Rout & Singh, 2010). In a cross sectional study of 246 children between 5 to 15 years of age in Surat, Gujarat, India, the average age of suspicion was 2.9 ±1.7 years, the first consultation was at 3.5±1.5 years and the average age of intervention for hearing loss was 7.8± 3.3 years (Patel, Moitra, Modi, Patel, & Kantharia, 2014). On the other hand in the prominent metro city of Mumbai, the age of identification (AOI) was found to be 27.77 months in 2008. A trend analysis of AOI among 510 children with hearing impairment born between 1989-2008, showed reduction in AOI by 9.59 months in two decades in the absence
of UNHS. The AOI reduction was attributed to increased number of pediatric hearing assessment centers in the city of Mumbai. In addition to such studies, anecdotal reports suggest that hospital based programs in major cities have been successful in lowering the AOI to well below 3 years of age. The reduction in AOI and the emergence of more facilities for audiological evaluation in urban areas is a positive trend. However, the AOI is much higher than the optimum age of 1 year and there exists a stark contrast in AOI between urban and rural areas.

Newborn hearing screening (NHS) programs were implemented in India as part of research studies to compare different techniques of screening and reported sensitivity, specificity, and cost of screening (Nikam & Dharamraj, 1971; Basavaraj, Kumudavalli, & Kamakshi, 1985; Ravi 2000; Maru, 2002; Yathiraj, Sameer & Jayaram, 2002). While these were research initiatives, a few hospital-based programs were also established. Most of these programs suffered poor follow-up for re-screening and diagnostic testing. Nagarajan, Janet, Bala and Binu (2008) based on a hospital based NHS program in Chennai, reported that only five out of the 41 babies referred for follow-up, came back for diagnostic testing. Only 28% follow-up for diagnostic testing was reported in an NHS program at a tertiary care hospital in Vellore, Tamil Nadu (Augustine et al., 2014). Nagarajan et al. (2008) attributed travel distance, inconvenience, and lack of parental interest as major factors for poor follow-up in their program. John, Balraj and Kurien (2009) reported that follow-up testing of babies was a challenge due to parents’ lack of perception of child’s hearing problem as it is a hidden handicap. KEM hospital, Pune (Vaid, Shanbhag, Nikam & Biswas, 2009), a group of twenty hospitals in Cochin (Paul, 2011) and
Christian Medical College and hospital, Ludhiana, (Jewel et al., 2013) are other tertiary care hospital based NHS programs.

Hospital-based hearing screening programs are on the increase and are a welcome initiative. However, in a country like India, due to the inequity in rural: urban population, hospital-based programs cater only to small sections of the society. In order to reach the country at large, the Ministry of Health and Family Welfare, in 2006, Government of India launched the ‘National Programme for Prevention and Control of Deafness’ (NPPCD) as a step to promote early identification of congenital and acquired hearing loss. Under this programme, both institution-based screening and community-based screening were implemented. At the grass root level, health workers, anganwadi workers, birth attendants were to be trained for creating awareness regarding prevention of hearing loss, and facilitate early detection using behavioural measures at immunization clinics and through home visits. To target older children with hearing loss, school screening camps were to be conducted by doctors with the assistance of primary school teachers. Diagnostic evaluations and management of children referred from the community and school screening were to be carried out by the ENT specialist, audiologist or audiometrician at the district hospital (Garg, Chadha, Malhotra & Agarwal, 2009).

This program is an important milestone as it integrates primary ear care with primary and district health systems, which has the potential to reach both urban and rural populations. The program started in a pilot phase in 25 districts in 2006 has expanded to 192 districts in 2013 (Agarwal, Chadha & Garg, 2013). However, an impact assessment by National Institute of Health and Family Welfare suggests that
infrastructural facilities, and equipment related shortcomings plagued the program in several states (Das, Mathiyazhagan, Yadav, Devrani & Mehra, 2013). In addition, shortage of centers with diagnostic testing facility and shortage of audiologists in all district hospitals were reported (WHO 2009).

While there are some initiatives for new born hearing screening in our country, due to a lack of a systematic infant hearing screening program, in low and middle-income countries like India, most children are not identified until they enter primary school or at a later stage (Davis, Carr, & Davis, 2014; Olusanya, Wirz, & Luxon, 2008; Rout et al., 2010, Patel et al., 2014). WHO recommends that national plans for infants, toddlers and children be prepared for the prevention and control of major causes of avoidable hearing loss within the framework of primary health care (WHO, 2009). In line with these recommendations, it is desirable that in countries like India, in the initial stages of a hearing screening program, infants and older children up to the pre-school age be included. As hearing screening programs improve the coverage of infants, the age of screening is likely to reduce.

In India, the challenges in implementing hearing screening programs include lack of availability of audiologists and infrastructure to cover the entire population. Seventy-two percentage of the population live in rural areas and only 28% live in urban areas, where health care services are predominate, posing considerable challenges in accessing healthcare facilities for those living in rural areas (Aitken, Bakliwal, Chang & Udeshi, 2013).
There are around 350 government-run hospitals in the country with tertiary care facilities. Of these 120 are reported to have diagnostic and rehabilitation facilities for early detection of hearing loss. Significant number of private centers offer audiological facility, however, they are not uniformly distributed in the country. There is a higher concentration in the cities than in towns and villages (WHO, 2007). Additionally, there is a stark contrast in demand for manpower versus the capacity, as the number of audiologist and audiometricians combined was found to be 1:500,000 (WHO, 2009).

Experts in hearing health and education for children with hearing impairment from the national institutes of India proposed a community based approach to screen infants, where anganwadi workers can be trained to provide prevention and early identification related services at the door step (Rehabilitation Council of India [RCI], 2007). A community-based program has the advantage of increased sustainability since the program can be designed based on existing resources accessible to all community members. Also, there is the added advantage of the involvement of local community leaders and volunteers, which can reinforce community-based programs (Sternin, Sernin, & Marsh, 1998).

The consensus among experts at the National Consensus on Prevention, Identification and Management of Hearing Impairment held at the All India Institute of Speech and Hearing, Mysore, in 2005, was that the activities towards prevention of hearing impairment should be carried out at the doorstep of each household by grass root level workers. In the absence of such workers, it was recommended that Education Guarantee Scheme (EGS) teachers or Lower Primary School (LPS)
teachers be assigned such roles, who would be supervised by a medical officer at the Primary Health Centre (PHC) (RCI, 2007).

Therefore, there is a need to explore methods to extend access and coverage of hearing screening services. Providing screening services using grass root level workers as envisioned by experts can increase coverage, however, the existing shortage of audiologist and infrastructure at rural centres makes newborn hearing screening unviable. When additional hearing services are needed after screening by grass root workers, parents are unlikely to travel to distant centres due to transportation, cost and cultural reasons. As a result, the diagnosis of infant hearing loss is delayed (Alberg J, Wilson K, Roush R., 2006 as cited in Krumm, Huffman, Dick, & Klich, 2008 page 102 ). In such a scenario, telemedicine or tele-practice may be a method that can ameliorate these problems.

Tele-medicine, which is the provision of health services from one location to another using telecommunication medium, offers real benefits in a country as vast as India where the majority of the population lives in remote areas. The Indian Space Research Organization (ISRO) started their telemedicine projects in 2001 with the aim of introducing telemedicine facilities to rural populations. ISRO provided satellite connectivity between remote district hospitals/health centres providing basic medical care, with super specialty hospitals. Also, mobile tele-van units were utilized to provide basic eye testing and community health for medical consultation and dispensing (ISRO, n.d.).
Telepractice services can be provided using relatively simple technology such as telephone conferencing, e-mail, or text message to more sophisticated systems such as wide area networks using high-speed "point to point" connections, premium interactive video systems and real-time testing using remote computing (Krumm, 2007). The advantage of tele-health services is that there is significant cost saving since the patients avoid expenses towards travel, stay, and for treatment at the hospitals in the cities (ISRO, 2005). Also, from an administrative perspective the cost of infrastructure development, personnel and equipment can be significantly minimized.

Two important requirements for tele-practice are: a) presence of a trained facilitator at the remote end to assist in establishing connectivity, handling clinical equipment and computers, and preparing patients for the testing process, and b) the availability of tele-communication connectivity between remote site and professional end (near site). Broadly two types of connectivity are available: satellite based, where a stable IP over satellite connection is available even in remote rural areas with minimum bandwidth of 256 Kbps, and the other is the broadband internet which is available through digital subscriber line (DSL), mobile phones and hotspots. Broadband Internet has greater speed and flexibility compared to satellite connectivity, however, internet penetration in rural areas is very limited (Belson, Leighton, & Rinklin, 2012).

There are two methods of tele-practice: store and forward (asynchronous) or real-time (synchronous). In the store and forward (asynchronous) model, patient data is obtained and sent to an audiologist or consultant for interpretation. In real-time
(synchronous) methods, the practitioner either conducts live video conferencing with the patient and/or takes remote control of clinical equipment to conduct live testing from the hospital/tertiary care centre. Store and forward method is routinely employed in disciplines such as dermatology (Kaliyadan & Venkitakrishnan, 2009) and ophthalmology (Verma, Raman & Mohan, 2009; Bai, Murali, Kim & Srivatsa, 2007) where images are captured, stored and forwarded to the radiologist and ophthalmologist, respectively. Real-time method is most commonly used for super speciality consultations where video conferencing is used. Tele-technology has been well explored in areas such as ophthalmology, dermatology, pathology (Baruha, 2005), neurology (Ganapathy, 2005), and cardiology (Sekar & Vilvanathan, 2007).

Alternative service delivery, before adoption into routine use, should be proved superior to the approach in practice that it is intended to replace; that is, it has to be cost-effective and telemedicine is no exception to this rule. (Roine, Ohinmaa, & Hailey, 2001). Evidence of the effect of telehealth on service use, costs, or cost-effectiveness is limited and most often reported in the U.S.A. Convincing evidence of effectiveness has been reported in teleradiology, tele-neurosurgery, telepsychiatry, transmission of echocardiographic images, and the use of electronic referrals enabling email consultations and video conferencing between primary and secondary health care providers (Roine et al., 2001).

Audiology as a profession has been a slow adopter of tele-practice, however, this form of service delivery is gaining more acceptance with availability of inexpensive video conferencing systems and better internet penetration (Krumm, 2007). In the western countries, researchers have studied the application of tele-audiology in
screening, diagnosis and intervention. Givens et al. (2003) demonstrated that real-time internet-based assessment of hearing can be accomplished. Krumm, Ribera and Schmiedge (2005) studied the implication of using tele-health for universal newborn hearing screening. The initial findings of the study supports the validity of tele-practice with infant hearing services, however, further examination of factors such as cost, clinician and patient acceptance of tele-testing and technical effectiveness is recommended. Krumm et al., 2008, found identical hearing screening results using face-face and telemedicine testing on newborns using ABR. Other areas include, a study by Krumm, Ribera and Klich (2007) for providing basic hearing testing using remote computing technology in adults, hearing screening via tele-health in rural elementary school (Lancaster, Krumm, Ribera & Klich, 2008). A few have also reported intervention-based applications such as hearing aid benefit in adults, and internet-based treatment for tinnitus (Smits, Kapteyn & Houtgast, 2004, Smits, Merkus & Houtgast, 2006, Bexelius et al., 2008). However, these are feasibility or experimental studies and are not applied in large programs. Till date, there is no reported tele-audiology study in India.

**Need for the study**

The prevalence of hearing loss in infants is more than twice that of other screened newborn disorders like congenital hypothyroidism and phenylketonuria (Michele, 2004 as cited in John et al., 2009, page 23). Considering the pervasive nature of hearing impairment and the large number of people affected, it is important that there is adequate access to screening, diagnostic and rehabilitation services to facilitate early diagnosis and intervention. Hospital-based hearing screening programs are less accessible and result in poor coverage, as well as high loss to
follow-up. Lack of manpower and infrastructure to cover all the districts is a setback for the national program. This necessitates a solution that has a potential to cover the large population, with less manpower and infrastructure.

WHO (2012) recommends integrating ear and hearing care into community-based rehabilitation programmes since it can improve coverage, especially in rural areas. While this may solve the issue of reaching screening services, application of telemedicine for diagnostic follow-up testing at the rural area can circumvent the problem of limited manpower and infrastructure.

In an attempt to reach rural areas and overcome the challenge of loss to follow-up for diagnostic assessment, this study explores the combination of a community based hearing screening program for infants and young children in remote rural villages with tele-Auditory Brainstem Response recording for diagnostic testing. Combining a community-based screening approach with a tele-audiological diagnostic testing, is also expected to reduce the cost incurred by the child and family member seeking services, in terms of travel, and wage loss due to time spent in accessing such services in distant places. The provider cost may also decline, as infrastructure for tele-medicine is transportable and can be a shared resource among communities as compared with building infrastructure, procuring equipment and employing professionals at each community.

Allocation of limited resources for health is prioritised based on returns to society. Cost-effectiveness of interventions is one of the key indicators of such return. Economic analysis seeks to identify and to make explicit a set of criteria which may
be useful in prioritising the use of scarce resources (Drummond, Stoddart, & Torrance, 1987, as cited in Shamanna, Dandona & Rao, 1998, page 170). The prospects of any immediate action to implement infant hearing screening programs are still uncertain in most developing countries without such cost data (Uus, Bamford, & Taylor, 2006). There is evidence from the West suggesting that the program cost outweighs the projected estimates of benefits, which include both short-term and long-term benefits, indicating that UNHS is a worthwhile investment for society (Keren et al., 2002). In developing countries, where scarce resources are to be allocated, contextual research evidence, is pertinent. In addition to cost-effectiveness, scientific evidence of acceptability of telemedicine support its integration into routine practice (Williams, May, & Esmal, 2001).

Therefore, the present study aims to develop a community-based model of hearing screening with tele-audiological diagnostic follow-up and evaluate its effectiveness as against the traditional face-to-face follow-up at a tertiary care hospital.